Sharing Findings on Sickle Cell Disorder in International Collaborative Biomedical Research: An Empirical Ethics Study in Coastal Kenya

DPhil Thesis

Victoria Mary Chuck Marsh

Kellogg College

Hilary Term 2012

ABSTRACT

Sharing Findings on Sickle Cell Disorder in International Collaborative Biomedical Research: An Empirical Ethics study in Coastal Kenya

Victoria Mary Chuck Marsh

Kellogg College

DPhil

Hilary Term 2012

Against the background of a dilemma experienced by researchers during a genomics study at an established biomedical research centre in Kenya, the broad aims of this thesis are to develop appropriate responses to important ethical questions on sharing information on a common and serious genetic condition, sickle cell disorder, and assess the responsibilities of researchers in this regard. Using an empirical approach to normative reflection across two phases of qualitative research, I explore the nature of important moral concerns related to sharing sickle cell disease information from researchers' and community members' points of view; and develop a bottom-up normative analysis around the questions generated. This analysis interweaves community experiences, processes of community reasoning and *ex situ* normative reflection; placing community views and values centrally while referencing these to wider ethical debates, commentaries and guidelines in the literature. Two main outputs of this thesis are to provide recommendations for information sharing on SCD findings in the genomics study in Kilifi; and to propose a set of key issues to consider for this type of information in other studies and geographic settings. I conclude that researchers have a strong responsibility to share SCD information on affected children with families as a form of ancillary service (validating tests, counselling and care); but less responsibility to actively share carrier information. Concurrent responsibilities are working collaboratively with the Ministry of Health/District General Hospital to plan and implement services for SCD; ensuring counselling services support family stability as far as reasonably possible; and to build forms of community engagement and informed consent that counter risks of diagnostic interpretations of research.

Table of Contents

Title page..... i
Abstract..... ii
Table of Contents..... iii
Table of Figures and Tables..... ix
Acknowledgements..... x

CHAPTER 1 Introduction, Background and Overall Aims of the Thesis

1.1 Introduction..... 11
1.2 The KEMRI Wellcome Trust Research Programme in Kilifi..... 13
1.2.1 The research programme..... 13
1.2.2 Kilifi District and local residents..... 16
1.3 The Kilifi Genetic Birth Cohort..... 18
1.4 Sickle Cell Disease in Africa..... 20
1.5 Thesis objectives..... 23
1.6 Literature on ethical issues in genetic screening and testing..... 24
1.7 Literature on the disclosure of genetic findings in research..... 27
1.7.1 Understanding the central role of 'clinical utility' for researchers' responsibilities..... 29
1.7.2 Recognising the influence of the availability of services..... 31
1.7.3 Providing a wider understanding of 'utility'..... 32
1.7.4 Understanding the role of parent-child relationships for participants who are minors..... 33
1.8 Literature on researchers' positive responsibilities for providing services in developing countries..... 35
1.8.1 Researchers' responsibilities to take account of background conditions of injustice..... 38
1.8.2 Researchers' non professional responsibilities for ancillary care..... 39
1.8.3 Researchers' responsibilities for ancillary care from a 'partial entrustment' model..... 40
1.9 Summary and overview of the thesis..... 43

CHAPTER 2 Methodological Approaches

2.1 Introduction and organisation of chapter..... 45
2.2 Overall study design..... 46
2.3 Study methods in Phase One..... 49
2.3.1 Study participants and sampling in Phase One..... 49
2.3.1.1 Initial sampling and study participants in Phase One..... 50
2.3.1.2 Later sampling and study participants in Phase One..... 51
2.3.2 Data collection and management in Phase One..... 54
2.3.3 Data analysis in Phase One..... 56

2.4 Study methods in Phase Two ... **56**
2.4.1 Theoretical considerations for methods in Phase Two ... 56
2.4.1.1 Using 'non expert' reasoning in normative analysis ... *57*
2.4.1.2 Using deliberative processes in normative analysis ... *61*
2.4.2 Study participants and sampling in Phase Two ... 62
2.4.3 Information sharing activities in Phase Two ... 66
2.4.4 Data collection in Phase Two ... 67
2.4.5 Data analysis in Phase Two ... 69
2.5 Limitations for methods ... **71**
2.5.1 Sampling in Phases One and Two ... 71
2.5.2 Data collection and analysis in Phases One and Two ... 72
2.6 Scientific and ethical approval ... **73**
2.7 Chapter summary ... **73**

CHAPTER 3 Phase One: Encounters with experience: Sickle Cell Disease and the Kilifi Genetics Birth Cohort

3.1 Introduction: Background, aims and organisation of chapter ... **75**
3.2 Community perspectives on SCD ... **76**
3.2.1 Community awareness and recognition of SCD ... 77
3.2.2 Beliefs about cause and looking for a cure: Anxiety and fears ... 78
3.2.3 Supernatural explanations for SCD ... 80
3.2.3.1 Supernatural explanations are likely but not universal ... *80*
3.2.3.2 Blaming others for supernatural causes ... *82*
3.2.3.3 The possibility that children are blamed for SCD: Curses ... *83*
3.2.4 Looking for a cure: Family surveillance and blame ... 84
3.2.4.1 Gendered family blame in SCD: High risks for mothers ... *85*
3.2.4.2 Questioning marital faithfulness and paternity: High risks for mothers ... *87*
3.2.5 Gendered impacts of blaming: Broken livelihoods and families ... 89
3.3 Community perceptions, experiences and attitudes towards KGBC ... **93**
3.3.1 Why people decided to participate (or not) ... 94
3.3.1.1 Perceived value as a health or SCD check for community ... *94*
3.3.1.2 Perceptions of access to high quality care through research: Expectations of reciprocity or perceived coercion? ... *97*
3.3.1.3 Concerns about KGBC: Rumours and trust ... *98*
3.3.2 Being given negative results: Anxiety of waiting and relief of learning ... 99
3.3.3 Being given positive results ... 100
3.3.3.1 Three families with no family history of SCD and a completely well child ... *101*
3.3.3.2 One family with no family history of SCD and a child with relatively mild symptoms before screening in KGBC ... *103*
3.3.3.3 Two families with an older child already attending SCD clinic ... *103*
3.3.3.4 Two families with a child known to have SCD before participation in KGBC ... *105*
3.4 Moral concerns arising from community experiences of SCD and KGBC ... **106**
3.4.1 Balancing harms and benefits of information sharing on SCD in Kilifi ... 106

3.4.2 The distraction of SCD screening in diagnostic interpretations of KGBC..... 109
3.4.3 Participant-research relations: Trust and power..... 110
3.4.4 Not sharing information on carrier status: A justifiable 'deception'?..... 111
3.4.5 Accounting for community views on morally important issues: Important gaps and the need for further research..... 112
3.5 Conclusions..... **113**

CHAPTER 4 Phase Two: Discussing information sharing for study findings on sickle cell disorder: Families of affected children

4.1 Introduction: Background, aims and organisation of chapter..... **116**
4.2 Local experiences, arguments and values..... **117**
4.2.1 Reasons for sharing SCD information: Limiting the harms of 'not knowing'..... 117
4.2.1.1 Increasing the likelihood of effective care for affected children, their families and the wider community..... *118*
4.2.1.2 Countering harmful beliefs about supernatural causes of SCD..... *119*
4.2.1.3 Addressing gendered blaming..... *120*
4.2.1.4 Removing fears about HIV/AIDS..... *120*
4.2.2 Reasons against information sharing: The risk of generating harm..... 121
4.2.2.1 Generating anxieties about an inherited condition: Hopelessness and reproductive risks..... *121*
4.2.2.2 Generating or strengthening gendered blame, including for misattributed paternity..... *124*
4.2.3 The influence of context on potential outcomes of information sharing..... 127
4.2.3.1 Individuals and family relationships..... *127*
4.2.3.2 Observable patterns of sickle cell disease within families..... *128*
4.2.3.3 Economic circumstances..... *128*
4.2.4 Emerging values..... 129
4.2.4.1 Protecting vulnerable children and family stability..... *130*
4.2.4.2 Openness and empowerment..... *131*
4.2.4.3 Researchers' responsibilities..... *132*
4.3 Reflecting on agreement and difference on sharing SCD information with parents of affected children..... **133**
4.4 Conclusions..... **137**

CHAPTER 5 Phase Two: Discussing information sharing for study findings on sickle cell disorder: Families of children with carrier status

5.1 Introduction and background..... **141**
5.1.1 Carrier status and screening in the Kilifi Genetic Birth Cohort..... 141
5.1.2 Organisation of chapter..... 142
5.2 Local experiences, arguments and values..... **143**
5.2.1 Reasons for sharing carrier information..... 143
5.2.1.1 Knowledge can reduce the child's future reproductive risks..... *143*
5.2.1.2 Knowledge can help to prepare for and minimise harms in future..... *144*

5.2.1.3 Knowledge can create a welcome demand for improved services *146*
5.2.1.4 Knowledge can help in maintaining the health of a carrier child *147*
5.2.2 Reasons against sharing carrier information 148
5.2.2.1 Living with worries about uncertain future risks *148*
5.2.2.2 Misinterpreting carrier status as adversely affecting health *151*
5.2.2.3 Increasing stigma for affected children and families *152*
5.2.2.4 Creating suspicions of misattributed paternity *154*
5.2.2.5 Questioning the reliability of parents in passing carrier information to children *154*
5.2.2.6 Influence of HIV/AIDS control policy on understanding of SCD carrier status .. *156*
5.2.2.7 Supernatural beliefs and carrier status *157*
5.2.3 Individual and contextual influences in interpreting carrier status 158
5.2.4 Revisiting key values: Protecting families, openness and researchers' responsibilities 159
5.2.4.1 Protecting vulnerable children and family stability *159*
5.2.4.2 Importance of openness as empowerment for individuals and society *160*
5.2.4.3 Researchers' responsibilities *162*
5.3 Reflecting on agreement and disagreement in sharing carrier information for SCD **164**
5.4 Conclusions **167**

CHAPTER 6 Phase Two: Discussing requests for paternal testing for sickle cell status arising during research

6.1 Introduction to chapter **170**
6.1.1 Background and aims 170
6.1.2 Organisation of chapter 172
6.2 Local experiences, values and reasoning on request for paternal testing **173**
6.2.1 Reasoning against sharing misattributed paternity information: Risks of harms to children, mothers and families 173
6.2.2 Reasoning in support of paternal testing and sharing misattributed paternity results 174
6.2.2.1 The importance of testing fathers to counter paternal denial of responsibility *174*
6.2.2.2 Severe harm may not result: Possibilities of forgiveness and alternative resources for mothers *176*
6.2.3 Revisiting key values: Protecting families, openness and researchers' responsibilities 182
6.2.3.1 The importance of truth/openness *182*
6.2.3.2 Protecting vulnerable children and family stability *188*
6.2.3.3 Researchers' responsibilities *188*
6.2.4 The influence of HIV/AIDS on perceptions of SCD as 'an epidemic' 190
6.3 Reflecting on agreement and disagreement on paternal testing and sharing misattributed paternity results **191**
6.3.1 The work of facts and values: Supportive and conflicting 195
6.3.1.1 Balancing the interests of 'faithful' and 'unfaithful' mothers *197*

6.3.1.2 Withholding misattributed paternity results to protect the interests of 'unfaithful' mothers and their children ... *198*
6.3.2 Is paternal testing likely to be commonly requested? ... 201
6.4 Conclusions ... **202**

CHAPTER 7 Framing responsibilities for researchers towards information sharing on sickle cell disorder during studies in rural Kenya and similar settings

7.1 Background and aims of chapter ... **206**
7.2 How can perceived benefits of sharing SCD information generate responsibilities specific to researchers? ... **207**
7.2.1 A professional (and personal) responsibility to assist ... 209
7.2.2 Responsibility from trust in researcher-participant relations ... 211
7.2.2.1 Trust and perceptions of study aims ... *213*
7.2.2.2 Trust and expectations of due concern and reciprocity ... *214*
7.2.2.3 Reciprocity or perceived coercion? ... *215*
7.2.2.4 Risks of obfuscation in sharing carrier information ... *217*
7.3 Potential for conflation between the roles of researchers and providers ... **218**
7.4 Challenges to realising expected benefits of information sharing ... **222**
7.4.1 Communication challenges ... 222
7.4.2 The challenge of responding to requests for paternal testing ... 224
7.4.2.1 Is it reasonable to prioritise the interests of 'faithful' over 'unfaithful' mothers? ... *225*
7.4.2.2 Should researchers test fathers but withhold misattributed paternity results? ... *227*
7.4.3 The issue of resources: Influence on information sharing and partnership building ... 229
7.5 An influence from important emerging values ... **233**
7.5.1 The interests of vulnerable children ... 234
7.5.2 The stability of families ... 234
7.5.3 Openness as empowering individuals and society ... 236
7.5.4 Tensions between values of a duty of care and openness ... 237
7.6 Researchers' responsibilities for sharing SCD status information during KGBC and similar studies ... **238**
7.6.1 A strong responsibility to share SCD information with parents of affected children ... 238
7.6.2 Carrier findings: Balancing values ... 241
7.6.3 A strong responsibility to build partnerships with government health providers 243
7.7 Conclusions ... **244**

CHAPTER 8 Wider study implications: Settings, methods and further research

8.1 Introduction **248**

8.2 Key considerations for sharing SCD information in new studies and settings **248**

8.3 The work done by empirical methods in this thesis: Contributions and limitations **252**

8.3.1 Challenges of information sharing 253

8.3.2 Experiences of exploring diversity 254

8.3.3 Developing normative conclusions from community deliberations 255

8.4 Recommendations for further research **255**

8.5 Conclusions **258**

9.0 BIBLIOGRAPHY **259**

Appendices

Appendix A

A1: Phase One Interview Guide families with SCD/non KGBC Part 1 i

A2: Phase One Interview Guide families with SCD/KGBC Part 1 iii

A3: Phase One Interview Guide families with SCD Part 2 v

A4: Phase One Interview Guide families in KGBC/non SCD vi

Appendix B

B1: Phase One Coding framework vii

B2: Phase One: Sample Analysis Chart family interviews viii

Appendix C

C1: Phase Two Information sharing tools for staff: Fact sheet x

C2: Phase Two Information sharing for KCRs/affected mothers: Guide to information sharing xii

Appendix D

D1: Phase Two Discussion Guide/Scenarios for staff groups xiv

D2: Phase Two Discussion Guide/Scenarios for chiefs/KCRs/affected mothers' groups xvi

Appendix E

E1: Phase Two: Sample Analysis Chart KCR01 xviii

E2: Phase Two: Sample summary Analysis Chart KCR01 xx

E3: Phase Two Exploring themes (early) xxiv

E4: Phase Two Exploring themes (later) xxv

E5: Table 6.2 Summary of agreement and difference *by group type* for responses to requests for paternal testing xxvi

Appendix F

Figure 8.2. Assessing researchers responsibilities for sharing SCD information during studies: Questions about context and their implications xxxi

Tables and Figures

Tables

		Page
Table 2.1	Kilifi Genetic Birth Cohort field workers in Phases One and Two	53
Table 2.2	Phase One participants: Families	53
Table 2.3	Community members participating in Phase Two	65

Figures

Figure 1.1	Map of Kenya showing Kilifi District	16
Figure 3.1	Summary of the questions from Phase One taken forwards to Phase Two	117
Figure 4.1	Agreement and disagreement on 'facts' and values in sharing SCD information	136
Figure 4.2	Summary of community views on sharing SCD information in studies	142
Figure 5.1	Agreement and disagreement on facts and values on sharing carrier information	168
Figure 5.2	Summary of community reasons for sharing carrier information during studies	171
Figure 6.1	Agreement and disagreement on facts and values on paternal testing	195
Figure 6.2.	Summary of community reasoning about paternal requests for testing	207
Figure 7.1	Summary of researchers' responsibilities for sharing SCD information during KGBC	249
Figure 8.1	Key considerations for sharing SCD information in studies: Questions about context	252

Acknowledgements

I have many people to acknowledge for their support in the research for and writing of this thesis. In part this is because my ideas inevitably draw on experience in research and community engagement over many years of working at the KEMRI Wellcome Trust research programme in Kilifi alongside many inspiring colleagues and friends. Very key and specific support for the thesis has been given by staff in the Social and Behavioural Research and Community Liaison groups in the programme, particularly Isaac Charoh, Anderson Kahindi, Jane Kahindi, Gladys Sanga and Duncan Ngome who helped me with data collection, translation and transcription; Metrine Saisi, Willhelmina Mwamuye and Chrisbel Kanini who undertook many hours of transcribing; and Francis Kombe whose help was essential for the discussion methods used in this research, and who has also worked with me on analysis and writing. I am also grateful to Dr Thomas Williams for his help and encouragement, as the principal investigator for the Kilifi Genetics Birth Cohort study. At the heart of this study are the contributions of many participants - all local residents of Kilifi district - who have given their time, energy and careful thoughts to this project, for which I am very grateful. I thank the Kenya Medical Research Institute and the Wellcome Trust in the UK for providing funding for the research undertaken. Particular thanks go to my supervisors, Professor Michael Parker, Professor Ray Fitzpatrick and Dr Sassy Molyneux for their support, inspiration and continuous interest in and enthusiasm for this project. Finally to my family and friends, thanks as always.

CHAPTER 1

Introduction, Background and Aims of the Thesis

1.1 Introduction

In 2008, a large scale population-based genomic epidemiological study began at an established biomedical research centre in Kilifi district in Kenya. The study was preceded by several months of discussions with community leaders and representatives, and a series of community outreach meetings across the area where it would be conducted. Challenges in explaining this relatively complex study to potential participants and the wider community had been expected; these continued to require continuous community engagement support (Marsh, Kamuya et al. 2010). However, an unexpected feature emerged over time, leading to a dilemma for researchers around plans for sharing findings on sickle cell disease (SCD) generated as part of the genomics study. This dilemma forms the core of the enquiry undertaken in this thesis.

The genomics research was the Kilifi Genetic Birth Cohort study (KGBC), which aimed to examine susceptibility and resistance to a range of serious illnesses affecting children in Kenya and other developing countries. Predictive screening for SCD had been included as a form of benefit for participants and the community[1], given the recognised seriousness of this diagnosis[2]. Clinical services were available at Kilifi District Hospital, provided collaboratively by researchers and hospital staff. In KGBC, SCD findings for affected children were returned to participant families, with referral to the specialist clinic. After several months, however, it became clear from records that few families were bringing their child to the clinic for test validation, counselling and management of the disorder. Researchers and community engagement staff began to ask questions that led to the development of research in this thesis: Why was use of the SCD clinic so low

[1] These activities were developed and approved through processes of institutional, national and international scientific and ethical review, explained in more detail in section 1.2.1.
[2] See section 1.4 on sickle cell disease in Africa.

amongst participants? And, more fundamentally, what did this mean for the balance of benefits and harms to participants, and for the responsibilities of researchers in relation to sharing SCD information as part of the genomics study? Behind these questions lie three larger ethical[3] themes, explored in more detail in the thesis: ethical issues in international collaborative biomedical research; concerns in genetics and genomics research; and the value of empirical research in addressing ethical questions, including as a form of community[4] consultation.

In relation to the third theme, for many types of research, but particularly for international collaborative research, there has been wide recognition of the importance of community consultation to support the ethical conduct and scientific validity of research (Sharp and Foster 2000; Weijer and Emanuel 2000; Marshall and Rotimi 2001; Emanuel, Wendler et al. 2004; Gikonyo, Bejon et al. 2008; Fabsitz, McGuire et al. 2010). There is much less clarity on methods that should be used in these consultation processes. Deliberative approaches to public consultation have been used as a way of democratising and strengthening the ethical basis of normative decision making processes[5] in developed countries, particularly where the issues under consideration are contentious and/or relate to public policy (Abelson, Forest et al. 2003; Burgess 2004; Kim, Uhlmann et al. 2010). In general, debate around these methods - as for community engagement more broadly - is often underpinned by different views on the goals of consultation, including the extent to which decision-making might be shared with community members (Sharp and Foster 2000; Marshall and Rotimi 2001; Dickert and Sugarman 2005). Challenges have also been raised about which individuals could most appropriately 'represent' wider communities from which they are drawn (Juengst 2000; Weijer and Emanuel 2000).

[3] In this thesis, I use the terms 'ethical' and 'moral' in the following way. 'Ethical' is used where the concept is linked to formal reasoning in the bioethics literature on principles in research and health care. 'Moral' is used either to refer to less formal 'norms' that, for example arise from people's everyday experiences, or to higher level ethical principles.

[4] The term community is used here to denote the population of local residents who live in the catchment area of Kilifi District Hospital, cognizant of the many ways in which this term can be used, the nature of 'communities' as fluid and overlapping, and that the term itself embodies normative influences in drawing boundaries (see also Marsh et al 2011a).

[5] See Chapter 2 section 2.4.1.2

Illustrative of these challenges, questions about the value of sharing SCD information had not emerged through community consultation activities undertaken during planning for KGBC in Kilifi. From these experiences, it seemed likely that consultation on unfamiliar or potentially controversial aspects of research should include more deliberation, information sharing and structured probing (Marsh, Kamuya et al. 2010). One aim of the thesis is to explore such an approach to community consultation, including an initial phase to explore the context in which information sharing occurs; and a second stage of informed deliberation by community members. Chapter 2 provides a detailed account of these methods and their theoretical basis as a form of empirical ethics.

The following three sections in this chapter will provide background information on the setting for this study, including the KEMRI Wellcome Trust Research Programme (1.2); the specific genomics study that acted as the entry point for empirical research (1.3); and sickle cell disease in Africa (1.4). Building on these descriptions, section 1.5 will describe the overall and specific aims of the thesis. The three sections after these provide information from the literature that will be drawn upon in the thesis: ethical issues in genetic screening and testing (1.6); disclosure of genetic findings in research (1.7); and researchers' positive responsibilities towards providing care or services for participants and communities (1.8), with a focus on international research in developing countries. The final section (1.9) summarises key points from the chapter and provides an overview of the organisation of the remainder of the thesis.

1.2 The KEMRI Wellcome Trust Research Programme in Kilifi

1.2.1 The research programme

The Kenya Medical Research Institute (KEMRI) Wellcome Trust Research Programme was officially formed as a collaboration between KEMRI and the Wellcome Trust on the basis of a

Memorandum of Understanding in 1989. KEMRI[6] is a parastatal organisation within the Kenya Ministry of Health, mandated to undertake locally relevant biomedical research. The Wellcome Trust is one of the major international funders of research in biomedicine and the medical humanities, established as a charitable organisation in 1936 in the UK[7]. The KEMRI Wellcome Trust Programme currently includes a wide range of collaborative partners in Africa, both within and outside Kenya, as well as in other parts of the world.

The main site for this programme remains within Kilifi District Hospital, entailing close collaboration between researchers and the district Ministry of Health managers and providers, for example with the District Health Management Team responsible for promotive, preventive and curative services in the district, and the Hospital Management Team, responsible for district hospital services. Within this collaboration, the research programme provides long term support to many areas of clinical service at the hospital and surrounding peripheral health facilities where research is conducted. This *core support* is seen as generating a supportive environment for research in this setting, including ensuring that adequate health care is available to local residents independent of their involvement in research, that clinical researchers can respond to their ethical responsibilities as physicians as well as researchers, and that specific types of clinical services needed for different studies are provided in a way that supports rather than undermines local health service provision. Of direct relevance to this thesis, the sickle cell clinic at Kilifi District Hospital represents one such collaborative service, including the provision of testing (and validation) by haemoglobin electrophoresis, and counselling and care for the disorder by a research medical officer at a dedicated weekly clinic.

[6] Also see www.kemri.org
[7] Also see www.wellcome.ac.uk.org

Research in the programme is collaborative and multidisciplinary[8]. In Kilifi, several studies are generally in progress at the same time. A key feature is a linked system of clinical and population-based demographic surveillance. Clinical data from most admissions at Kilifi District Hospital are electronically linked to information from a regular population-based census - the Kilifi Health and Demographic Surveillance System[9] (KHDSS) - in the main geographic catchment area for the hospital. A population of around 260, 000 people live in this area, of approximately 891km^2 (Cowgill, Ndiritu et al. 2006).

As part of KEMRI, new proposals for research are reviewed by an institutional scientific coordinating group and national scientific and ethical review committees as well as by international bodies, where the latter are required by sponsors. Community engagement is a prominent activity in the programme, seen as processes of interactivity between researchers and community members that support consultation and information sharing in relation to the research progamme overall (including over time) and for individual studies. For specific studies, interactivity is supported at all stages of the research cycle, including research design and planning; community outreach; informed consent processes; responding to questions and concerns throughout studies; feedback of findings, where relevant; and closing down studies (Marsh, Kamuya et al. 2008; Marsh, Kamuya et al. 2010; Marsh, Kamuya et al. 2011a). A team of five full time community facilitators supports these processes of engagement, working with - amongst other community 'links' - a large group of volunteer KEMRI Community Representatives (KCRs). KCRs are selected by their local communities (usually one person for a group of villages) through public meetings organised by the local administrative chief, and have a role to support consultation on research-related issues through regular or *ad hoc* meetings with community liaison staff and/or researchers. Their ability to make inputs in this way is supported through annual workshops covering aspects of research and

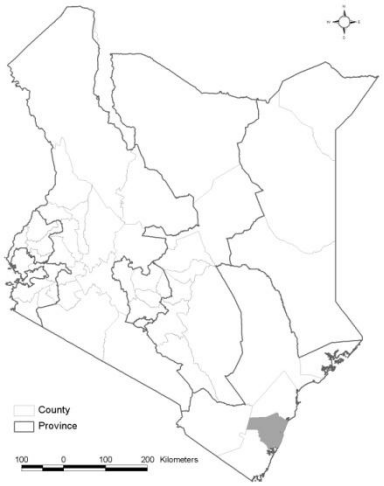
[8] See www.kemri-wellcome.org
[9] The KHDSS is part of INDEPTH, a global network of members who conduct longitudinal health and demographic evaluation of populations in low- and middle-income countries (http://www.indepth-network.org)

the research institution, including ethical review processes. 170 community members were elected as KCRs for a period of three years at the last round of elections in 2011.

Training and support supervision of staff at the research centre who work at the interface with community members is part of this agenda of community engagement work (Molyneux, Kamuya et al. 2010; Marsh, Kamuya et al. 2011b). A key group of staff are called 'field workers', being local residents with at least 12 years of formal education who are employed full time in the research programme. For many studies conducted within the community, and for some in clinical settings, field workers undertake informed consent processes and conduct study procedures such as collecting information, taking measurements and capillary blood sampling. Within this role, their familiarity with local languages and culture are seen as essential skills. As will be seen later, community facilitators, KCRs and field workers have been important contributors to the discussions described in Chapters 4, 5 and 6 in this thesis.

1.2.2 Kilifi district and local residents

Figure 1.1: Map of Kenya showing Kilifi District (shaded)



Kilifi district[10] is part of the coastal province of Kenya, bordering the Indian Ocean along an approximately 60 km coastline (see Figure 1.1). The district lies close to the equator (around latitude 3º S and longitude 40 º E), the highest part being approximately 700 metres above sea level. The population is approximately 653,144 of which around 34,000 people live in Kilifi town, the district headquarters. A deep sea creek is a major feature of the town, and fishing and subsistence farming form the major livelihoods in the district alongside low level tourism,

[10] The term 'district' is about to be converted to one of 'county' as part of the introduction of changes to governance in the new Kenyan constitution. In this thesis, I use the existing (old) boundaries and populations in describing the district. The new county will have a population of around 1.1 million.

although there is increasing prominence of service industries in Kilifi town. The research centre employs the largest number of people in the town. In 2009, a college of further education, the Kilifi Institute of Agriculture, was upgraded to Pwani University as part of one of the main national universities (the Jomo Kenyatta University of Agriculture and Technology) with a student body of 2,500 from across the country.

More inland, focal areas of coconut farming provide relatively high return on land use. The remainder of fertile land is used in subsistence farming, primarily of maize, beans and green vegetables. Particularly arid central areas only support livestock, mainly goats, which can be sustained on sparse and otherwise inedible vegetation, and charcoal making. According to surveys in 1999, parts of Kilifi district were amongst the most poor in Kenya, with reported high depth as well as incidence of poverty: three-quarters of the population falls below the poverty line in 24 out of 34 locations'[11] (CBS, World Bank et al. 2005). More recently, a consortial spatial mapping initiative in Kenya described that members of more than 65% households in Kilifi in much of the district live below the poverty line[12], and between 55% and 65% of households do so within the remainder (Virtual Kenya 2011).

The main population group in Kilifi district is the Mijikenda, comprising seven subgroups of which three - Giriama, Chonyi and Kauma - form the local majority, dominated numerically by the Giriama. Along the coast there is a relatively high influence of Islam, but across the district the majority population describe themselves as Christian (47%), compared to 13% Muslims and 24% traditionalists[13] (Kamuya 2007) . The people of Kilifi district are served by the main district general hospital in Kilifi, a sub district hospital in a rural part of the district (Ganze), and 30

[11] A 'location' is a subdivision of a district, which number 16 in Kilifi.
[12] In the CBS/World Bank report referenced, the poverty line was 'determined and based on the expenditure required to purchase a food basket that allows minimum nutritional requirements to be met (set at 2,250 calories per adult equivalent per day) in addition to the costs of meeting basic non-food needs...for urban and rural households respectively' (p8).
[13] 14% described other religions, and no response was indicated for 4% in this round of the KDHSS in 2007.

peripheral health facilities[14]. For many people in remote areas, village shops provide an important source of over-the-counter medication (Snow, Peshu et al. 1992; Marsh, Mutemi et al. 1999). As I will go on to describe in Chapter 3, traditional healers of different types are also important health providers within the district (Mwenesi, Harpham et al. 1995). Free primary school education was introduced as a national policy in Kenya in 2003. However, 45% of adults reported being unable to read a newspaper or letter during a random survey of 329 local residents, conducted as part of community engagement planning at the research centre in 2005.

1.3 The Kilifi Genetics Birth Cohort

The Kilifi Genetics Birth Cohort study (KGBC) was the genomic epidemiological study around which the empirical data in this thesis was collected. KGBC aimed to assess genetic susceptibility and resistance to several common serious illnesses in children through genomic analysis of samples from children matched to phenotypic data from ward admissions at the district hospital. This passive longitudinal follow up study was based on the linkage between clinical surveillance and the KHDSS. While working on a broader range of diseases of interest, KGBC was also a partner in the MalariaGEN consortium, involving collaborators from 27 different countries, to study genomic influences on the risks of childhood malaria, including severe disease and death. The MalariaGEN consortium placed ethical, legal and social issues in research as a central focus, to explore in depth and to set up systematic ways of addressing these concerns for specific consortial studies (Chokshi 2005; Chokshi, Parker et al. 2006; Chokshi, Thera et al. 2007; Parker, Bull et al. 2009). The MalariaGEN group provided support to researchers and ethics review committees in partner countries in identifying and addressing issues that were seen as particular concerns for this type of research, including for KGBC.

[14] 28 are dispensaries, run by a nurse; and 2 are health centres, where the presence of a clinical officer increases the range of services provided. Clinical officers are a paramedical cadre of staff with at least 4 years of medical training. They provide a major element of clinical services in the government health sector in Kenya.

In KGBC, healthy children under one year of age were recruited through visits to their homes for a single capillary blood sampling between late 2007 and 2011. Since follow up for the genomics study was passive, the only further contact with participating families was to return findings of SCD screening. During the development and review of the proposal for KGBC, which included consultation with community leaders and KCRs as well as institutional, national and international stages, the *initial* plan for sharing SCD information was that:

- Findings in children found to have the disease (around 1%) were to be shared with parents, with referral to the SCD clinic at Kilifi district hospital for validation of the test, counselling on the findings and initiation of long term management. This decision was based on recognition of the serious nature of this condition for the health of the child, and the improved prognosis associated with early diagnosis and management[15];
- Findings on carrier status (affecting around 18% children) and normal haemoglobin (around 81%) were not to be returned[16]. The basis for this decision was the relatively benign nature of carrier status[17]; low awareness of the disorder in the community; the lack of clinical or social services to support information sharing, including the absence of a national or district SCD health policy or public access to screening[18]; and the research resources that would be needed to validate and provide counselling on findings in a relatively high proportion of the population.

After initiation of the study, however, there were strong requests from the community, via KCRs and study fieldworkers, for results to be actively returned *in all cases* since parents were concerned that positive results might accidentally go astray, leaving them unaware of this serious condition.

[15] See section 1.4 on sickle cell disease.
[16] As a result, the concept of carrier status for the condition was not specifically explained either as part of the feedback of findings, nor during the informed consent process.
[17] The main agreed importance of being a carrier for SCD is an increased risk of future children (of a carrier) being affected by the disease, where both parents are carriers. This is described as an increased reproductive risk.
[18] Only physician-led diagnostic testing was available at Kilifi district hospital. See section 1.4 on sickle cell disease.

The feedback plan was therefore changed so that all findings were actively taken back to the home, but that carrier and negative findings were both explained as 'not SCD'. This approach was recognised by researchers as a compromise between responding to reasonable community requests for universal feedback of findings and the many reasons, given earlier, that returning carrier findings was seen as unlikely to be helpful. Given the resources needed for this feedback exercise, non urgent results were distributed with the next round of the census, meaning up to four months delay in returning results for the majority.

1.4 Sickle Cell Disease in Africa

Sickle cell disease (SCD) is one of the most common and serious single gene disorders affecting people's health worldwide, with its central impact on the structure of haemoglobin within red blood cells. It has been estimated that between 300,000 and 500,000 children are born with SCD worldwide every year (Weatherall, Akinyanju et al. 2006), and that three quarters of these births occur in sub Saharan Africa (Sadarangani, Makani et al. 2009). Its high incidence in this part of the continent is accounted for by a well recognised evolutionary link between SCD and increased resistance to malaria. In these areas, there is little evidence on the prevalence, patterns and burden of the disorder and frequent calls for more research (Weatherall and Clegg 2001; WHO 2002; Makani, Williams et al. 2007). In addition, it has been noted that its relatively high prevalence has been masked by the much greater burden of infectious diseases and conditions such as malnutrition in these settings. Weatherall and Clegg (2001) point out that genetic diseases account for about 40% of the requirements for chronic care in paediatric practice in developed countries, and that a similar pattern is likely to emerge in developing countries in future given the 'unusually high frequencies of haemoglobin disorders' (Weatherall and Clegg 2001) (p711).

SCD is inherited as an autosomal recessive condition. Affected children inherit two copies of the abnormal haemoglobin gene, one from each parent. The condition is generally[19] transmitted from one generation to the next through individuals who are healthy but carry one copy of the abnormal gene. The situation of carrying one abnormal sickle cell gene is known as carrier status. Where both parents of a child have a single abnormal gene, there is a 1 in 4 chance that any of their children will inherit two copies of the gene, resulting in the disorder. There is a 1 in 2 chance that any of their children will inherit one copy of the abnormal gene from a parent, leading to transmission of carrier status. In fact, SCD refers to a group of conditions with three main variations described by molecular differences in the structural haemoglobin defect; most commonly the underlying defect results from the presence of two sickle haemoglobin genes (HbSS), but the syndrome can also be produced where a sickle haemoglobin gene is combined with another abnormal haemoglobin gene, giving rise, for example, to haemoglobin SC (HbSC) or haemoglobin Sβ thalassaemia (HbSβ) (Williams, Mwangi et al. 2005).

The physical effects of SCD are largely caused by shortened survival of red blood cells and a tendency for these abnormal cells to block small blood vessels. The disorder leads to chronic anaemia and can cause multisystem effects, including attacks of bone pain, sequestration of blood into the lungs, liver or spleen, or thrombosis of cerebral vessels leading sometimes to stroke (Weatherall, Akinyanju et al. 2006; Makani, Williams et al. 2007). However, there is considerable variation in the severity of physical symptoms, with both environmental and genetic influences. Studies have shown, for example, that some of the difference in severity is linked to variable expression of the HbSS gene (Chui and Dover 2001).

The mainstay of SCD management in developed countries includes early detection through neonatal screening, comprehensive care programmes and early detection and management of crises, with more recent therapeutic interventions of blood transfusion therapy and the use of

[19] Affected individuals as well as carriers can also pass the gene on to their children.

hydroxyurea. Where these programmes are in place, adult survival has reached a median of 48 years (Makani, Williams et al. 2007). In the absence of preventive strategies and management of the complications of this disorder, as is common in many part of sub Saharan Africa, it is likely that very many children die of this condition undiagnosed (Weatherall and Clegg 2001; WHO 2002). Amongst 124 children attending SCD clinic in Kilifi followed up over 13 months, two died and seven were lost to follow up (Sadarangani, Makani et al. 2009). Highlighting a lack of evidence, Makani (2007) notes that the prevalence of SCD in older people in some malaria endemic parts of Africa is surprisingly high given presumed mortality rates in childhood (Makani, Williams et al. 2007).

Although more common than SCD, carrier status is generally seen as a benign condition (Ohene-Frempong 2005), with physical complications being described as uncommon or mild (Kark 2000). Relevant for malaria endemic settings, presence of the gene majorly reduces risks of mild and severe forms of malaria[20] (Williams, Mwangi et al. 2005). The evidence surrounding other physical effects is to some degree mixed (Tsaras, Owusu-Ansah et al. 2009); and the possibility of a psychosocial component is well recognised (Fullwiley 2006). *The main agreed importance for carrier status is the risk of having a child with SCD in future, where both parents are carriers* (WHO 2006). For this reason, premarital carrier screening programmes for SCD have been set up in some parts of the world in order to allow individuals to understand their risk, mainly in the Middle East (Alswaidi and O'Brien 2009; Hartley 2009; Memish and Saeedi 2011) and to a lesser extent in parts of West Africa (Bazuaye and Olayemi 2009; Ghana Sickle Cell Foundation 2010; Nigeria Sickle Cell Foundation 2011). In addition, worldwide, SCD screening targeting couples before marriage has become increasingly available through the commercial sector (Eastern Biotech and Life Sciences 2011).

[20] This phenomenon is the basis of its evolutionary advantage, and therefore prevalence, in malaria endemic areas.

Where SCD screening or testing is undertaken in children, however, the main objective is generally the detection of the disease, and in this context carrier status is an incidental finding[21] (Wolf, Paradise et al. 2008) generated by the fact that information on SCD and carrier status cannot be established independently using the common screening methods (Laird, Dezateux et al. 1996). In recent years, wide support for newborn SCD screening programmes has followed clear evidence that early initiation of management has an important positive impact on outcome, including the chances of survival (NIH 1987; King, Fraser et al. 2007; Telfer, Coen et al. 2007). As a result, newborn screening programmes including SCD are now in place in many parts of Europe, the Americas and the Middle East (Bain 2009). Newborn SCD screening has also been established in parts of Ghana, (Ohene-Frempong 2005) and there are reports of pilot studies from other countries in Africa (Kafando, Sawadogo et al. 2005; Munyanganizi, Cotton et al. 2006; Mutesa, Boemer et al. 2007; Rahimy, Gangbo et al. 2009). Worldwide, carrier status may therefore be most commonly discovered incidentally during screening for SCD in newborn infants.

In relation to the disorder and carrier status, their known and different clinical importance, as described in this section, underpinned decisions on sharing these findings during KGBC. The subsequent and unexpectedly low uptake of referral for care for the disorder generated the dilemma around SCD information sharing outlined at the start of this chapter, and questions on how researchers should handle information of this nature.

1.5 Thesis objectives

Against the background of a genomics study in Kilifi, the broad aim of the thesis is to develop appropriate responses to important ethical questions about disclosure of SCD findings during

[21] Wolf (2008) defines an incidental finding as "a finding concerning an individual research participant that has potential health or reproductive significance and is discovered in the course of conducting research but is beyond the aims of the study."

international collaborative biomedical research, including the responsibilities of researchers in this regard.

Specific aims are:

- To identify the moral concerns of participants and community members around participation in a genomics study in Kilifi through an exploration of community perceptions, beliefs and experiences of participating in the research and sickle cell disease, including where screening was conducted through participation in research;
- To build a 'bottom up' normative analysis of the moral concerns identified, including the ways in which the perceived value of sharing SCD findings in research can be taken to inform responsibilities for researchers;
- To propose specific recommendations on sharing SCD information (for affected children and those found to be carriers) for the genomics study, including their ethical basis; and key considerations in planning for SCD information sharing in other types of studies and different malaria endemic settings;
- To explore the potential contribution of descriptive and deliberative empirical approaches to resolving ethical dilemmas in research, including as a form of community consultation.

1.6 Literature on ethical issues in genetic screening and testing

As background to an enquiry into ethical concerns for sharing genetic information in research in this thesis, I begin by describing some wider issues raised for genetic screening and testing, separate to questions of disclosure. Genetic *screening* is described as a process of looking for the genetic basis of disease in subpopulations at risk (which could include non genetic tests, such as biological indicators), while genetic *testing* - drawing on a 1997 report from the UK Advisory

Committee on Genetic Testing[22] - aims to detect the presence or absence of, or change in, a particular gene or chromosome (WHO 2006).

Many social and ethical issues have been debated internationally for genetic screening or testing, for the 'post genomics' era (Collins 1999; Hoedemaekers, Gordijn et al. 2006; Parker, Bull et al. 2009) that began with the initiation of the Human Genome Project in 1990 and in the field prior to these developments (Markel 1992). Most commentaries and guidelines on genetic screening or testing draw on developed country experience (Richards 1993; WHO 2002; NCOB 2006), with the notable exception of a 2006 World Health Organization (WHO) report on Genetic Services in Developing Countries (WHO 2006). This report outlines seven key areas for ethical issues in developing countries, including distributive justice; the social context in developing countries related to genetic testing and screening, including consanguineous marriage, religion and gender; risks of stigmatisation and discrimination; protection of privacy and confidentiality; facilitating patient decision-making; the importance of education; and patient safety and quality assurance in genetic testing. Within these themes, specific issues include the implications of: consanguineous marriage; sex selection and bias against female children; directive genetic counselling and traditions of paternalism in medicine; discrimination and stigmatisation on the basis of carrier status for recessive genetic conditions; and the role of commercial genetic testing.

Stigmatisation and discrimination in genetic testing emerge as important concerns across the literature in all settings and at different times, at the level of the individual within a family, for the family within a wider community and for communities themselves (Phoenix, Lybrook et al. 1995). Much of the initial recognition of the risks of discrimination through genetic testing arose from experiences of sickle cell disorder testing in programmes in the 1970s in the USA. Collectively, these programmes have been described as 'a public policy disaster' (Duster 2003) and as a

[22] Advisory Committee on Genetic Testing (1997) *Code of Practice and Guidance on Human Genetic Testing Services Supplied Direct to the Public* (London: Health Departments of the United Kingdom).

Dickensian axiom of 'How Not To Do It' (Markel 1992). Greater understanding of the way in which stigmatisation can occur in genetic conditions, including testing, is important in developing strategies to counteract risks, including counselling, informed consent and disclosure processes. This has been emphasised in developed country settings as part of the introduction of SCD screening within public health programmes (Borry, Nys et al. 2007; Miller 2009).

There is a rich literature from social science around the social meanings and impacts of genetic knowledge on concepts of family and kinship. Much social science literature in the 1990's, again following the mapping of the human genome, paid increased attention to the influence of genetic understandings on social domains, including the family. This focus on the role of genetic knowledge led to the development of terms such as 'geneticisation' to refer to an increasing trend to account for human problems as genetic in origin (Lippman 1992; Featherstone, Atkinson et al. 2006), alongside concepts of 'medicalisation', described as 'drawing into the biomedical domain of physical aspects and behaviors such as antisocial conduct that could be understood alternatively as a sin, a crime, a moral fault, or a disease' (Finkler 2005) (p175). As Conrad and Gabe (1999) wrote: 'The *rising genetic paradigm* is influencing how we think about life, including disease and disability, human capacities and failings, social problems, kinship and the quality of life." (Conrad and Gabe 1999) (p3).

More recently, social scientists have reflected on more complex and interrelated ways in which concepts of genetics, family and kinship seem to work together in practice. In a recent special edition of Social Science and Medicine (2011), authors have described a 'post genomics' nature for these new interpretations, including the importance of pre-existing family dynamics on the way that new genetic knowledge is interpreted and given form (Chilibeck, Lock et al. 2011); co-production between social and genetic influences, and the role of numerous mediating factors, including specific characteristics of the national or local health care systems and political culture (Horstman and Finkler 2011); and the importance of features of the genetic condition under

consideration as well as the work done by families to limit the potential for genetic knowledge to become 'all-encompassing' (Geelen, Van Hoyweghen et al. 2011). In keeping with notions of complexity and the influence of many variables - including the nature of the genetic disorder - in 'post genomics' theories, Tekola (2009) describes community views on the importance of wide family consent for research on the heavily stigmatised genetic condition of podoconiosis in Ethiopia (Tekola, Bull et al. 2009).

There is however relatively little empirical data on experiences of living with genetic disorders or genetic testing from Africa. The 2006 WHO report on genetic services in developing countries illustrates this gap well; only 4 of its 278 referenced articles provide reports from this continent. Much of the social science literature is based on studies in developed countries (Rapp 1988; Featherstone, Atkinson et al. 2006), some with specific attention to issues of discrimination in high risk communities (Atkin, Ahmad et al. 1998; Shaw 2006), but much of the data on experiences of genetic conditions in Africa draws on quantitative surveys and illustrates issues in limited geographic areas, particularly West Africa (Bamisaiye, Bakare et al. 1974; Akenzua 1990; Ohaeri, Shokunbi et al. 1995; Adeodu, Alimi et al. 2000; Assimadi, Gbadoe et al. 2000; Ohaeri and Shokunbi 2001; Ohaeri 2002). Exceptions - apart from the study in Ethiopia described above (Tekola, Bull et al. 2009) - are qualitative accounts of SCD beliefs as reincarnation in Nigeria (Nzewi 2001) and Ghana (Allotey and Reidpath 2001), a description of biosocial illness associated with SCD carrier status in Senegal (Fullwiley 2006) and fathers' experiences of SCD in Ghana (Dennis-Antwi, Culley et al. 2011) highlighting changing knowledge and attitudes towards the disorder.

1.7 Literature on the disclosure of genetic findings in research

Given central questions for the thesis on sharing SCD findings during research, the literature and guidelines for researchers on the disclosure of genetic findings in studies form an important

component of the background literature. There are a number of international guidelines for the ethical conduct of research involving human subjects that outline researchers' responsibilities in relation to sharing genetic results generated during the course of studies. A review of these recommendations was undertaken by Knoppers in 2006, showing that consensus has moved over time towards recommending greater openness in relation to disclosure of findings (Knoppers, Joly et al. 2006). Knoppers suggests this position has, in part, been relatively recently driven forwards by the advent of large scale population based genomic epidemiological studies, of which KGBC is an example. Knoppers also shows that this movement represents a general shift in attitudes towards greater disclosure of individual findings across all types of research. To illustrate this point, Knoppers compares the Council for International Organizations of Medical Sciences (CIOMS) International Guidelines for Ethical Review of Epidemiological Studies in 1991 and 2002, describing that the earlier guidelines 'maintained that being informed of findings 'that pertain to their health' is one of the 'reasonable' benefits of participation for 'communities, groups and individuals' in research' while the 2002 version states that 'individual subjects will be informed of any finding that relates to their particular health status' (Knoppers, Joly et al. 2006)(p1173).

Thus, as commentators have both lauded and opposed, there has been a shift towards seeing disclosure of individual information generated in research as a greater positive responsibility for researchers, drawing on principles of respect for persons and beneficence, and more controversial ones of reciprocity and justice (Fernandez, Kodish et al. 2003; Ravitsky and Wilfond 2006). Arguments against disclosure of genetic findings have also drawn upon principles of justice, as fair utilisation of research resources given the costs of disclosure, and risks of promoting a therapeutic - or more accurately a diagnostic - 'misconception' of research, again with a potential to undermine autonomy.

In this way, although many commentators agree that there are ethical principles that support the importance of disclosing genetic findings in research, these have been argued as insufficient to

generate an obligation for researchers to share findings[23] (Ossorio 2006). Therefore other opposing principles - or interpretations of the same principles - could be considered to carry more weight. To illustrate, Ravitsky (2006) draws on a principle of justice to prioritise the autonomy and interests of individual participants, while Ossorio (2006) uses the same principle to prioritise society's 'long term interests in advancing knowledge' over 'participant's short term interests in obtaining particular individual medical information.' (Ossorio 2006) (p25).

While this literature seems primarily to concern researchers' responsibilities in developing country settings in relation to genetic findings[24] with less obvious clinical significance than SCD, it is useful to questions about information sharing on this disorder in Kilifi in four main ways. These are described in the following paragraphs.

1.7.1 Understanding the central role of 'clinical utility' for researchers' responsibilities

The literature pins down a core agreement on the critical influence of 'clinical utility'[25] in determining the strength of a researcher's responsibilities to share genetic findings in studies (Knoppers, Joly et al. 2006; Ravitsky and Wilfond 2006; Fabsitz, McGuire et al. 2010). In fact, whether or not these are genetic, a criterion of clinical usefulness seems to cross-cut researchers' responsibilities to share individual findings during studies. A typical description of a responsibility for information sharing in relation to clinical usefulness is given in the WHO 2003 report on 'Genetic Databases: Assessing the Benefits and the Impact on Human and Patient Rights' which states that information should be shared where 'the data have been instrumental in identifying a

[23] As implied here, in this thesis I will use the term 'obligation' to imply an absolute requirement, whereas a 'responsibility' suggests a variably strong role in undertaking an action. A very strong responsibility (for example, for SCD testing or sharing information) that is not opposed by equally strong reasoning against this action could arguably raise a responsibility to the level of an obligation.

[24] I also note commentaries on the need to avoid genetic exceptionalism, as potentially strengthening common community perceptions about the 'fundamental' nature of genetics at the expense of understanding equally important environmental influences. These commentators have called for the same policies to be used for the disclosure of genetic and non genetic findings in research (Sharp 2006).

[25] In this thesis, I will generally use the words 'usefulness', 'importance', or 'benefit' to capture the meaning of utility as described in the literature on disclosure of genetic findings, since utility may be taken to imply an association with the ethical theory of utilitarianism.

clear clinical benefit to identifiable individuals; and the disclosure of the data to the relevant individuals will avert or minimise significant harm to those individuals' (WHO 2003)(p23). Clinical usefulness of genetic findings is also widely interpreted to include future reproductive benefits. For example, Ravitsky's (2006) definition includes results expected to be valuable to participants' physical or psychological well being, to their reproductive decision making or to their life planning (Ravitsky and Wilfond 2006). Further, most definitions of this value work with an assumption of a valid testing process, itself a criterion that draws on a long established requirement in medical testing in general (Fabsitz, McGuire et al. 2010).

One outstanding issue for clinical usefulness in sickle cell disorder that deserves flagging is controversy in the literature over the health implications of carrier status. Current evidence suggests health risks are uncommon and serious complications are very rare. For example, a recent review in the American Journal of Medicine (Tsaras, 2009) concludes that there is convincing evidence of an association between carrier status and certain very rare but serious conditions[26], but no evidence of an association with other important less rare conditions or of an impact overall on life expectancy (Tsaras, Owusu-Ansah et al. 2009). However, some patient information sources point to a risk that people with carrier status may develop symptoms, including when exposed to low oxygen levels during an anaesthetic or in a depressurised aircraft at altitude. For example, the National Health Service (UK) Sickle Cell and Thalassaemia Screening programme's public information area of their website explains to parents of a baby found to be a carrier for SCD:

> "Your child is healthy and will not need any treatment, although carriers of sickle cell will need to take extra care when having a general anaesthetic or in situations where there might be a lack of oxygen (like deep sea diving or high altitude mountaineering)."[27]

More evidence on the health implications of carrier status could change perceptions of its clinical and social importance both for community members and researchers[28]. It is notable that, apart from

[26] For example, a higher incidence of a rare form of renal cancer.
[27] http://sct.screening.nhs.uk/cms.php?folder=2446 accessed 26/09/2011

a report on 'biosocial suffering" in Senegal (Fullwiley 2006), there is little published evidence on the health or social implications of carrier status in Africa, where conditions of dehydration and extreme physical exertion that have been proposed as risk factors for carriers can at times be a routine part of life. However, the consensus for carrier status is that the main importance of this finding relates to indirect future reproductive risks (of having a child affected by SCD) and to direct benefits of protection from the risks of malaria. Analysis of the value of sharing carrier findings in this thesis is based on this consensus.

1.7.2 Recognising the influence of the availability of services

While agreeing on the central role of clinical importance in defining researchers' responsibilities to share study findings, commentators have also pointed to limitations in using a theoretical form of usefulness without certainty that the services available will deliver the expected benefits. For researchers in all settings, this 'availability' has been held to include their ability to interpret and explain the findings, to understand the nature of clinical importance and to provide services. This possible limitation seems particularly applicable where researchers are not clinically trained. Even where researchers have a medical background, the interpretation of the significance of some types of findings might require specialist knowledge and experience. Some emphasis is therefore also placed on the ability of researchers to refer participants with findings they believe might be clinically important to specialist genetic services (Ravitsky and Wilfond 2006). As Sharp and Foster (2006) point out, however, there is a dilemma in classifying findings as having clinical benefits when the services needed to achieve these are not available, including genetic counselling, given the sensitivity and sometimes the complexity of the information to be communicated (Sharp

[28] There is a recognised dilemma around responding appropriately to the possible health implications of SCT, since over-medicalisation is a risk and concerns have been raised about increased potential for stigmatisation of carriers, for example in employment and insurance. The USA military recently removed a requirement for new recruits to be screened for SCD, instead electing to advise all new recruits as if they might carry this trait, in order to act against a form of stigmatisation that might be seen as racially based. In Africa, SCT stigmatisation would not have the same potential for racial discrimination, but could carry other discriminatory implications that would have to be weighed against health and autonomy gains.

and Foster 2006). In other words, one critical feature of this debate relates to the resources available to support information sharing on findings in research, and therefore also to issues of resource prioritisation in and between research and health service provision.

The dilemma around *achieving* clinical benefits brings the situation surrounding SCD information sharing in research in developing countries into sharp focus. In Kilifi, services are available to validate tests and provide basic medical care and some counselling. As noted[29], these services are significantly supported by core research resources, albeit in collaboration with the district general hospital. Screening and providing services across the population involved in KGBC (260, 000 people, including around 8000[30] new births per year) includes the costs of reimbursing transport for a first visit to the SCD clinic, validating tests and providing counselling and medical care for children affected, including screening for siblings where requested.

Further, looking outside the set up in Kilifi, other research sites may not have these facilities or research core funds to support their implementation. In this situation, it is less clear what the responsibilities of researchers should be towards disclosure of findings on SCD. In this way, the debate on researchers' responsibilities for disclosing genetic findings is linked to a closely related set of questions about their responsibilities for providing the types of services that would make clinical usefulness achievable. This linked issue is a cornerstone in the debate about researchers' responsibilities for ancillary care, as will be described in section 1.8.

1.7.3 Providing a wider understanding of 'utility'

Putting aside issues of how clinical benefit could be achieved, there is now general agreement that the term 'clinical utility' should take account of the views of research participants, and sometimes the public in general, in deciding whether a particular type of finding is useful (Knoppers, 2006).

[29] See section 1.3.1 and 1.4

[30] Within this population, approximately 80 babies with the disorder and 1440 babies with carrier status are likely to be born each year.

As early as 1993, the Nuffield Council on Bioethics produced a report on genetic screening indicating that an important criterion in selecting screening programmes was the value of the knowledge gained *for those being screened* (NCOB 2006). Bioethicists have pointed out that principles of respect for persons and autonomy underpin the importance of understanding wider meanings of usefulness, including rights to know and not to know (Ravitsky, 2006). For these reasons, at an extreme, commentators have claimed that 'the investigator should be compelled to offer results based on the investigator's inability to obtain a complete conception of clinical utility' (Lavieri, 2006) (p48).

1.7.4 Understanding the role of parent-child relationships for participants who are minors

KGBC involved the screening of minors, with parents consenting for their children to be included in the research. Genetic testing in children in general is a topic of much ethical debate. Borry (2006) published two systematic reviews of international guidelines or position papers on presymptomatic or predictive genetic testing on children (Borry, Stultiens et al. 2006) and on carrier testing in minors (Borry, Fryns et al. 2006) that are helpful to the analysis in this thesis. For 27 publications on presymptomatic testing, Borry reports a consensus on the need to avoid genetic testing in children, unless this is seen to be of clear and compensating direct benefit to the child themselves, given the potential for harming a child's self-esteem, creating depression or anxiety, distorting the family perception of a child, stigmatising the child, breaching the child's confidentiality and privacy, ignoring the specific moral status of the child and downplaying respect for the autonomy of the child or adolescent, including a right not to know. In relation to carrier status, the guidelines reviewed by Borry (2006) agreed that children under 18 years should not be tested for carrier status, but that this should be deferred until children themselves were old enough

to make this decision (Borry, Fryns et al. 2006). Five guidelines provided for exceptional cases where a compelling family reason for testing a child existed[31].

The ethical principles drawn upon in recommending deferral of carrier testing concerned the lack of benefit for the child in testing before reproductive age; the breach of privacy, confidentiality and autonomy of the child that testing would incur; and the potential for testing to negatively impact the mental wellbeing of the child. In 2006, Borry noted that there was no empirical evidence to support concerns of psychological harm, but that this absence *per se* would not undermine the status quo of the ethical analysis presented (Borry, Fryns et al. 2006).

While the paragraphs above report on guidelines for decisions about the acceptability of genetic testing in children, including carriers, the advice surrounding disclosure of genetic carrier information generated *incidentally* during research is more complex. Amongst the 14 publications on carrier testing in minors, four sets of guidelines - from the UK, Europe and North America - dealt directly with this more difficult position[32]. The situations described in the guidelines were concerned with incidental carrier findings during research and newborn screening, and therefore seem analogous to that of screening healthy young children for SCD in KGBC. In addition, Miller and colleagues (Miller 2009; Miller, Hayeems et al. 2009; Miller, Robert et al. 2009; Miller, Paynter et al. 2010) have more recently written a series of papers on the subject of disclosing carrier findings from newborn screening that includes SCD carrier status based, in part, on research in a non western setting in India. Drawing on these two sources, the arguments for and against sharing *incidentally generated carrier information in children* in the literature can be summarised as follows:

- *Sharing information is seen as important to*: respect the child's and parents' rights to information, including knowledge of future genetic risks within the family; avoid the over

[31] Such as extreme anxiety on the part of parents.
[32] I note that in research in developing countries, SCD carrier status may not be classed as an incidental finding, since it is often included in study design as a risk factor.

paternalism that withholding information would seem to constitute; acknowledge a right to ownership of such information, seen by some as particularly important given the 'fundamental' nature of genetic information; and enable informed choices (for the parents and child) about reproduction in the future.

- *Withholding information is seen as important because of*: the risk of carrier information being misunderstood, leading to harms of over-medicalisation, and stigmatisation; and to the detection of misattributed paternity where parental testing follows. Further, sharing information through parents risks infringing the autonomy and confidentiality of the child, as well as the right of the parents and child not to know. The latter may be particularly important where the validity of informed consent processes is challenged. Little support is offered for arguments that information sharing would support parental decision making, since parents could be tested independent of their child; and the child's interests are regarded as central in the instance of testing a minor.

In summary, this literature presents a controversy around the question of whether or not carrier findings in children should be disclosed in research that generates such information, but some pragmatic - and questioned - consensus that carrier results should generally be returned, after checking through the informed consent process that this is what the parents would wish to happen. This position draws on the ethical principle of autonomy, and counters charges of medical over-paternalism.

1.8 Literature on researchers' positive responsibilities for providing services in developing countries

In the preceding section, I discussed the concept of 'utility' for disclosure, and its limitations. In the current section, I look at a separate but connected literature on the way that researchers' positive responsibilities for providing medical services have been seen, given the relationship between the

proposed benefits of genetic findings and the availability of services to ensure these are achieved (Sharp and Foster 2006; Beskow and Burke 2010). This relationship seems particularly important in developing countries where the medical services needed *may not be available through the public health sector*.

In a broad sense, the responsibilities and roles of researchers have traditionally been considered through a professional lens, stemming from the evolution of biomedical research as a branch of medical practice. The most widely used formulations of these responsibilities follow the principles of biomedical ethics outlined by Beauchamp and Childress (Beauchamp and Childress 2001), as seen in the guidelines for biomedical research involving human subjects issued by the World Health Association (World Medical Association 2004), the Council for International Organizations of Medical Sciences (CIOMS 2002) and the Kenya National Council for Science and Technology (NCST 2004). Within the bioethics literature, the concept of responsibilities for researchers being either positive or negative is linked to a distinction between ethical principles of beneficence and non maleficence. Beauchamp and Childress (2001) define beneficence as 'a group of norms for providing benefits and balancing benefits against harms and costs'; and non maleficence as 'a norm of avoiding the causation of harm' (Beauchamp and Childress 2001) (p12). Accordingly, researchers have negative obligations in relation to norms of non maleficence and positive ones in relation to those of beneficence; they must avoid causing harm and they should aim to maximise benefit as much as possible. As this implies, a further difference between a negative and a positive responsibility for researchers is that the former is automatically seen as an obligation, while the conditional nature of the latter means that a convincing justification would be needed to see any specific action as a positive responsibility.

More specifically, the positive responsibilities of researchers *in relation to providing services to participants outside that needed as part of study procedures* has been extensively debated as part of a literature on researchers' ancillary care responsibilities. This literature is particularly useful

because it encompasses a careful analysis of reasons why actions seen as important, such as sharing SCD information, might be a specific responsibility of researchers as opposed to other stakeholders. If a responsibility is seen as specific to researchers, this increases its strength.

Researchers' responsibilities to share SCD information, and provide the services needed to ensure this information is useful to participants, may therefore be importantly informed by theories within the literature on their responsibilities for ancillary care. Of particular relevance is that ethical dilemmas generated by research in developing countries has been the driving force for the debate on ancillary care, particularly around provision of care during research on HIV/AIDS (Richardson, 2008). There is an inevitability about this dilemma; the need for biomedical research to tackle global health inequities that affect developing countries and the limited provision of government health care frequently encountered in the same settings generates a potential responsibility for researchers to make up any shortfalls met through providing ancillary care.

A distinction between the 'obligatory' and 'ancillary' care that researchers might have to provide is given by Richardson and Belsky in two frequently cited articles published in 2004 (Belsky and Richardson 2004; Richardson and Belsky 2004). Ancillary care is defined and explained as 'that which is not required to make a study scientifically valid, to ensure a trial's safety, or to redress research injuries.' (Belsky and Richardson 2004) (p1494). In 2006, participants at a workshop at Georgetown University in the USA considered the ancillary care responsibilities of researchers in developing countries[33] (Georgetown University Workshop 2008). According to their report, drawing on guidance from the Nuffield Council of Bioethics[34] and the Joint United Nations

[33] The fifteen workshop participants at the Georgetown workshop were mainly drawn from academic institutions in the USA, but with representation from the UK, India, Malawi and Ethiopia as well as from the pharmaceutical industry.

[34] Nuffield Council on Bioethics (2002) 'The ethics of research related to healthcare in developing countries'. http://www.nuffieldbioethics.org/go/ourwork/developingcountries/publication_169.html; and 'The ethics of research related to healthcare in developing countries: A follow-up discussion paper'. http://www.nuffieldbioethics.org/go/ourwork/developingcountries/publication_169.html

Programme on HIV/AIDS[35], the participants concluded that 'the ancillary care obligations [of researchers in developing countries] are not nil' (p0710). The report went on to describe the moral basis for such a claim as: i) Due Concern for Welfare, drawn from the Declaration of Helsinki[36]; ii) A duty of rescue (discussed in section 1.8.2); iii) Justice; or some responsibility to take into account background injustices that impact on research in developing countries; and iv) Entrustment; or the extent to which 'research participants automatically entrust certain aspects of their health into the researchers' care' (p0711). These last two concepts will be outlined in the following paragraphs, including the way these relate to community perspectives.

1.8.1 Researchers' responsibilities to take account of background conditions of injustice

There are some strongly held and polar positions within the debate on the importance of researchers taking background injustices into account when defining their ancillary care responsibilities that are of obvious relevance for research in developing countries. One prominent position draws on philosophical accounts of wrongful exploitation to exclude background characteristics completely from this concept (Wertheimer 2008). According to this position, exploitation in research has a procedural component that relates to the validity of the informed consent process; and a substantive component that is dependent on the notion of 'fair exchange'. The latter is described as notoriously difficult to define but claimed as independent of the background context for the exchange (Wertheimer 2008).

The polar position is represented by the proposal of an *obligation* for researchers to address background global injustices, as for example argued by London (2005) in calling for a 'human development approach to international research ethics' (London 2005) and by Benatar (2002)

[35] Joint United Nations Programme on HIV/AIDS (2000) Ethical considerations in HIV preventive vaccine research. http://data.unaids.org/publications/IRC-pub01/JC072-ethicalCons_en.pdf.
[36] World Medical Association (2000) Declaration of Helsinki: Ethical principles for medical research involving human subjects. Revision adopted by the 52nd World Medical Assembly. http://www.wma.net/e/ethicsunit/helsinki.htm.

similarly proposing a role for research ethics in 'promoting moral progress and improving global health' (Benatar 2002). Ijsselmuiden and colleagues (2006) anticipate that 'the next 'era' in international research ethics will involve an intensification of focus on the role of research in achieving global justice' including researchers' responsibilities for ancillary care (Ijsselmuiden, Kass et al. 2010) (p1). Arguments moderating a 'research as development' approach were identified by the participants at the Georgetown workshop as: the challenge in seeing why this is a specific responsibility for researchers rather than other citizens; the impact that the extra demand for resources would have on the viability of the research enterprise; the risk that ancillary care benefits would act as undue inducement and undermine voluntariness; and the risk of undermining host governments through the provision of such services (Georgetown University Workshop 2008).

On this basis, the consensus of the workshop was that an intermediate position should be taken, to acknowledge that researchers should take account of background injustices but that such responsibilities should be determined on a case by case basis linked to other considerations related to the scope of the research and the strength of such a requirement, as described in more detail in section 1.8.3. Importantly, responsibilities were seen as existing for funders and sponsors of research as well individual researchers. Separately but similarly, Lavery (2010) supports an intermediate position, described as a 'relief of oppression' principle (Lavery, Bandewar et al. 2010). Lavery's central point for this debate is that a 'fair benefits' model could and should include benefits that aim to address background injustices as far as possible.

1.8.2 Researchers' non-professional responsibilities for ancillary care

Participants in the Georgetown workshop derived responsibilities for researchers to provide ancillary care from two sources: general moral norms that would apply to anyone confronted with a person in need of help, and those associated with their profession. The former were described as a 'duty of rescue'. Some commentators have linked the idea of a responsibility to help others to the

Kantian maxim that people should not be treated merely as a means to an end, but also as an end in themselves, implying a fundamental respect owed to all people that could generate a requirement to help in this situation (Richardson 2008). Other authors liken this responsibility to that of a 'Good Samaritan'; not helping someone in distress when this is reasonably easily within reach, by the Good Samaritan account, would demonstrate a profound disrespect for that person (Beskow and Burke 2010).

An obligation to assist has perhaps been most clearly spelled out by Singer (2011), using the analogy of the moral responsibilities most people would see for a passer-by to rescue a child drowning in a shallow pond[37] (Singer 2011). The obligation to assist rests on an agreed moral pre-eminence of preventing harm, assuming no equally important moral costs are implied by the action of assisting, and strengthened where there is no one else in a position to help. These analyses of a general responsibility to assist others might be seen as linked to the types of good citizen responsibilities described in the previous section to address existing background injustices as far as possible. Although there is a challenge in assessing how much assistance would be due, and what costs and competing obligations might reasonably limit an obligation, there is wide agreement in the literature as to the value of the underlying principle.

1.8.3 Researchers' responsibilities for ancillary care from 'a partial entrustment' model

The entrustment model discussed at the Georgetown workshop derives from original papers by Richardson and Belsky on researchers' ancillary care responsibilities, referenced at the start of this section. The authors described a 'partial entrustment model' as a rationale for this responsibility, based on the nature of the professional relationship between researchers and research participants (Belsky and Richardson 2004; Richardson and Belsky 2004).

[37] This principle was articulated to account for an obligation for those living in the rich parts of the world to support those living in poverty, but has been more generally interpreted.

Although these earlier publications drew attention in more detail to the features of researcher-participant relationships that would count towards understanding the strength of a researcher's responsibility to provide ancillary care, later publications on this model place focus primarily on three factors; scope, engagement (including features of the researcher-participant relationship that relate to benefit) and cost, subsuming other features under these three main considerations (Richardson 2007). Overall, the steps for researchers implied by the model would be to consider whether the ancillary care needed was within the scope of the original research design, the strength of the obligation as dictated by the degree of engagement with participants (including needs) and the cost of meeting it. An important challenge to this partial entrustment model is presented by Dickert's (2007) subsequent observation, based on experience of research in developing countries, that the strength of an obligation should not only be considered for ancillary care needs within the scope of the research since, for example, an obligation to meet an easily satisfied ancillary care requirement might also validly operate outside this domain (Dickert, DeRiemer et al. 2007).

In earlier more detailed descriptions of the rationale for the partial entrustment model, Richardson and Belsky (2004) seek to establish an ethical basis by which researchers might have positive ancillary care obligations (Richardson and Belsky 2004). Their approach starts from the premise that research-participant relationships are characterised by particular types of power imbalances directly related to the level of trust placed by participants in researchers (entrustment). In a situation of power imbalance, two aspects of the agreement strengthen researchers' responsibilities to a positive level. The first is the participant's agreement to allow the researcher special privileges - specifically those linked to privacy - that would not be allowable outside this professional engagement. The second is the researcher's tacit acceptance of this responsibility through the process of professional engagement[38] (Richardson 2008). As a result, the nature of entrustment within the researcher-participant relationship leads to moral responsibilities being intrinsically

[38] In contrast with, for example, any responsibility the researcher would have towards another individual if stopped in the street and offered private information that s/he had not solicited.

generated for researchers. As the name 'partial entrustment model' implies, other types of relationships can have more or less entrustment. For example, complete forms occur in fiduciary relationships such as guardianship; lesser forms - that are still greater than for researcher-participants relations - occur in physician-patient relations; and sales agreements illustrate a low form of entrustment. Across these types of relationships, the diminishing level of entrustment reflects diminishing imbalances of power, and consequent reductions in the responsibility owed by one person to the other.

In the model, two broad features of the context then describe the extent to which researchers can be seen to have positive responsibilities in relation to ancillary care; whether the findings fall within the scope of the agreement underlying entrustment, and the strength of the obligation implied by the relationship. According to Richardson (2008), the strength of obligation depends on the following features of that relationship: "(1) how much difference getting care would make to the welfare of the individual ("vulnerability"); (2) how dependent such an individual is on the research team for getting that care ("dependence"); (3) how intense and long-lived the relationship expectably is between researcher and participant ("engagement"); and (4) whether the researchers owe the participants any debt of gratitude for their willingness to undergo procedures that may be risky, painful, or inconvenient ("gratitude")." (Richardson 2008) (p11). The final element to this model is cost, seen as a morally relevant issue given the social importance of research. There is concordance with the cost issues raised in the literature on the disclosure of genetic findings in research (section 1.7), seen there as a balance between the 'public good' nature of research and individual participants' autonomy and interests. Taken together, these features of scope, the nature of the relationship between researchers and participants and the costs of providing ancillary care provide a useful starting point for considering the responsibilities researchers in Kilifi, and other similar settings, may have to share information on SCD during studies, and provide services to ensure usefulness of this knowledge.

1.9 Summary and overview of the thesis

In this first chapter, I have described a specific dilemma arising during a genomics study at a biomedical research centre in Kenya on sharing SCD findings with the parents of participant children. Early experience of SCD information sharing led to uncertainty about the extent to which anticipated benefits could be achieved, and about how the process could be strengthened. Awareness of this dilemma, and recognition of the need to take community members' perspectives into account in finding a solution, has generated the broad objective of this thesis to develop appropriate responses to important ethical questions about disclosure of SCD findings during international collaborative biomedical research, including the responsibilities of researchers in this regard[39]. The importance of these questions is highlighted by the fact that many different types of biomedical research in settings where malaria is endemic may include screening and analysis of SCD status; and limitations in services provided for this disorder in the public health sector.

The study methods, including research over two separate phases, aim to draw strongly on community members' experiences and informed deliberation within a process of normative analysis. These methods will be described in Chapter 2, including their theoretical basis as a form of empirical ethics.

Chapter 3 largely presents data from research in the first phase (Phase One), concluding by identifying three ethically important questions around SCD information sharing in research. In Chapters 4, 5 and 6, respectively, I describe the outcomes of community deliberations in the second phase (Phase Two) around these three key questions. Within Chapters 4 to 6, I describe the role of three important emerging values of: protection of vulnerable children and (relatedly) family stability; openness as a form of empowerment; and researchers' responsibilities. I also explore

[39] See Chapter 1, section 1.5 for the specific objectives of the thesis.

difference and agreement amongst community members, and whether normative conclusions can be drawn from these discussions.

In Chapter 7, I turn to a different perspective[40] in focusing more on *my reasoning* around the conclusions from community deliberations in the previous three chapters. Drawing on my earlier analysis, the chapter frames my reflections on researchers' responsibilities for sharing SCD findings during KGBC, for affected children and those found to be carriers. As part of developing these arguments, I take account of a series of issues: important values emerging from discussions; the way perceived benefits could be seen to generate responsibilities; the potential for conflation between the roles of researchers and health providers; and challenges to achieving benefits from communication issues, perverse outcomes and resource prioritisation. This process includes resolving divergence within community debates by drawing on a wider literature, including that in Chapters 1 and 2.

In the concluding chapter, Chapter 8, I provide a summary of the thesis findings and discuss three areas of wider implications: key considerations for planning different types of studies that generate SCD findings, including in other settings; the lessons learned from the empirical approaches to normative analysis undertaken in this study, particularly for community consultation; and recommendations for future research. The overall conclusions of the thesis are therefore informed by those in Chapters 7 and 8, taken as a whole.

[40] Although I recognise this 'change' in perspective is more an adjustment, since the conclusions in Chapters 3 to 6 are also influenced by my perspectives, as described in the methods in Chapter 2.

CHAPTER 2

Methodological Approaches

2.1 Introduction and organisation of chapter

In Chapter 1, I described the overall aims of this thesis in exploring a research question with its origins in a dilemma experienced around disclosure of sickle cell disease (SCD) findings during a genomic epidemiological study at an international collaborative research centre in Kilifi. I explained that the thesis would address this dilemma through an empirical approach, with the intention of ensuring that a wide range of perspectives of the value of disclosing SCD findings was taken into account (Knoppers, Joly et al. 2006). Drawing on the literature on the disclosure of genetic findings in research and researchers' responsibilities for ancillary care, I concluded that the empirical process needed to support this analysis should provide a rich description of the context for the dilemma and take account of the reasoning and values of those directly affected. I also described that a further aim of the thesis is to draw upon and contribute to debates on empirical approaches to normative analysis.

Taken together, these aims made it clear that the overall methodological approach in this study would need to draw on the disciplines of both social science and bioethics. The focus of this chapter is to describe the methodological approach to collecting and analysing data in the thesis. Recognising a strong current of debate in the literature on empirical ethics on whether and how social science and bioethics might be considered compatible fields of study, I describe the way I have responded to this challenge. The underlying model for ethical reflection draws upon acknowledgement of the complexity of human experiences and values (Fulford, Dickenson et al. 2002). This seems particularly important in international collaborative biomedical research, given

partnerships between researchers from countries with different social, cultural, economic and political environments.

In the following sections, I begin by describing the overall design of the study (section 2.2); followed by the methods used for data collection, management and analysis for each of two phases of research separately (sections 2.3 and 2.4). In describing the analytic processes for the second phase of research, I describe the way that community members' reasoning on appropriate responses to dilemmas around SCD information sharing in research was incorporated into normative reflection for the thesis overall, including the way that differences and agreement were analysed, and the normative approach taken to framing responsibilities for researchers in the light of this analysis. In section 2.5, I discuss limitations to the methodological approach. The process of external scientific and ethical review of the proposal describing this work will be described in section 2.6. The conclusions of this chapter form the final section (2.7).

2.2 Overall study design

The empirical research for this thesis was qualitative and conducted over two separate phases with different but complementary aims. The findings from Phase One formed the basis of discussions with community members in Phase Two. During the analysis of data, there has been some iteration between phases to support reflexivity and trustworthiness (Patton 1999; Pope and Mays 2006). A description of the study area for both phases is given in Chapter 1 (section 1.3.2).

Phase One, conducted in early 2009, aimed to explore community perceptions of SCD and the Kilifi Genetics Birth Cohort study (KGBC), including SCD screening and information sharing in that study. The underlying purpose of this exploration was to identify direct or indirect moral concerns generated by participation in the study. Phase One was descriptive, and included the gradual development of themes for focused enquiry around a largely narrative approach of listening

to families' experiences, and progressive identification of participants, as a form of modified grounded theory (Flick 2006).

Phase Two, conducted in late 2009/2010, aimed to take forwards key moral concerns from Phase One into a process of normative reflection by community members in Kilifi. As will be shown in Chapter 3, the questions that emerged from Phase One reflected those already raised by researchers on sharing findings on SCD status. In addition to questions on whether researchers should share findings on SCD for *affected children* and for *carriers*, a concern arose over the potential for sharing genetic information on SCD to raise suspicions of misattributed paternity, and requests for paternal testing. A third question therefore concerned the nature of researchers' responsibilities in this situation, and whether researchers should share *findings from paternal testing*, given the potential to show misattributed paternity (where fathers were found not to be carriers). Deliberations in Phase Two aimed to explore community members' views about these three questions, through a process of informed reasoning that might reasonably be taken to account for important features of the range of opinions within the community as a whole. While challenging, the approach taken to this phase combined two broad elements, working with a form of methodological hybridisation described elsewhere as 'the pragmatic use of methodological principles and avoidance of a restricting subscription of a specific methodological discourse' (Flick 2006) (p25). The first element is familiar to many methods in qualitative research, reflecting aims to show diversity through purposive sampling; build rich descriptions to enhance understanding, including the potential for transferability of emerging theories; and to seek some level of redundancy through repeated enquiry (Flick 2006). The second element was more novel, based on a deliberative approach including information sharing activities and systematic probing to support and explore processes and outcomes of informed reasoning. A similar methodological hybridization was undertaken by Ives and colleagues in exploring concepts of fatherhood with

groups of men in the UK, described there as a cross between a focus group discussion and a philosophy seminar (Ives, Draper et al. 2008).

Where the aim of normative analysis is to address an issue in real life, descriptive understandings around a particular moral concern clearly form the basis of reflection (Ives and Draper 2009). To this extent, the role of social science in providing rich descriptions of context around a moral issue - as occurs in Phase One - is uncontroversial. The relationship between social science and ethics becomes more contested in identifying *the analytic process* by which a solution can be identified (Hope 2008; de Vries and Gordijn 2009; Leget, Borry et al. 2009). In the bioethics literature, these different aspects of a moral problem (understanding versus resolving) are often distinguished by reference to what 'is' the case, as seen for example through a rich description, and what 'ought' to occur, as identified through ethical analysis (de Vries and Gordijn 2009). Phase Two is therefore more controversial in its normative aims. Fundamentally, an argument could be made about the way I will define the *appropriateness* of solutions proposed through community deliberations, and against what ethical benchmark such a claim might be made. If the definition of 'appropriate' depends on the views of community members in Kilifi, for example, what would that mean if the emerging values conflicted with the views of people in other situations, or of those widely held as 'objective', such as respect for all persons? (Sheehan 2007) And how, in any case, can differences or conflicts in values *within* a community be taken into account in defining a measure as appropriate?

For these reasons, *the final stage of analysis* in the thesis is based upon my normative reflection on the findings in both phases in relation to the three main questions posed. This analysis can be seen as a form of reflective equilibrium[41] (Beauchamp and Childress 2001) that is strongly led by the views and reasoning of community representatives, community leaders, staff groups and affected families in earlier phases, but represents my account of the way that these can be brought together

[41] Also see Chapter 1, section 2.4.1.1

around specific questions for the thesis, drawing on the literature and ethical theory more widely. As I will go on to discuss, one implication of this approach is that further rounds of deliberation on the thesis conclusions, involving a wider group of stakeholders, would be important to take the thesis conclusions forwards into policy.

Throughout this study, reflexivity has been important in taking account of the influences I, and other staff who supported me, have had on data collection and analysis, and vice versa. All the facilitators in this study (including myself) are long term residents in Kilifi, who were either born in this area or - in my case - have lived in the district for more than 20 years. My professional background is in medicine and health systems research, with a focus on community-based interventions and engagement in research over the past 15 years. Our positions working within a large and powerful biomedical research programme in the district, seen both as an important source of medical care and employment, may have influenced the information community members decided to share with us, as well as the way we interpreted 'findings'. I and the main facilitator (Francis Kombe) who assisted with Phase Two data collection were able to build upon established relationships with community leaders and many community representatives, as part of the group normally responsible for community engagement in research within the programme. Debriefings after interviews, taking several viewpoints into account in data coding and categorisation during analysis, and sharing of drafts during the writing of thesis chapters with facilitators have aimed to strengthen the trustworthiness of these processes, described in more detail in the following sections.

2.3 Study Methods in Phase One

2.3.1 Study participants and sampling in Phase One

In total, 34 community members participated in this phase of study, including five families affected by SCD, 17 families who participated in KGBC, seven field workers responsible for recruitment

into the study and five community facilitators who had undertaken community outreach activities for KGBC. In keeping with the exploratory nature of this phase of study, the identification of participants for interviews was partly *a priori*, but was also based on identifying emergent issues over the phase of study, as will be described in the following paragraphs. Tables 2.1 and 2.2 at the end of section 2.3.1.2 provide a detailed description of participants in Phase One of this study.

2.3.1.1 Initial sampling and study participants in Phase One

Early in 2009, using SCD clinic records at the district hospital and information from the research centre's KHDSS[42], the first interviews were conducted with *five families with one or more child affected by SCD but not participating in KGBC,* for two main reasons. One was to begin the process of gathering data on beliefs and perceptions about the disorder to support understanding of the implications of SCD screening in KGBC in the community. A second, underpinning the timing of these interviews, was to gather initial data on the disorder that would support planning for later interviews, including ways in which SCD was locally recognised, and the language used to talk about it, as well as who should be interviewed to support understanding of emerging concerns.

Although the number of families in this first group was small, purposive sampling drew on criteria seen as likely to underpin diversity in this community. Families were selected from rural and urban parts of the community, with different faith backgrounds, frequency of clinic attendance and numbers of affected children. The influence of an urban/rural distinction was described by community facilitators as likely be important, as also seen in KHDSS data, in relation to wage employment in town, levels of formal education and formal religion. Kilifi town has a higher proportion of Muslims than in more rural areas of the district. Areas taken as 'urban' were those within Kilifi Township as well as some focal areas of building and service development within the surrounding largely rural locations. Organised faith was seen as a likely influence on diversity; for

[42] Kilifi Health and Demographic Surveillance System, see Chapter 1 section 1.3.1

example, many Christian churches locally strongly discourage traditional supernatural beliefs and use of traditional medicine. It was seen as important to capture the perspectives underlying different choices about clinic use; and having more than one affected child was thought likely to lead to both different interpretations of the disorder as well as different burdens to the family. The first family visited was closely related to one of the field workers at the research programme who participated in this study; in so doing, we hoped to minimise the sensitivity of the first interview through building on this existing relationship. All five families were visited twice, a week apart; both parents were interviewed unless the father worked or lived away from the home visited. Each visit lasted between one and three hours.

Following the interviews described above, *eight families who had participated in KGBC and whose child had been found to have SCD following screening* were interviewed. We continued to use a similar purposive approach to that taken for the first five families, that is, sampling for differences in location (urban/rural), formal faith and use of clinic since screening. Where information was available from the clinic, we also included the criterion of the number of affected children. As will be shown in Chapter 3, the stories of participation for these families were heavily dominated by the experience of learning about SCD in their child. Five children had in fact been tested diagnostically prior to their inclusion in KGBC; only three were predictively screened as part of the study. Given this gap in understanding the way KGBC would be seen in the absence of a positive SCD test, we therefore identified and interviewed a further *nine families who had participated in KGBC and been given a result of 'not SCD'*, using criteria similar to those described for the earlier sampling.

2.3.1.2 Later sampling and participants in Phase One

Later in 2009, following an initial analysis of the data from families, twelve full time members of staff from the research centre, who were also local residents, were interviewed in groups or individually on community perceptions of SCD, its inheritance and impacts on family relations,

drawing on emerging themes from family interviews. Seven staff were field workers with the genomics study responsible for explaining the study and informed consent processes; two also had coordinating roles in the weekly SCD clinic at the District Hospital. Five staff were community facilitators undertaking community engagement activities across a range of studies, including the genomics study. Both groups were chosen on the basis of their particular experience with KGBC and relationship to the community. These interviews were seen as important since the perceptions of participant families and field workers about KGBC and SCD screening could be considered together as a form of triangulation ((Pope and Mays 2006). In addition, field workers were seen as able to report on perceptions related to a much larger number of families, having visited many thousands of homes by this stage to talk about the genomics study.

Table 2.1 Kilifi Genetic Birth Cohort field workers in Phases One and Two

Phase One ID	*Phase Two ID*	*Gender*[1]	*Age (years)*	*Education (years)*	*Religion*[2]	*Roles*
FW1	IDI03/P1	M	34	12	M	Field worker/SCD clinic coordinator
FW2	IDI03/P2	M	34	12	C	Field worker/SCD clinic coordinator
FW3	IDI04/P1	M	27	12	C	Field worker
FW4	IDI04/P2	M	34	12	C	Field worker
FW5	IDI06/P1	M	30	12	M	Field worker
FW6	IDI06/P2	M	31	12	C	Field worker
FW7	IDI06/P3	M	33	12	C	Field worker

1. Gender: M=Male; F=Female
2. Religion: C= Christian; M=Muslim; T=Traditional; N=None given

Table 2.2 Phase One participants: Families

ID	*Child's age (years/months)*	*Child M/F*	*Mother's age (years)*	*Mother's education (years)*	*Number siblings with SCD*	*Religion*	*Urban/ Periurban/ Rural*	*Symptoms before screening*	*Reported SCD clinic use*	*KGBC status*
1	4y 9m	F	30	6	1	C	PU	Y	regular	Non study
2	3y 9m	F	28	6	1	C	R	Y	regular	Non study
2b[1]	14y	M	50	6	0	C	R	Y	regular	Non study
3	3y	M	28	12	0	C	R	Y	regular	Non study
4	5y 7m	M	?50	0	2	C	R	Y	regular	Non study
5	5y 6m	F	31	0	0	T	R	Y	occasional	Non study
6	1y 2m	F	24	6	1	C	U	Y	regular	Study; SCD pos
7	2y 10m	F	30	6	0	C	PU	N	regular	Study; SCD pos
8	2y 9m	M	30	12	0	N	PU	Y	regular	Study; SCD pos
9	1y 11m	M	DK	0	2	M	R	N	occasional	Study; SCD pos
10	2y (twins)	M	?25	0	0	C	R	N	occasional	Study; SCD pos
11	2y 5m	F	DK	0	1	C	R	M	regular	Study; SCD pos
12	1y 3m	F	39	DK	DK	C	U	N	none	Study; SCD pos
13	1y 8m	M	20	0	0	M	R	Y	none	Study; SCD pos
14	12m	F	19	0	NA	N	U	NA	NA	Study; not SCD
15	13m	F	23	6	NA	M	U	NA	NA	Study; not SCD
16	11m	M	19	0	NA	M	PU	NA	NA	Study; not SCD
17	17m	M	30s	0	NA	C	R	NA	NA	Study; not SCD
18	11m	M	22	6	NA	C	R	NA	NA	Study; not SCD
19	13m	M	35	4	NA	N	U	NA	NA	Study; not SCD
20	11m	M	19	6	NA	N	PU	NA	NA	Study; not SCD
21	13m	M	22	4	NA	N	R	NA	NA	Study; not SCD
22	1y 5m	F	27	0	NA	C	U	NA	NA	Study; not SCD

1. 2b was teenage aunt to ID 2; her mother's story was also included in the overall analysis

2.3.2 Data collection and management in Phase One

For all *family interviews* in Phase One, discussions took place in the family home, and involved both parents (except in a few instances when the father worked away from home and could not be met) and often other members of the family who were at home at the time of the visit. In this traditionally patrilineal[43] cultural setting (Shaw 2006), many participants live in extended family groupings centred on the home of the household head, a senior member of the paternal clan. Several 'nuclear' families (parents and children) live in this common wide household, although with separate housing structures. While other family members were welcomed to join in the discussions at times, particularly where a family history could be drawn upon in this way, care was taken to ensure that any sensitive information was discussed in privacy, particularly for the mother. For families with an affected child, interviews were conducted over two separate visits to the home, usually a week apart, each time lasting between two and three hours. The interviews encouraged narrative accounts of experiences with the genomics study, SCD screening and the disorder itself over time, including perceptions of cause, practices, effects and patterns within the wider family.

The family interviews were largely held in Kigiriama, the local mother tongue, with use of some Kiswahili, the national language. The main interviewer, one of four field workers who supported this first phase of data collection, used an interview guide with a detailed probing structure (see Annex A1 to A4: Phase One Interview Guides). The tool was developed over the course of these interviews in response to emerging issues as well as being informed by *a priori* themes for enquiry. It was seen as essential that the final version included a structured reminder about fixed probes, rather than allowing for more flexibility as might have been preferred in an exploratory qualitative study, because I was unable to conduct the interviews in the Kigiriama language needed to facilitate open discussion for this group of participants. All four interviewers shared the language and culture of interviewees, and had experience of qualitative data collection, but their understanding of the theoretical background to the study was clearly less than mine. An alternative

[43] A patrilineal culture is one in which social and political identity and ancestry are constructed in the male line (Shaw, 2006).

approach would have been to work through a strict 'real time' translation process, but at the risk of interfering with the quality of the data. This situation is not an uncommon dilemma in qualitative research, where a choice has to be made between interviewers having cultural and language skills in common with interviewees, or having a strong theoretical perspective on a study necessary for flexible inductive processes in qualitative research (Molyneux, Goudge et al. 2009). The ideal situation would be that both sets of skills could be found in the same individual. My approach was to build the interviewers' understanding of the background to the study as far as possible through a series of group discussions, which included joint development of the interview guide, role play interviews and repeated amendments before the first family was approached. Debriefing after the interviews enabled us to discuss areas in which more probing would have been informative; and for many families follow up of issues could occur in the second interview. In addition, in all interviews I was accompanied by a second field worker whose main role was to help me to follow conversations in Kigiriama through verbal hints and sharing written notes. I have sufficient familiarity with Kiswahili to follow conversations and support probing where this language was used. For staff members, I was able to conduct the interviews in English.

Interviews were digitally recorded, and immediately after each interview, the interviewer either handwrote or typed up a translation in English - where indicated - from the recording. The interviewers and I discussed English translations, including referring back to the voice recordings where issues were unclear. The handwritten versions were typed up in Microsoft Word by data entry clerks, before being checked for accuracy by the field worker responsible for the original version, and by me for coherence. Any unclear issues from the first interview were clarified with the family at the second visit.

2.3.3 Data analysis in Phase One

Data from Phase One included approximately 28.5 hours of recordings of family narratives and 6.5 hours of discussions with staff in the programme. Analysis was based on an initial immersion in the data through repeated reading of all transcripts, while making notes about the background, overall content and key issues by interview, including noting unexpected findings and possible patterns and themes. Five transcripts that were particularly rich in content were then identified for line by line coding using the constant comparative method to support progressive categorization (Flick 2006). An initial coding scheme was developed in this way by two researchers (myself and my local supervisor) working independently. After comparison of these preliminary coding plans, I continued to code the remainder of the transcripts in the same way, adapting the initial framework as new data dictated. Other supervisors also independently read and commented on four transcripts from this phase to support validation of codes and themes. Data were managed with Nvivo 8 software to facilitate discussion and audit. Following coding and the development of themes, analysis charts were drawn up to identify the contributions of individual participant within groups of codes, and support the comparison of these findings across the interviews. The final coding framework and an example of an analysis chart for one group of codes from the family interviews are included in Appendix B (B1 and B2).

2.4 Study Methods in Phase Two

2.4.1 Theoretical considerations for Phase Two

As described in the overview of this chapter, the aims of Phase Two were different to those in Phase One, being to explore processes and outcomes of normative reasoning amongst community members in Kilifi on ways of resolving moral concerns. I described that the move from Phase One to Two represents a shift from a less to a more contentious approach to empirical forms of ethical analysis. A first area of contention for Phase Two is the introduction of a theoretical risk of moral relativity, given its nature as a form of non expert *in situ* ethical reasoning. The risk, in the absence of external ethical benchmarks, is that *any view that is widely enough held* might have to be seen as morally acceptable. A linked set of challenges for Phase Two are captured in critiques of

deliberative ethics, given the close relationship with the deliberative methods planned in Kilifi. Approaches in the thesis to both sets of challenges are discussed in the following paragraphs.

2.4.1.1 Using 'non expert' reasoning in normative analysis

In the introduction to this chapter, I described a fundamental challenge seen by some commentators in basing normative conclusions on empirical processes of reasoning - as aimed for in this thesis - given theoretical concerns about risks associated with moral relativism. For example, Fulford and colleagues - although defending complexity - describe the philosophical concern that: 'if a substantive system of ethical norms is replaced with diversity of values, ethical process will be left rudderless…and risks ethical meltdown.' (Fulford, Dickenson et al. 2002) (p16). In contrast, social scientists tend to see morality in general as a social construction, such that there is less logic in appealing to a putative transcendental meaning, whereas there are many reasons to value wider forms of human reasoning and to see a fundamental challenge in any approach that does not recognise the formative influence of context (Hedgecoe 2007; Hoffmaster and Hooker 2009). This sociological position has led to the discipline of moral philosophy itself being described as in need of greater reflexivity (Ives and Dunn 2010).

A sociological critique seems particularly relevant for global forms of health research that involve partnerships between researchers from different countries, where differences challenge, at least in principle, the notion that understood values and methods of reasoning developed through a history of scholarly endeavour in one setting will have similar moral traction in another. For example, Hoffmaster (2009) argues against the exclusive use of a rationality based on the 'formal rules' of moral philosophers, instead advocating a prescriptive role for empirical understandings that include constructivist and naturalist accounts (Hoffmaster and Hooker 2009). These concerns are further politicised by awareness of the way that existing economic inequities between global partners risk dominance of one view, at the cost of understanding and allowing sufficient respect for difference. By illustration, Tangwa (1999) asks the question 'How is 'global bioethics' to be possible in a world inhabited by different cultural groups whose material situation, powers, ideas, experiences

and attitudes differ rather markedly and who are not, in any case, equally represented in globalization efforts and fora?'(Tangwa 1999) (p218)

Concurrent with controversy, there have been calls to understand, value and build on epistemological differences between social scientists and bioethicists (Hedgecoe 2004; de Vries, Turner et al. 2006) and to debunk some of the critiques seen as false through overgeneralization (Ives and Draper 2009). Part of this debunking includes pointing out that both disciplines in fact incorporate a variety of academic positions relevant to the debate, rather than presenting monolithic stances. Both disciplines have contributed to a literature on the exact way that empirical and normative domains could and/or should be brought together (Molewijk, Stiggelbout et al. 2004; Widdershoven 2007; Parker 2007a; Ives, Draper et al. 2008; Carter 2009; Dunn and Ives 2009; Hoffmaster and Hooker 2009; Ives and Draper 2009; Leget, Borry et al. 2009), including a series of publications on different typologies in empirical ethics (Hope 2008; Kon 2009; Parker 2009; Sugarman, Kass et al. 2009). The normative analysis in this thesis, drawing on processes of description, community 'deliberation' and reflective equilibrium (as will be described in more detail later in this section) can be seen as theoretically aligned with the group of empirical approaches that endeavour to bring social realities to bear on ethical reflection, including pragmatic bioethics, narrative ethics, ethics of care, communitarianism and hermeneutic ethics.

Hermeneutic approaches to ethics seem particularly relevant to the wider set of questions to which this thesis aims to contribute, that is, on policy around SCD screening in health research. Hermeneutic ethics aims to make everyday processes of interpretation around ethical questions explicit through dialogue between various stakeholders, with an ultimate goal or philosophy based on a Gadameric concept of 'fusing horizons'(Molewijk, Stiggelbout et al. 2004; Widdershoven 2007; Molewijk, Abma et al. 2008; Abma, Molewijk et al. 2009; Widdershoven, Abma et al. 2009; Widdershoven, Molewijk et al. 2009). Widdershoven (2009) describes the way that dialogue and facilitation in hermeneutic ethics are underpinned by reference to formal ethical principles, theory and forms of reasoning. Normative analysis becomes a process drawn outside the traditional

domain of bioethicists, while the critical importance of bioethics expertise to ground local debates remains. As well as the involvement of a wide range of stakeholders, the process is likely to rely on strong interdisciplinary partnerships between bioethicists, social scientists and experienced facilitators.

Two additional positions in the literature seem to provide particularly important support for the approach I have adopted to bringing together empirical findings and normative conclusions in this thesis, and counter theoretical concerns about 'ethical meltdown' that might follow the use of non expert processes of ethical reasoning. Firstly, Fulford and colleagues (2002) reassuringly suggest that placing diversity of values 'uncompromisingly' at the centre of the ethical action (although in reference to health care) has 'little risk of ethical chaos', since human values themselves are already diverse in every aspect of human life - law, aesthetics, sport and medicine are the examples given - without bringing chaos. Description and evaluation, or fact and value, are seen as 'twin logical elements, woven together as warp and weft in the conceptual framework' of healthcare ethics (Fulford, Dickenson et al. 2002) (p17). Fulford and colleagues' *interweaving* emphasises a movement for normative analysis away from applying rules and towards processes, with communication having a *substantive* rather than an *executive* role. On this basis, Fulford and colleagues (2001) provide a useful description of ethical analysis as a process based on understanding 'principles reasoning not as generating ethical conclusions directly, but rather as providing a four-dimensional "ethical space" in which the relevant particulars of a given situation can be worked out.' (Fulford, Dickenson et al. 2002) (p9). The analysis in Chapter 7 can be seen as such a process of 'working out' in relation to a theoretical ethical space.

A second way of considering the theoretical underpinnings of ethical analysis in this study, seen most directly in Chapter 7, is as a form of reflective equilibrium, defined by Beauchamp and Childress (2001) as a 'reflective testing of our moral beliefs, moral principles, theoretical postulates and other relevant moral beliefs in order to make them as coherent as possible' (Beauchamp and Childress 2001) (p 398), drawing on Rawls' original description of this concept in 'A Theory of

Justice' (1999)[44] as well as other sources. In other words, the outputs of a reflective equilibrium primarily presented in Chapter 7 are grounded in the views of community members in Kilifi, understood from the methods used in Phases One and Two, and described in Chapters 3 to 6. Central theoretical postulates in this process are those described in literature (including the principlist[45] approach to normative analysis and areas described in Chapter 1) and those related to the findings of the study (such as important emerging values).

In this way, as a process of reflective equilibrium, the analytic approach does not set out to depend *entirely* on the reasoning and values of the people who participated in this study but to also draw on external ethical benchmarks in deciding on the most appropriate way of resolving dilemmas. As Ives (2008) describes for his analysis of data from the hybridised empirical ethics method referred to section 2.2: 'Of course, once we have this information we do not have to agree with it or accept it at face value. It is information - it is there to inform, not to govern... in using these opinions and preferences, in accessing the values that inform them, and in having 'encounters with experience', we can make our ethical and philosophical deliberations more applicable, more useful, and more real' (Ives 2008)(p5).

My own approach in this thesis has drawn more strongly on the perceptions and values emerging as important within discussions with people in Kilifi, perhaps better described as allowing this to *lead* if not to *govern* development of my ethical reasoning as a process of reflective equilibrium.

2.4.1.2 Using 'deliberative' processes in normative analysis

Areas in common between the community deliberations planned here and methods of deliberative ethics described in the literature have allowed me to build on critiques of the latter. In his account of deliberative bioethics, Parker (2007b) describes this as an approach where 'public discussion

[44] Rawls J. A (1999) Theory of Justice: Cambridge, MA: Harvard University Press, 1971; version 1999.
[45] Beauchamp and Childress (2001) Principles of Biomedical Ethics: Oxford, UK; Oxford University Press.

should be central to the identification and analysis of bioethical issues...based on the concept of an inclusive deliberative democracy'. Deliberative ethics is contrasted with other approaches to ethical analysis that work through 'other democratic methods, or applying deontological analysis or consequentialist calculation by experts *ex situ*' (Parker 2007b) (p185).

Fundamental principles for deliberative ethics *per se* include the involvement of free and equal citizens in dialogic processes that support transformation of preferences through reasoning on the basis of rationality and impartiality, without coercion or deception (Parker 2007b). Parker (2007b) describes two important dimensions along which approaches to deliberative bioethics may vary. The first is the extent to which decisions are reached through procedures (procedural), as opposed to placing some restrictions on the types of outcomes allowable (substantive). The second is whether or not consensus building is seen as essential (consensual), with fair ways of living with diversity as a contrast (pluralist).

Key issues I have taken into account in developing study methods are those most associated with approaches that are both procedural and consensual, including the risk that morally important principles may be 'sacrificed' to a majority opinion; the distorting influence of power, at individual and structural levels, including the relative disadvantage to those less able to argue rationally; and that some voices may not be included at all. Effective deliberative processes have also been argued as occurring in relatively homogeneous groups. Parker (2007b) describes that key methodological elements to countering these challenges are a focus on reflexivity, capturing marginalised or overlooked voices, focusing on difference (as opposed to consensus), aiming to develop 'thick descriptions' through the use of additional ethnographic methods, and adopting a self consciously critical approach to established, agreed and uncontested practices, attitudes and beliefs. The way in which these elements have been drawn upon will be described in the remaining sections on methodology for Phase Two, with the exception of the use of 'thick descriptions' which was outside the scope of this thesis. The iterative nature of the analysis between Phase One and Two,

however, do provide some support for a rich understanding of the issues experienced and discussed by a range of community members and families.

One specific way in which I have aimed to reflect and understand diversity within the empirical findings from discussions in Phase Two has been to draw upon a concept described by Ruth Macklin (1999) in her book 'Against Relativism: Cultural diversity and the search for ethical universals in medicine' (Macklin 1999). Macklin argues that differences in moral reasoning within or between communities or societies often arise in three different ways that are important to normative analysis, related to differences in perceptions about relevant facts, differences in moral principles or values that are drawn upon, and differences in the ways that such principles or values are weighed against each other. Although I have already described debate about the relationship between areas of 'fact' and value in section 2.4.1.1, including that overall I have interpreted these as interwoven rather than distinct, I have adopted and extended Macklin's account of a distinction *as an organising concept* in the analysis to aid mapping out and assessing both *agreement* and *difference* in the community views described. Within Chapters 4, 5 and 6, I have included figures showing the mapping of agreement and difference in 'facts' and 'values' across the data, as will be described in more detail in section 2.4.4 on data analysis.

2.4.2 Study participants and sampling for Phase Two

Overall 62 community members participated in individual and group discussions in Phase Two. These were 18 KEMRI community representatives (KCRs); 17 local administrative leaders (chiefs or assistant chiefs); four members of the District Health Management Team; 20 people working within the research programme; and three mothers with affected children. Detailed information on participants in Phase Two is given in Table 2.3 (also see Table 2.1 for field workers who participated in Phases One and Two).

Sampling in Phase Two was purposive and, to a greater extent than the earlier phase, drew on *a priori* principles. The main aim in selection was to reflect of diversity of opinion by including

different types of community members (community representatives, community leaders, mothers with affected children, health managers and research staff) and individuals with different personal characteristics (gender, age, religion). In addition, including people within groups who had some familiarity with each other was thought likely to encourage openness and greater depth of deliberation[46].

Of the types of community members selected, KEMRI community representatives (KCRs) were seen as an important given their identity as 'reasonably' typical community members[47], with an existing role in consultation on research-related issues (Kamuya 2007). KCRs' existing relationship with KEMRI staff, understanding of basic features of research and familiarity with each other seemed likely to support both openness and informed reasoning in the planned discussions. Community leaders were selected on the basis of their typical role as gatekeepers and opinion leaders within their areas of work. KEMRI staff who were also local residents were important given their perspectives on common concerns and attitudes in the community - their own as well as those of friends, relatives and neighbours - and of researchers. The staff groups chosen were largely field workers with KGBC (some of whom had participated in Phase One), community facilitators and the group of four field workers who had conducted the interviews with me in Phase One. I also interviewed an early career researcher and two data entry clerks, the latter providing an opportunity for piloting the interview guide whilst also contributing their insights. Amongst the staff members, as Table 2.3 shows, there were several people with direct experience of SCD in their families; these individuals were seen as particularly important to bring into the discussions.

The final group, of three affected mothers, was brought cautiously into this phase of the study. The views of people with direct experience of living with the disorder were important in these discussions, particularly in describing vulnerabilities that might otherwise be overlooked in this

[46] Against this view, established interpersonal dynamics could also influence contributions negatively, for example, where someone familiar is already established as 'more knowledgeable'. At the same time, interpersonal dynamics are a feature of all group discussions, whether or not the members know each other beforehand.

[47] This is the main criterion that community members are asked to consider in electing KCRs, although it is likely that KCRs are slightly more educated than the average person in their community (Kamuya, 2007)

minority group (Parker 2007b). As suggested, several community members from the groups described above had direct experience of SCD, but it seemed important to ensure that this perspective was explored in greater depth. At the same time, I had concerns about bringing affected mothers into a discussion of this nature. Given the need to build understanding of the biomedical nature and inheritance of SCD to inform reasoning, and the probing needed around moral concerns including conflict in families, it was possible that participation could add to the anxieties of some mothers of affected children by appearing to challenge their beliefs, or asking them to voice concerns that might stem from distressing personal experiences, particularly in the presence of other community members. For this reason, I chose to invite mothers who had participated in Phase One, whose understanding of SCD and family responses were therefore reasonably known to us, and who seemed likely on this basis to respond positively to the topics of discussion[48]. These mothers, as with other community members[49] who had direct experience of SCD, were offered the choice of being interviewed individually or as part of a group. All those offered a choice preferred to participate in a group discussion. A fourth mother from Phase One declined participation since her affected child had sadly died in the period between these phases of study.

Table 2.3 Community members participating in Phase Two[¥]

ID	*Age (years)*	*M/F*	*Education (years)*	*Religion*	*Role/comments*
ID01	28	F	12+	C	Data entry clerk
ID02	33	F	16+	C	Community facilitator
IDI05	27	F	17+	C	Early career researcher
IDI07/P1	26	F	12	C	Field worker; sister has 2 children with SCD
IDI07/P2	27	F	12	C	Field worker
IDI07/P3	36	M	12	C	Field worker

[48] In fact, this approximately three hour group discussion was clearly valued by the mothers for the support they offered to each other, as well as the opportunity to ask questions.
[49] Apart from two staff members who were interviewed individually and not asked if they would prefer to contribute in a group for logistical reasons.

IDI07/P4	31	M	12	C	Field worker
IDI08	36	F	12+	C	Data entry clerk; carrier, 2 brothers with SCD, one died; carrier child
IDI09/P1	51	M	12+	M	Community facilitator
IDI09/P2	35	M	12+	C	Community facilitator
IDI09/P3	43	M	12+	C	Community facilitator
IDI09/P4	37	M	12+	C	Community facilitator
IDI10	51	M	12	C	Field worker; 2 affected children, 2 children died SCD
IDI 11	37	M	16+	C	Health manager
IDI 12	48	M	16	C	Health manager
IDI 13	43	M	16+	C	Health manager
IDI 14	42	F	16	C	Health manager
Chiefs01/P1	39	F	12	C	Urban assistant chief
Chiefs01/P2	37	M	12	C	Urban assistant chief
Chiefs01/P3	38	F	12	C	Urban assistant chief
Chiefs01/P4	49	M	12	C	Urban assistant chief
Chiefs01/P5	45	M	12	M	Urban assistant chief
Chiefs01/P6	56	M	12	C	Urban assistant chief
Chiefs02/P1	41	M	12+	C	Rural assistant chief
Chiefs02/P2	46	M	10	C	Rural assistant chief
Chiefs02/P3	40	M	12	C	Rural assistant chief
Chiefs02/P4	53	M	12	C	Rural assistant chief
Chiefs02/P5	48	M	12	C	Rural assistant chief
Chiefs03/P1	52	M	12	C	Rural/urban chief
Chiefs03/P2	48	M	12	C	Rural chief
Chiefs03/P3	55	M	12	C	Rural chief
Chiefs03/P4	49	M	12	C	Rural chief
Chiefs03/P5	50	M	12	C	Urban chief
Chiefs03/P6	52	M	7	C	Rural chief
KCR01/P1	59	F	7	C	KCR urban; Vice chair
KCR01/P2	32	F	16	M	KCR urban member
KCR01/P3	24	F	8	C	KCR urban member
KCR01/P4	53	M	12	C	KCR urban member
KCR01/P5	65	M	3	M	KCR urban member
KCR01/P6	50	M	12	C	KCR urban; Chairperson
KCR02/P1	35	M	7	T	KCR rural; Chairperson
KCR02/P2	36	F	12	C	KCR rural: Secretary/child with SCD
KCR02/P3	66	F	8	C	KCR rural member
KCR02/P4	30	M	10	M	KCR rural member
KCR02/P5	36	F	6	C	KCR rural member
KCR02/P6	36	M	12	C	KCR rural; Vice secretary
KCR03/P1	32	F	7	C	KCR rural; Vice chairman
KCR03/P2	34	M	8	C	KCR rural; Vice secretary
KCR03/P3	57	F	7	C	KCR rural; Secretary
KCR03/P4	57	M	8	M	KCR rural member
KCR03/P5	24	F	8	C	KCR rural member
KCR03/P6	58	M	12	C	KCR rural; Chairperson
Mot/P1	24	F	6	C	2 affected children (phase 1 Family F06)
Mot/P2	27	F	6	C	2 affected children (phase 1 Family F02)
Mot/P3	28	F	12	C	1 affected child (phase 1 Family F03)

[¥]Also see table2.1 for field workers in Phases One and Two

2.4.3 Information sharing activities in Phase Two

As described in section 2.4.1, an important element to data collection was the establishment of an informed basis for reasoning, that is, that this should be based on an understanding of central elements of the dilemma around disclosure of findings on SCD status in research. These elements included:

- The biomedical nature of the condition, including its prevalence, health implications and forms of management;
- Inheritance of SCD, particularly the means of transmissions across generations through healthy carriers, the risks of children being affected where both parents are carriers and the implication for affected children that both parents have the same role in transmission (as carriers).

The methods used to share this information varied between groups, depending on their pre-existing understanding of these topics. For example, field workers and community facilitators were already largely aware of these issues from previous training in preparation for informed consent and community engagement processes in KGBC, whereas community representatives and leaders had much less awareness of these issues, including the mothers of affected children.

In all cases, some form of information sharing - based on exploring current knowledge and perceptions - took place prior to the main discussion around the dilemma posed for SCD disclosure, often supplemented during discussions when clarifications were needed during reasoning. These information sharing activities were most formalised for community leaders and KCRs, reflecting the generally lower levels of awareness of the issues identified as relevant in these groups. For community leaders, the main 'deliberative' activity was immediately preceded by information sharing in the form of a group discussion, drawing on a short overview using visuals in a PowerPoint presentation (see Appendix G1). For KCRs, a separate information-sharing group discussion was held a few days before the main 'deliberative' activity. Points from the first discussion were raised and clarified immediately prior to this second activity. For the preliminary meetings with KCRs, the main facilitator used a set of visuals as laminated A4 cards to support information sharing and subsequent discussion in Kiswahili. These cards were developed from the

PowerPoint presentation, and used similarly to support information sharing (see Appendix C2), but also as the basis of a participatory game on the roles of parents in SCD inheritance, including the role of chance. A copy of these cards is given in Appendix G2.

2.4.4 Data collection in Phase Two

Community members of all types were involved in discussions around three main sets of questions identified from Phase One concerning: disclosure of findings on SCD status for *affected* children and *carriers*, and on responses to *requests for paternal testing*[50]. Following information sharing, data on opinions, reasoning and underlying perceptions were collected primarily through group discussions, given the potential for interactions within the group to support debate and exploration of opinions (Burgess 2004; Kitzinger 2006). Overall, discussions were held across ten groups of community members, where groups contained between three and six members, as well as nine individual and two 'pairs' interviews. The tool used to support these discussions is included in Annex D1 and D2.

Two parts of the discussion were each initiated by considering two different 'scenarios'. The first scenario[51], aimed to explore perceptions of SCD and its implications for families in order to support reasoning on the importance of information sharing for children affected by the disorder. This scenario involved diagnostic testing of a symptomatic child, and was later linked to a situation where this finding was generated through participation in research. Community members were asked to consider exactly what information should be given, how and for what reasons. In the second scenario - discussed after a refreshment break - community members discussed the case of a child found to be a carrier during screening in research. As for the first scenario, the main questions explored reasoning on exactly what information should be shared. As will be shown in Chapter 3, a key dilemma that was raised within these discussions - from both scenarios - was the risk that disclosure of the genetic basis of SCD would generate claims of misattributed paternity. This issue

[50] As described in section 2.2
[51] Both scenarios are included in the question guide in Appendices D 1 and D2.

was also explored in depth, with questions asked about whether requests for paternal testing were a likely consequence of disclosing SCD status information and how researchers should respond to such requests, including the nature of any information that should be given where paternal testing was undertaken.

An important feature of these discussions was that all participants were encouraged to contribute freely and openly, and that the views put forwards were explored carefully by the facilitator to establish as detailed an understanding as possible of underlying reasoning. To further support an informed and rational process, the main facilitator used probes and non judgmental forms of 'challenges' to the views put forwards, sometimes by drawing on differences in opinion emerging within the group and sometimes through the introduction of alternative ways of reasoning. As the tools show, these probes primarily drew upon considering the nature of outcomes of disclosure/non disclosure for different people in the short and long term, the responsibilities and rights that individuals in this situation might have, and how the interests of different people might be weighed against each other. For example, where some community members believed that information showing misattributed paternity for affected children should be withheld, I asked for discussion on how the father's position should be taken into account, including how participants would feel if they were the father involved in this situation. In this way, I aimed to support a process of informed reasoning that took different moral perspectives into account in developing conclusions. The method strongly drew upon Ives' (Ives and Draper, 2008) description of a similar process used to explore concepts of fatherhood in the UK, referenced earlier in this chapter (section 2.2).

As in Phase One, language presented a challenge for me in some of these complex discussions. For this reason, meetings with staff members and community leaders were held first, as I could conduct these in English, with some Kiswahili where needed. My role as the main interviewer/facilitator meant that I could explore emerging issues in the discussions directly, and strengthen the method and tool based on experience over the initial conversations. Later discussions with KCRs and affected mothers were held in Kiswahili/Kigiriama, limiting my capacity to guide and input in this

way. For groups of community leaders, KCRs and affected mothers, I worked alongside a lead interviewer/facilitator (Francis Kombe) who was a fluent Kigiriama and Kiswahili speaker, being a local resident as well as an experienced community facilitator within the programme (and a contributor to a staff discussion earlier in Phase Two as well as in Phase One). Mr. Kombe observed and supported the interviews I led with community leaders in English; all meetings were followed by a debriefing to discuss main emerging issues and any areas of difference between us. In the KCR and affected mother groups, my co-facilitator led the discussions while I listened and supported probing where possible. In these later group discussions, we were accompanied by a field worker (who had supported Phase One interviews) who kept notes based on general observations, and documented coded identities of participants for all contributions made throughout the discussions. Discussions were digitally recorded, transcribed, checked and translated - where necessary - as for data in Phase One. Participants in group discussions were given a code number as an identifier (P1, 2 3, etc) and asked to wear a badge with this number to support the note taker's tasks. Participants were only represented by these codes in the transcriptions developed. During Phase Two, I recorded approximately 23 hours of discussions with staff members, 22 hours with chiefs, KCRs and mothers of affected children and 3.5 hours with health managers.

2.4.5 Data analysis in Phase Two

Analysis of data in Phase Two was undertaken differently to that in Phase One, since a constant comparative approach based on inductively taking line by line meanings directly from transcripts was seen as less appropriately applied to data on processes of reasoning within groups. In the latter situation, the use of broader deductive themes were seen as more useful analytic approach from the outset, within which data from individual participants might move over time and in response to certain types of inputs from other participants or the facilitator during a discussion. At the same time, emergent issues were constantly looked for.

As in Phase One, the analysis began with an immersion in the data through repeated readings as well as discussions between main facilitators where relevant. An overview summary was made of each discussion, including overall comments on features of context that might be important (such as late-comers or noisy environments), group dynamics, the main emerging issues, areas of agreement and difference, and any surprising data. For discussions in Kiswaili or Kigiriama, Francis Kombe and I made summaries separately, then compared and reconciled these versions. A detailed analysis chart was made for each interview or group discussion, with data organised for each participant under broad themes of *reasoning for* and *against* information sharing. A series of subthemes were generated in this way describing the varied forms of arguments used 'for' and 'against' disclosure, by participant, as well as shifts in opinion and any important factors that seemed to prompt change. Accompanying these charts, I summarised the range of sub-themes emerging in each group and described the extent of agreement and difference within the group (for example, a particular community leader group), as well as across each type of group (for example, across all three community leader groups). Extracts from detailed and summary analysis charts for one KCR group are included in Appendix E1 and E2, respectively, to illustrate these stages of the process. At this time, I also began to develop a series of different visual representations to support my exploration of the relationship between the subthemes and the types of values that were emerging within different strands of reasoning (see Annex E3 and 4 for early and late explorations of visual representations of emerging values).

The next stage of the analysis drew on Macklin's (1999) understanding of the role of differences in perceptions about relevant facts, differences in moral principles or values that are drawn upon, and differences in the ways that such principles or values are weighed against each other as an organizing concept, as described earlier (2.4.1.2). The detailed analysis charts and summaries were used to support this process of mapping, including assessing the extent to which agreement and difference seemed to be a feature of particular groups or associated with other forms of diversity included in the sampling process. Figures showing the mapping of agreement and difference about

'facts' and 'values' are included in Chapters 4, 5 and 6, respectively. In addition, Table 6.2 in Appendix E5 was drawn up to examine agreement and difference *between types of community members* for the set of questions generating greatest controversy, that is, those addressed within Chapter 6.

2.5 Limitations for methods

There are several different types of limitations to the methodologies described in this chapter. In what follows, I discuss limitations that are associated with qualitative research methods in general, including the potential influence of sampling, data collection and descriptive and explanatory forms of analysis and positionality in both phases, as well as some specific limitations arising from the approach taken to normative analysis in Phase Two.

2.5.1 Sampling in Phases One and Two

The process of sampling throughout the study has been purposive, aiming to provide a range of different viewpoints on the issues under discussion. However both for the descriptive methods in Phase One and the discussion-based activities in Phase Two, the findings cannot be considered to represent the full range of perceptions or attitudes within the community. For example, it is likely that KCRs are slightly better educated and likely to have more prominent leadership roles within the community than typical community members (Kamuya 2007). Of relevance, a key finding of the study concerned heterogeneity amongst community members. Given the scope and intensity of the study, it was not possible to seek redundancy within the findings by extending the numbers of participants. Instead the sample in this study may provide more of a 'snap shot' of a wide range of experiences and opinions that exist across the community. A key group where opinions may be under-represented in Phase Two, as explained in section 2.4.1.2, is mothers of children with SCD who did not accept biomedical interpretations of the disorder or who had particular vulnerabilities within their families; these mothers were deliberately not targeted in this study given ethical

concerns described earlier. However, the community members who did participate were encouraged to consider the positions of vulnerable groups who were not included in the study.

2.5.2 Data collection and analysis in Phase One and Two

The analysis in this study largely drew upon reported data, although this was derived from a reasonably long period of study including several phases over two years, and sometimes interviews/group discussions with the same participants. No additional methods were used to triangulate findings, although comparisons between Phases One and Two were possible for many questions. A key issue already discussed that presented a limitation to flexibility in data collection was language limitations. This also had implications for processes of translation, which were strengthened by ensuring that the translation was undertaken by the main facilitator at the interview/discussion and that there were always two or three facilitators present at each interview/discussion who could discuss and corroborate findings.

The influence of group dynamics was a key potential influence in both phases. In Phase One, this particularly occurred when several family members were present, in which setting mothers were less likely to talk freely particularly about sensitive issues. In Phase Two, the issues were typical of group discussions in general (Kitzinger 2006); open and respectful discussion was however generally not difficult to achieve, partly given the existing professional relationships within groups and between groups and facilitators. At the same time, these existing relationships could have influenced the nature of discourses (Flick 2006). The main measures used to strengthen openness and reliability were the use of experienced facilitators, all of whom (apart from me) shared the culture and mother tongue of the community members in the study, and maintaining high levels of reflexivity in relation to all forms of interactivity. However, the methods used in Phase Two were both hybridised and to some extent innovative; the lessons learned from this process will be discussed in Chapter 7.

The analysis of data in Phase Two was both more complex and made less easily auditable given the decision not to use a qualitative data analysis software programme to manage data. Instead, the analysis relied on the development of detailed charts; these were complex to develop given the richness of data, the diversity in reasoning, changes over time and influences within groups.

2.6 Scientific and ethical approval

Scientific and ethical approval for this study was given by the institutional review committee and national scientific and ethical review committees in Nairobi (KEMRI Scientific Coordinating Committee and National Ethical Review Committee).

2.7 Chapter summary

Having described the nature of a dilemma emerging in a genomic epidemiological study in Kilifi in Chapter 1, along with its potential wider implications for other types of research in malaria endemic settings, this chapter has provided a description of the way in which I aim, in this thesis, to develop normative conclusions on resolving the dilemma based on empirical research. The methodological approach combines two phases of data collection: exploring experiences and context in Phase One (reported in Chapter 3), and deliberation by community members in Phase Two (Chapters 4, 5 and 6). In a final phase of normative reflection, in Chapter 7, I give my conclusions on researchers' responsibilities in these three areas.

This analysis interweaves community experiences, processes of community reasoning and *ex situ* normative reflection, described here as a form of reflective equilibrium. In this process, I aim to place community experiences, views and values centrally in the analysis, while referencing these to wider ethical debates, commentaries and guidelines in the literature. This focus on methodology addresses thesis objectives to explore the role of this form of empirical research in normative analysis, as well as to develop conclusions from its use. Given the relatively novel nature of these methods, in Chapter 8, I return to discuss experiences of working with these approaches to

normative analysis, including the way that the emphasis on community perspectives was achieved in practice (section 8.3).

CHAPTER 3

Phase One. Encounters with experience: Sickle Cell Disease and the Kilifi Genetics Birth Cohort study

3.1 Introduction: Background, aims and organisation of chapter

As described in Chapter 1, the broad aim of this thesis is to develop appropriate responses to important ethical questions about the disclosure of findings on sickle cell disease (SCD) generated during international collaborative biomedical research, including on the responsibilities of researchers, as part of exploring approaches to community consultation. Empirical research in the thesis focuses on SCD information sharing in a genomic epidemiological study (KGBC) at an international collaborative research programme in Kenya. This chapter aims to address the first specific objective: To identify important moral concerns of participants and community members around participation in a genomics study in Kilifi through an exploration of community perceptions, beliefs and experiences of participating in the research and sickle cell disease, including where screening was conducted through participation in research[52] (Chapter 1, section 1.5).

Research was divided into two main phases, as described in detail in Chapter 2. This chapter reports on the findings from Phase One[53], including:

- The experiences of families with a child affected by SCD in relation to the disorder;
- The perceptions and attitudes of *participant* families towards KGBC, including some families where children were found to have SCD;
- Perceptions of KGBC field workers from their interactions with participant families.

[52] As argued in Chapters 1 and 2, it is important to begin this investigation by situating questions about SCD screening and information sharing within a broader picture of the moral concerns of participants and community members related to this study.
[53] Given the iterative nature of analysis between Phase One and Two, explained in Chapter 2, some findings in this chapter draw on supportive data from Phase Two.

This chapter has two main sections. The first section (3.2) explores community beliefs, perceptions and practices in relation to SCD. The second section (3.3) describes community perceptions and attitudes towards KGBC. These are followed by two shorter sections which identify direct and indirect moral concerns emerging from these accounts (3.4) and the questions taken forwards for investigation in Phase Two (3.5).

3.2 Community perspectives on SCD

In this section I explore community members' perceptions and experiences of SCD in Kilifi from interviews with eight families whose child was found to have SCD during participation in KGBC; five families where the condition was identified following diagnostic testing at Kilifi District Hospital; and seven field workers with KGBC[1]. For children participating in KGBC who were found to have SCD, elements of their stories about study participation are described in section 3.3.

In many cases, the child identified for inclusion in Phase One - either through their positive screening in KGBC or through attendance at the SCD clinic at Kilifi District Hospital - was not the first child in the family to have been affected. During interviews, affected families described their experiences of and reactions to SCD over time, including those of their first child affected. Parents (and sometimes grandparents) described the way perceptions about the nature of this 'problem' shaped their care-seeking behaviour and impacts within the family.

Given the central interest within this thesis on researchers' responsibilities in SCD information sharing, a key feature explored in these stories is the *influence of genetic knowledge*, whether inferred from observations within the family or information passed on by others, including relatives, neighbours and biomedical or traditional health providers. In keeping with the literature in Chapter 1 (section 1.6) this includes ways in which family dynamics and perceptions and beliefs about genetic knowledge can be inter-related (Chilibeck, Lock et al. 2011).

3.2.1 Community awareness and recognition of SCD

In families interviewed in Phase One whose children had eventually been diagnosed as having SCD, including participants and non participants in KGBC, no family members - including grandparents where they joined in the discussions - had recognised the emerging intermittent signs of sickle cell disease (SCD) as a specific disorder or given a name to this collection of events in their first affected child. Instead, the various symptoms were initially recognised and managed over time as disconnected entities with varying perceived causes and in different ways.

Homa ya mifupa is the Kiswahili term widely used for sickle cell disease (SCD) in medical facilities in Kenya, or *ukongo ya misozani* in Kigiriama, the local language used by the majority Giriama population in Kilifi district, translating in both cases to 'fever/illness of bones'. During interviews, few families had heard of a condition called *homa ya mifupa* prior to being told about this condition, either as part of participation in KGBC or at the district hospital. For those who had heard of the condition, two described limited understanding ('inherited and affects bones') from the experiences of close neighbours or relatives with SCD. Others could only report having heard about it.

Low awareness and confusion about the nature of SCD was a striking and consistent finding throughout my research. For example, in Phase Two, a member of the District Health Management Team (DHMT) who grew up in Kilifi explained:

> "But people now wonder where is this disease [SCD] coming from?...whereas it has been here from time immemorial. So first of all the community are not aware of this disease." (IDI14)

Low general awareness of the condition was also reflected in KGBC field workers' perceptions, based on their initial experiences of communicating about the genomics study and screening for SCD with thousands of families across the study area. Over time, in response to their repeated individual explanations and the research programme's wider community engagement activities in support of KGBC, field workers described an increase in community understanding of at least

some biomedical aspects of SCD. They expressed relief that their task in explaining this condition, as part of the informed consent process for KGBC, became correspondingly easier over time.

3.2.2 Beliefs about cause and looking for a cure: Anxiety and fears

Inevitably, many children with SCD went on to experience more serious effects of the disorder over time, including severe pain due to crises, ill health associated with serious infections, frequent admissions to hospital, chronic tiredness, embarrassment and discomfort due to abdominal swelling and difficulty in maintaining normal social and school lives. Parents' responses to these varied and often severe symptoms differed between families, and over time within families, influenced by the child's current state of health and their beliefs or suspicions about the cause of symptoms.

Amongst affected families, perceptions of the underlying causal mechanisms were broadly of two types: natural causes, with explanations drawn from biomedical traditions, such as a form of malaria; or supernatural causes, particularly ancestral curses (*laani*), spirits (*majini*) or devils (*shetani*). Initial perceptions of the cause of SCD were generally in keeping with the usual health beliefs of that family, but the lack of a cure for this genetic condition led to many families moving pragmatically between different types of health care over time, with similarly switching beliefs about cause. Pragmatic combinations of biomedical and traditional health seeking practices were therefore frequent. Most commonly, a sequence of different actions was undertaken over time on a trial and error basis in an often highly anxious search for ways of relieving symptoms, particularly where symptoms were severe. Uncertainty has similarly been described as underpinning 'switching' care seeking patterns for other conditions and settings (Nyamongo 2002). The severity, unfamiliarity and intermittent but long term nature of the symptoms generated high levels of worry and distress in some families, strengthening the perceived need to find a remedy, and also to ascribe a cause. In this way, a family with a more severely affected child would, through their anxiety and distress, be more likely to try out many different remedies and to engage hard in

identifying a cause. As the father in Family F03 explained about his state of mind during a period of confusion and anxiety before a diagnosis of SCD was made in his child: "I was like a mad man."

One mother, in Family F06, gave a detailed description of her care seeking efforts before a diagnosis of SCD was made in her first child; this account is highly illustrative of the anxiety and desperation experienced by parents in this situation, as the following extract shows:

> "They tested the child for malaria but the child had no malaria. They gave the child normal drugs like paracetamol, amoxin, these are the drugs the child was being given. You would give the drug to the child, but the child would continue crying all the night, even if you then take the child to the hospital. The doctors would say I didn't give the child the proper dose. I used to tell them I'm giving the child the right dosage but the child doesn't recover. Another doctor prescribed some drugs after several visits to the hospital and told me to go and buy them. The drug cost 500/= [about $5] and I went to buy it. [Interviewer: How many times did you go before he was tested?] It's too many times, even at night I would go, because I went until my book [outpatient record book] got finished. I can't say how many times I went until the book got finished. I would go even at night. [Child's] time was very hard to me. I'm telling you the book got finished, we bought another book and the child didn't recover or feel at least better. It was so stressful and troublesome. I even said if this child is not a blessing, he should just die." (Mother, F06)

Later, in Phase Two, the same mother further highlighted the implications of low awareness of SCD for care seeking, the likelihood of a supernatural explanation and for anxiety caused by uncertainty about the cause:

> "…this information that we are talking about here [the nature of SCD] if it could have been there during the time of our grandparents, we couldn't wonder about, we couldn't go to traditional healers because I went to traditional healers until my child was one year and some months and I was absolutely confused, the child was sick suffering and I had not known. When I went to the elderly people they told me that I had demons…I lacked knowledge and also my parents had no such knowledge, there was nobody with such knowledge. If there could be anyone with knowledge s/he could have explained to me to go to the hospital for the child to be tested. But because there was no knowledge, I spent most of the time at the healer, moving from here to there." (Mot01/P3)

3.2.3 Supernatural explanations for SCD

3.2.3.1 Supernatural explanations are likely, but not universal

As suggested by the quotation above, the most common initial interpretation of SCD symptoms in a young child in families was as a supernatural condition. As a member of the DHMT later pointed out:

"…the majority of our community when somebody is sick, the first thing is witchcraft" (IDI14).

Supernatural explanations for misfortune are common in many settings in Africa and elsewhere, and have been described in detail for a similar nearby Giriama community in the late 70s and mid 80s (Parkin 1991). In Kilifi, families talked about supernatural causes of illness as resulting from various forms of witchcraft, curses, devil or 'natural' spirit activity. These could arise directly through a malign influence or be generated by breaking important taboos. Particular types of traditional healers (diviners) would undertake the task of 'diagnosing' a problem thought to be related to a supernatural cause, often with the result that another individual would be identified as the source of the problem. Ancestral curses were a likely option for SCD, given the possibility of recurrence across generations. As a mother of a child with SCD shared, with some frustration:

> "According to my belief and what I am hearing, I think we can say it's like a curse from the lineage. It could be from the fore fathers or ancestors, I can't even explain what mistakes they could have made so that the curse follows these grandchildren."(Mother, F03).

KGBC staff and other community members in Phase Two pointed to aspects of SCD that would make it particularly prone to interpretation as having a supernatural cause. These included low awareness of alternative biomedical explanations for the disorder; its long term and therefore 'incurable' nature; the severity of symptoms; the diagnosis of anaemia, interpreted generally as 'lack of blood'; the potential need for blood transfusions; and the possibility of unexpected and sudden death in young people[54].

The association of SCD with 'lack of blood' and transfusions was seen as particularly indicative of the work of devils or evil spirits. Such beliefs have in fact been implicated in rumours about blood and blood sampling during research in the KEMRI Wellcome Trust programme over many years (Molyneux and Gikonyo 2005; Gikonyo, Bejon et al. 2008), and are similarly reported from several other similar settings (Marshall and Rotimi 2001; Geissler and Pool 2006; Leach and Fairhead 2006). A common rumour in Kilifi links the practice of blood taking as being on behalf of devils or spirits, with the generation of wealth as a reward or payment to the intermediary who collects the

[54] These considerations are discussed in greater depth in Chapter 4.

sample. As a result, people within the community who were wealthier than others, even to a minor degree, were particularly likely to fall under suspicion of being 'devil-worshippers'. As a community facilitator pointed out:

> "There is one belief in our communities that you see, that if it [an illness] affects the blood, there is this belief of *jinis* [spirits] taking the blood. If its transfusion or what in blood, but always that *jini* is there, and that might either affect the same family or the neighbours, because they might think somebody, somewhere is playing with my kid, there is also that belief" (IDI09/R2).

At the same time, it is difficult to know how widely supernatural beliefs and related practices would apply across the very mixed community in Kilifi district. A district health manager in Phase Two qualified his description of the prevalence saying:

> "There are *some* places where people are *still* really believing in the those very old cultural beliefs, and that is really affecting the service provision now because they still believe such diseases especially like sickle cell diseases or blood related diseases or inherited diseases are actually as a result of a curse that is following through a certain generation" (IDI11) (my italics)

Community members who described these influences were nearly always talking about a sub-section of the community, specifying - where asked - those with a 'lack of education'. Others suggested that shifts in traditional beliefs were now occurring, tied into larger developmental trends, most notably driven by education and economic progress. However, some well educated and articulate participants described their own ambivalent feelings when others raised the issue of witchcraft, for example, as a possible cause of health problems. Supernatural causes seemed to be able to generate some fear or apprehension even for those who did not openly accept their influence. A possible indication of this ambivalence was that a common way of talking about these supernatural influences was as stories or rumours, almost always generating laughter amongst listeners.

The common recourse to supernatural explanations was further heightened by low recognition of SCD amongst local health service providers. It was common for families to describe several visits being made to clinics before the condition was recognised, usually after referral to the district hospital. Even here, the condition was not necessarily picked up straight away amongst out-patients

(see for example mother F06 story in 3.2.2). Where a diagnosis of SCD was made following contact with biomedical health facilities, a label of '*homa ya mifupa'* was given by health staff and was then generally also used by the parents and family, whether continuing care was biomedical or traditional.

Low awareness of SCD amongst health providers was also described by health managers. For example a DHMT member suggested that SCD is:

> "…one of the chronic illnesses that most often goes unnoticed in the community, many people suffer the disease and they succumb, and they go to facilities, they go to health facilities, they go to a dispensary, they go wherever, *and the appropriate diagnosis is not made*."(IDI13) (my italics)

Perhaps most tellingly, SCD does not appear in the national register of diseases that are reported on monthly from district to national levels as part of disease surveillance. This report is compiled from all government health facilities in the district by the District Disease Surveillance Officer, and includes a section on chronic non infectious disorders such as epilepsy and anaemia, but there is no mention of SCD.

3.2.3.2 Blaming others for supernatural causes

Typically, some types of traditional healers were described as working through the identification of a person responsible for a child's ill health. Some community members explained that elderly relatives were particularly at risk of accusations of being 'witches' in these circumstances. An indication of the commonness and type of circumstances that might lead to these perceptions is captured by a community facilitator who suggested:

> "I think I would say it's quite usual for chronic illnesses, where people run around looking for how to help the child for quite some time, and initially when the child falls sick they will you know go to the medical doctor yeah modern medicine, but after that has failed they'll start seeking other remedies and that's where the problem begins, because they'll go to the soothsayers [traditional healers/diviner] and they will start telling them well the person who did this is just from your family, or from your neighborhood, because a lot of the time that is what comes out from those soothsayers. And that is where now you start thinking okay who could it be that is causing this problem? First from my family, and then of course the blame goes straight to those who are a bit older, because you know they have nothing to do, they are idle, they are seen around with weird, weird things [referring to objects associated with both traditional medicine and witchcraft] and that is where it begins." (IDI09/P4)

3.2.3.3 The possibility that children are blamed for sickle cell disease: Curses

The examples of supernatural beliefs described so far involve blaming an individual near to an affected child for causing the symptoms associated with SCD. Chiefs in one group and some field workers also raised the possibility that in some families who believe that children born with physical abnormalities are cursed these beliefs may lead to the child themselves being victimised. A chief recollected a recent incident in his community involving the prosecution of a father who killed his newborn child as a result of this type of belief. Other chiefs in this group agreed that issues of this nature did arise from time to time. This was discussed as an unlikely concern for SCD, given that children do not develop any symptoms until at least six months of age, but one that should be kept in mind. Other participants, asked directly, could not recollect any experience or story of a child with SCD being blamed themselves for the condition. In contrast, I note a report from Nigeria of children with SCD being harmed as a result of supernatural explanations linked to perceptions of malign forms of reincarnation (Nzewi 2001); and reports of harm to others seen as 'spirit children' as a result of a range of health problems in Ghana (Allotey and Reidpath 2001). More subtle forms of discrimination may occur however, as shown by a community representative in Phase Two saying that:

> "So now, the parents must know that this child of ours, it's because they inherited this from me, and they inherited this from the father…they are now a sickler. For us, being the way we are, we feel we're fine, but our child has this condition. So we must, or it's our duty to bring them up well without any worries, *knowing that they are one of our family members*." (KCR03/P3) (my italics)

Given assumptions of the culpability of an individual within many supernatural explanations for ill health, there would generally be negative consequences for the person implicated. These impacts, which could also affect the relationships and livelihoods within the wider family or community in which the person was situated, reportedly depended on the perceived seriousness of the act 'committed', but could vary from persuading that person to accept their role and make financial

reparations to the family to acts of physical violence against the presumed offender, including fatal wounding.

3.2.4 Looking for a cause: Family surveillance and blame

In this genetic condition, a further important influence on family perceptions of cause was the existence, or memory, of another family member believed to be (or have been) affected by the same condition across current and earlier generations. My interviews with affected families suggest that observations of SCD in a child could lead to a range of other health problems (such as polio, cerebral palsy, malnutrition and forms of arthritis) being linked to its occurrence. In seeking an explanation for the child's health problem, members of both the paternal and maternal clans would commonly scrutinise the extended family across several generations, interpreting observations as pointing to the source of the problem, including that one or both sides of the family were responsible. As suggested, these observations could generate blame through misconceptions about cause, sometimes underpinned by negative attitudes towards the individuals implicated. A good illustration of the way that surveillance can occur in families is given by the following story, related by a community representative, and quoted here almost in full to provide a picture of the way that the wider family and neighbours can be drawn into these questions of blame:

> "One day the child had that condition [SCD], the mother was the one at home attending to the child. When the neighbours asked the mother, she said this condition keeps coming back but I don't know what causes it. So, one mother told her "Maybe you were born with it, didn't you have this condition…maybe in your family there…?" The mother said, "No, I wasn't like that, maybe it's his father." The mother in-law was also there and she said, "My child [the husband] did not have that condition." So there were such arguments, and in the end another mother came up and said, "If everyone is saying it's not them, then who is to blame here? How did the child get this condition?" They were told that maybe when the husband comes, they sit down and talk, or they go to the doctor for a checkup…When the husband came…he was told about the child's condition, how it happens often, so he too said, "Maybe it's the mother, it might be her mother, because for me I haven't had that problem even when I was young. So she is the one who brought that problem." They were told that they will be forced, both of them, to go to a doctor for advice so as to be explained the cause of this illness and that's where it ended. Still they haven't gone to a doctor, and the condition comes back all the time." (KCR02/P4)

This practice of looking closely at family members over generations for signs of inheritance in genetic conditions, including a 'lay' tracking of features thought to be associated with familial

conditions, is not unique to this community. For example, the same phenomenon has been described for genetic disorders in South Wales in the UK, in which affected families described an intense process of scrutinising current and earlier generations for clues on the origins and pathways for the disorder, termed 'surveillance' by the author[55] (Featherstone, Atkinson et al. 2006).

3.2.4.1 Gendered family blame in SCD: High risks for mothers

Many families described the pressure placed on relationships between parents by the distress and confusion of having a child with SCD. At the same time, these types of disruptions were also seen as strongly influenced by the nature of the parent's relationship overall, as well as other contextual issues and the influence of changing times. In addition, to some degree blaming was described as a normal part of humour in family life. As one community facilitator described:

> "… blames in families are just like part of life…we always blame each other in families for various reasons including a naughty child, you joke around by saying "you look like your mother because she used to be as naughty as you are when she was little!" I mean these sort of things. So I think…it's [blame] kind of evenly distributed to all the family members, both the parents and even forefathers and all chains of you know in the family trees" (IDI09/P4)

However, a strong implication of gendered blame often resulted where mothers had more than one affected child, particularly where no previous generations were known to be affected. For example, two children in family F09 had been diagnosed as having SCD at Kilifi district hospital. At the time of interview, the family suspected that the earlier death of an older child, who had been treated by a traditional healer, had also been as a result of this disorder. When I visited, the mother had recently given birth to a fourth child, too young to be tested for SCD. Her sister in law described her certainty of the mother's responsibility for all her children's suffering, telling me:

> "The baby is still young but will come for the test too, because *all her children are born that way*. That one who is still young will then be in trouble." (my italics)

In addition, community members in both phases described a common general attitude in which mothers would be more likely to be blamed than any other family member for the occurrence of any health problem in their children. As a KGBC field worker commented:

[55] Also see Chapter 1, section 1.6

> "I think…we men, you know, are…hard to take things, in fact, things which are on the negative side, I will try to pull myself out." (FW2)

Similarly, a community facilitator explained:

> "Let me be one of the fathers, us fathers, we normally if anything happens [to the child] in terms of diseases, we normally put a blame to mothers." (CF4)

The likelihood of fathers being resistant to accepting their role in SCD was also described by most field workers and community facilitators, as shown by the following comments from a field worker during a group discussion:

> FW7: "…most men from the community don't think they can, that their blood can be having this sickle cell disease, so they're always blame shifting, they always shift the blame to the woman"
> Moderator: "Are we just talking about sickle cell disease?"
> FW7: "Sickle cell plus other diseases…he will always look for loopholes to blame, that is how the majority of men in the community behave."

The tendency for gendered blame was described as due to various perceived influences, such as the mother's prolonged period of contact with an unborn child during pregnancy; her potential to act as a channel for supernatural forces through contact; aspects of mothers' parenting skills perceived as inadequate; and the mother's role as representing an 'outsider' coming from the maternal into the paternal clan. As will be explained next, her sexual fidelity might also be called into question. Illustrating ideas about the importance of maternal-foetal contact, a (female) community member explained:

> "Mostly in such situations, women are mostly blamed, like us, because if there is anything at home that, if they don't know where it's come from, or if they don't understand, they start blaming the mother, maybe it's her, because I am the one who carries the pregnancy. I mean I carry the child in my womb. So if anything happens, they will say it's her." (KCR02P5)

3.2.4.2 Questioning marital faithfulness and paternity: High risks for mothers

An important source of blaming towards mothers of children with a health problem, including SCD, which was perceived as reasonably common, was the occurrence of doubts about paternity and common attitudes of jealousy in men towards their wives. As will be suggested in this section, elements of this attitude are likely to be structural. In this regard, the Mijikenda community is

described as a patrilineal society by anthropological convention, or one where social and political identity and ancestry are constructed in the male line (Shaw 2006). Particularly in rural settings in Kilifi, marriage has traditionally been arranged between families. Although cultural attitudes and practices are inevitably mixed and in a process of continuous change, this arrangement builds in inequities in power, notably by the giving of a bride price by a man's family to his prospective wife's father. Although geared towards maintaining continuity of the paternal family line through the successful bringing up of children (Parkin 1991), the system also makes women's lives and livelihoods vulnerable to their husbands' use of power. Jealousy and accusations of marital unfaithfulness are therefore potentially very damaging, leading at times to mothers being sent back to their maternal homes with the 'outside' child[56]. Social support for such a mother is not guaranteed in the maternal home, particularly if demands are made for the bride price to be returned.

The phenomenon of marital infidelity was generally referred to as 'going out', in the sense of 'going outside marriage'. When a young child developed health problems, doubts about the cause thought to be related to wives 'going out' could arise in three main ways, one of which could in fact implicate either parent. This first way was through the locally recognised condition '*chirwa'*. *Chirwa* is related to the breaking of an important taboo on parents of a relatively new born child having sexual relations outside marriage during a fixed period of time following birth. It is characterised by a general failure to thrive in the young child that may lead in severe cases to their death in the first year of life. If a child is suspected to have *chirwa*, the father is most likely to be seen as responsible, although traditional healers would be able to specifically identify the wrong-doer. An interpretation that *either parent* might experience this doubt is illustrated by a community representative saying:

> "So If I happen to get a child with that condition [SCD], there definitely will be conflicts there, so one [of the parents] might say, "Ah! You are the one who went out and brought this illness because you were moving from one person to the other, and then you came to me, so I think you are the one who brought this illness." (KCR02P2)

[56] This topic will be discussed in more depth in Chapter 6

Other ways in which a mother perceived as 'unfaithful' could more specifically be blamed for adversely affecting the health of her child were through bringing HIV/AIDS into the family or simply through misattributed paternity. In addition to the cultural tendency towards blaming mothers, an underlying understanding that fathers are *biologically* more vulnerable to deception about parenthood was expressed by a community representative using a local saying that '*mwana ni mameye*'[57] (KCR01/P1), meaning 'only a child's mother knows who the real father is'. Similarly, two (male) community representatives jokingly hinted at this attitude in the following extract, taken from a discussion of how commonly children in the community might be born out of wedlock:

> P4: "Women are very clever, she can go outside and she gets pregnant, and then when she comes back she pretends so much that the child is yours, then this, ha, his nose is like yours, when I look at his nose it's not like mine (laughter) while she has persisted saying his nose or his fingers . . ."
> P5: (interrupts) "...they are from their grandfather who died!" (all laughing)

Similarly, a field worker described this general tendency in the following story:

> "Let me share an example. In our area there was a lady who was married, the wife's complexion was brown and the husband was black in complexion. And she gave birth to three children and two of them were brown in complexion. When the man saw that the children are brown, they had to divorce because the man was saying that 'these are not my children, why are they brown when I am black? So you have to go back'…up to now she is living with her parents." (IDI07/P2)

An example of jealousy and its relation to structural gendered attitudes with the community is provided by the exchange below, quoted in full in order to understand the context of the discussion for such a potentially sensitive issue, including participants' ways of speaking about power and its impact in marital relations, and the way that humour was often a component of such discussions. The final contribution to this exchange was made by a woman (P3).

> P2: "Aa, some groups don't easily blame each other, while some groups it's easy, like for us Mijikendas, that habit is very common."
> P4: "Jealousy!"
> P2: "We blame mothers mostly, but there are other groups which even if they see a thing like that [suspect marital infidelity], they take it very easily so they don't blame that much. So, you will see they carry on with their activities."

[57] Literal meaning: 'the one [known parent] is his/her mother'

Interviewer (A Mijikenda man): "But the Mijikendas tend to blame their wives a lot?"
P2: "A lot."
Interviewer: "Why? Why do Mijikendas blame mothers mostly?"
P2: "Like P4 said, it's jealousy"
Interviewer: "Jealousy, jealous about what, mh?
P4: "If you leave your wife at home, you want to find her there when you come back, but if she is not there, you think that maybe she's gone to her 'friend' [meaning lover]. That's our problem, us Mijikendas. Lack of trusting each other!"
P3: "Mh, here's an example of that jealousy, a man being jealous of his wife. Even us mothers feel jealousy. There's an example at X, of a man who used to sweep his compound with a broom backwards when he left his home until he is completely out. He sweeps as he leaves, backwards so that he leaves the place without any mark of any person's foot! (All laugh). So he continues to sweep even his own footprints so now that wife, the husband has studied the wife so he knows her footprints, the way they are so now, he should not find any other footprints when he comes back. He should not find any other footprints there or the wife will get a beating because it will be 'Whose footprint is this one? Whose is this one?' So, that's jealousy, because he doesn't want people to go to his home or he doesn't want other people *to play with his property*. So, he wants it to be his alone." (my italics)

3.2.5 Gendered impacts of blaming: Broken livelihoods and families

As the earlier description of structural imbalances of power between some husbands and wives shows, although both blaming and jealousy clearly could work both ways, attitudes of blame often have greater implications for women in this community (Marsh, Kamuya et al. 2011b), as also suggested by the story above. If, for any of the reasons discussed in this section, mothers were blamed for causing their child's condition their position could often be very vulnerable where - as was commonly the case - they lacked economic independence. If separation occurred, mothers with greater means would be able to manage their and their child's lives independently, while others with fewer resources were likely to return to their maternal homes. For many families, this would be economically difficult and seen culturally as a failure on the part of the mother and the maternal clan. The affected child would be likely to be sent with the mother, particularly where the husband believed that he was not the father, adding to the demands placed on the maternal home. Most often, the mother would have to leave any other children behind with their father, in keeping with traditional marriage agreements. As before, any demands for return of the bride price would worsen the mother's situation.

One outcome of paternal denial of responsibility and gendered blaming was that fathers might seek to be tested for SCD, in order to support claims of misattributed paternity. In this way, the father in family F09 denied his responsibility for his children's condition, even though a paternal history of SCD was known. The father argued for a negligible influence of the paternal clan through comparing the *proximity* of affected generations on both sides of the family, saying:

> "You know in my father's generation and my generation sickle cell is not there, but on my wife's side, it is there [in her generation]."

His negative attitude to his wife persisted for many months, leading to his making several forceful requests for paternal testing at the SCD clinic, to *show* whether or not he was responsible for the children's condition. These requests were refused, generating heated exchanges.

Highlighting the gendered basis of this process of blaming, both parents in family F06 perceived that the father was largely responsible for their two children having the condition, since many family members on his side were similarly affected and none on hers. However, the mother did not express any blame towards her husband, but rather described her relief in his acceptance of the situation, saying:

> "Since their father has accepted them, and also me I have accepted their condition, we are finding it very easy. If a child is sick, the father goes to work and by the evening he comes in bringing fruits, milk… so it's like *he has accepted*." (my italics).

As can be understood from the preceding sections, the impact of SCD on families included those related to the symptoms experienced by the child, taking care of an affected child and impacts on relationships within the family. These impacts were therefore psychological, physical and economic in nature. All these types of impact were accentuated where children were severely affected, including high levels of emotional distress as well as economic costs of home care and treatment seeking.

However, while the entire family would be affected by the costs of caring for a child with a chronic lifelong disability, economic losses particularly impacted on mothers, who were unable to establish or maintain income generating activities as a result of the high levels of child care needed, and had

relatively few options in coping with the disorder within their families. In this way, a mother described her life at home with two affected children, saying:

> "They will cry till you won't be able to do anything… I won't wash clothes and I won't be able to do anything, I will stay with the child the whole afternoon … the problem with them if they are sick is they don't want to sit, don't want to sleep but only want to be carried" (Mother, F06).

Another mother talked of the limitations these types of worries placed on her life:

> "When he has gone to school, like now, I don't ever leave the house and go far until he comes back home."(Mother, F03).

The loss of income these restrictions entailed was described by a mother with affected twin boys saying:

> "If the children are sick, will you leave them? I was frying potatoes but when my children started getting sick then I stopped that." (Mother, F10).

Those who were most secure financially were least affected by this challenge to an independent livelihood. A mother who ran a small busy general shop, explained*:*

> "When this [a SCD crisis] happens, I don't see this [finding someone else to run the shop] as important, I just close the shop. I haven't looked for anybody, even the time I was in the ward I closed it, till I was discharged is when I came to open the shop." (Mother, F08).

In the situation in family F06, where both parents perceived the father as responsible for their children having SCD, the mother's account of her powerless to improve or remove herself from this situation contrasts sharply with that of a husband who has the option to send his wife back to the maternal home. Her child care responsibilities prevented her from seeking financial independence, her own family was reluctant to take her home with two chronically ill children, even for short periods, and she could not countenance leaving the children with her husband and a future substitute 'mother'. As she explained:

> "It's me and my husband who are to take care of our children but to tell the truth if I leave here and go to my home, the mother who will come [another wife] won't live well with my children… and if I decide to take my children to my home there won't be any one who will offer to take care of my children". (Mother, F06)

The form of blame captured by these stories of mothers' experiences of SCD moves beyond a negative attitude towards individual mothers to include the structural gendered influences described

earlier in this section. These influences relate not only to attitudes of blame but also to mothers' ability to manage their (and their children's) lives in the presence of blame, potentially leading to cycles of harm. Structural forms of blame are similarly discussed for discrimination or stigmatisation in some of the literature on HIV/AIDS in developing countries. In this way, Parker (2003) describes a structural concept of stigma as one that 'feeds upon, strengthens and reproduces existing inequalities of class, race, gender and sexuality' (Parker and Aggleton 2003). This concept takes account of social theories describing the work of power, culture and difference in producing and reproducing stigma and discrimination, where discrimination is defined as 'acts or omissions derived from stigma and directed towards the stigmatized' (UNAIDS 2005). As a result, in the field of HIV/AIDS research, the structural basis of gender discrimination for women in many developing countries is acknowledged, including that 'women are (already) economically, culturally and socially disadvantaged and lack equal access to treatment, financial support and education. Being outside the structures of power and decision-making, they may be denied the opportunity to participate equally within the community and may be subject to punitive laws, norms and practices exercising control over their bodies and sexual relations' (UNAIDS 2000) (p13).

This wide concept of stigma, incorporating structural forms of both blame and discrimination that work at a 'macro' level, seems applicable to the situation of some mothers of children with SCD in Kilifi. It contrasts with a much longer established concept of stigma as a 'discrediting attribute' that focuses on the 'micro' level responses of individuals and those in relationships with a stigmatised individual (Goffman 1963). At the same time, stories of SCD in Kilifi underline the importance of influences at both macro and micro levels. In this study, the physical signs and symptoms of SCD, which could be interpreted as 'discrediting attributes', clearly led in some families to increased concern and pressure on relationships within the family. As a father explained:

> "To tell the truth, when we knew there was a problem, I even said 'it's better I didn't marry you' because I felt very bad, and when things started to calm down then we sat and decided that we just leave this to God and he will know what to do." (Father, F02).

Similarly, individual characteristics such as the mothers' educational and economic status were described by staff as impacting on both the likelihood of maternal blame and the gender discriminative effects of SCD. An important element described was the ability to make financial contributions to the household, and this was linked to educational background. An influence of individual characteristics of personality and the nature of relationships is well illustrated by the father in family F03 who, despite believing his wife to be mainly responsible for their child having SCD (related to closer generational proximity of affected family members on the maternal side), explained:

> "So we thought maybe from her, that's why it has come to us, but, okay, this is an illness, and even if I follow it up, where will I get the answer? The only important thing is to find treatment for it. That's where I ended." (Father, F03)

3.3 Community perceptions, experiences and attitudes towards KGBC

In this, the second main section of this chapter, I will move away from an exploration of community perceptions of SCD to investigate understanding of, and attitudes towards, KGBC drawing upon interviews with 17 families in Phase One. Nine of these families' children had been given feedback on a 'negative' SCD screening result; the remaining eight were given positive results. I begin with an exploration of reactions of all families whose child participated in the study (3.3.1) and then go on to investigate specific responses of those whose child was found to have SCD (3.3.2).

3.3.1 Why people decided to participate (or not) in KGBC

3.3.1.1 Perceived value as a health or SCD check for community

The predominant reported perception of KGBC amongst all the families interviewed was that the study aimed to provide "assistance" to the community through surveying for health problems in young children, often with SCD as the main or one of the disorders being addressed. Participation was therefore generally well understood to be voluntary, but perceived as important for the sake of the child.

Thus the main perceived benefit of participation in this study was access to a screening test for either SCD or an undefined health condition, with an assumption that treatment would be made available by the research centre to either cure[58] or ameliorate the effects of any disorders that were found. In the group interviewed, families did not recollect that the aim of KGBC was to learn more about children's intrinsic susceptibility or resistance to common and severe diseases in Kenya[59]. The decision to participate or not was reportedly sometimes made by the mother and sometimes required consultation with a husband or older male member of the family.

KGBC field workers looked at this situation slightly differently, explaining that families varied in their reactions to and understanding of the study. All agreed that the study had been very difficult to explain, particularly at the start, with discussions sometimes taking more than two hours. However all felt that at least some participants did understand the genomics part of KGBC, but that their decisions to participate were often influenced by other factors. These factors became dominant for those who had more difficulty seeing the primary scientific goal of the study. For KGBC, the most commonly occurring 'distraction' of this nature was SCD screening. All field workers saw that this was the feature of the study parents were most interested in, as well as being best understood and remembered. As one field worker explained:

> "At the end of the consent you'll then ask questions to see if they have understood the whole thing, you'll find most of the questions or the answers are just from the sickle cell part." (FW02 Phase One)

Similarly, another described that:

> "Okay you might go deep, giving all those examples and all that, but just send someone tomorrow and ask there, okay? Okay there are some, a few probably, they'll tell you about the inheritance issues and all that, but quite a good number they will tell you about '*pima homa ya mifupa*' (testing for sickle cell disease)" (FW03 Phase One)

Families' focus on SCD was often seen to be prompted by field workers' own explanations about this condition, as part of the informed consent process:

[58] This perceived 'treatment' role of the research centre is closely linked to common community perceptions of the research programme as providing high quality medical services, as we have reported elsewhere (Molyneux, 2005b).

[59] See Chapter 1, section 1.3, for more information on KGBC

> "It's just from what my colleague was saying because you can just tell the whole story about the study but at the end they just stick to sickle cell even they can start explaining, oh this is what happened to another kid at the neighborhood! So they just accept the study because of the benefit, so and also they don't want the kid to be like that of neighbour so they just accept because of that." (FW01 Phase One)

The fact that some participants did have some understanding of the main scientific goals of the study, but that SCD screening was perceived as the main benefit, is also reflected in some reported community reactions to negative tests. As a KGBC field worker described:

> "There are already some questions, you know ... there are parents also asking, okay, fine, my child is negative, yeah, so how about the child? Why do you just leave the child like that? Okay he participated, yes, but he is not positive. Does that mean it's all over for him? We are waiting for maybe vaccines or maybe for better tabs in future? You see so those already are questions that are lingering in the people's heads…" (IDI07, Phase One)

In addition to the inclusion of SCD screening, field workers perceived that general attitudes to and expectations about the research centre acted as an important influence on decisions to participate in KGBC. One such perception, which has been recognised more widely from other research settings as well as within other studies in Kilifi, was that the research centre had a primary role to act as a high quality hospital (Molyneux, Peshu et al. 2004; Molyneux and Gikonyo 2005), where 'quality' was implied by the high level of resources and standards of care often associated with studies. This perception of research as treatment, which has been referred to as a therapeutic misconception of research (Appelbaum, Roth et al. 1987), arises largely from low familiarity with the nature of research, much greater familiarity with health care, and the fact that these two activities are sometimes overlapping. These types of concerns and rumours have been reported extensively from other similar settings (Marshall and Rotimi 2001; Leach and Fairhead 2006). Diagnostic 'misperceptions' (Clayton and Ross 2006) or interpretations of research have similarly been problematised[60]. A diagnostic interpretation of KGBC was particularly strong for a mother whose child had been discharged from the district hospital a few days before a field worker visited her at home to request participation in the study. As she explained:

> "He [the KGBC field worker] said he had come to take blood to go and test to see about any child's health if it was fine or not, or to see if the *virusi* [virus/bacteria] are finished or they are

[60] See Chapter 1, section 1.8

> still there, or if there is another illness coming they could also know. I didn't ask him where the test would be done because my heart was seeing that because he has come with [child's] name, it's because I was in the ward recently at the hospital. And in the ward the KEMRI [research] people were really visiting me so I knew… he has come from the hospital with the name of [the child]." (Mother, F19)

Even where no particular predisposing influences - as above - existed, community perceptions of KGBC show that inclusion of SCD screening could lead to diagnostic interpretations or 'misconceptions' about the purpose of the study. The quotes from KGBC field workers in the previous paragraph, however, suggest a more subtle influence of SCD screening on community perceptions, in that - for some families at least - this was less to do with a straightforward misconception and more to do with a very high perceived importance for SCD screening that acted to crowd out interest, understanding and possibly even remembrance of the wider study. Although not specifically explored with all field workers, one staff member described an instance where - following an explanation of the study and screening for SCD - a mother refused to participate on the basis that she did not want to know if her child would develop an illness in the future; she was content to know that he was currently well. This attitude stands in contrast to the appreciation that most families seem to have shown towards early detection of this disorder, at least from a prospective position. As I will go on to show, some whose children were found to have SCD did not always subsequently value this 'warning'.

3.3.1.2 Perceptions of access to high quality care through research: Expectations of reciprocity or perceived coercion?

According to KGBC field workers, an additional influence on decisions to participate linked to perceptions of the research centre as a high quality hospital was the expectation from some families that agreement would increase their chances of getting access to this 'high quality care' if this was needed in future. Two field workers drew on this phenomenon to explain why families in more rural areas were often more willing to participate than those from urban settings, even sometimes requesting participation when not invited:

> FW01: "Maybe the reason [that urban families are more likely to refuse to participate] is nearness to the hospital so sometimes when they are sick they just rush. So as compared to somebody who is in X [a very rural area] and you are recruiting that guy, they say I just want to be involved because if my kid falls sick *I may see you in the hospital and you can assist me*… so those sorts of things." (my italics)
> FW02: "Even you can go to homestead, the kid is not in the child list you are looking for, but the parent is very willing [for you] to recruit the kid."

This perception that participation might lead to a greater willingness on the part of researchers to assist if needed later could be interpreted as an effort to build a relationship within which some obligations of reciprocity might later emerge. Other accounts of the influence of views about the research centre as a quality health provider more strongly imply a form of perceived coercion. Where this occurs, the perceived nature of SCD, as explained during informed consent for KGBC, might strengthen a feeling of powerlessness to refuse to participate. The difference between building an expectation of reciprocity and feeling pressured to participate through fear of losing access to future care might at times be quite subtle. However, a perceived form of coercion is clearly illustrated by a father's account of his and his wife's decision to join the study, while reflecting on the information they had been given by a field worker:

> "…we had a long discussion and then she [the mother] said if we refuse, if she [the child] is affected [by SCD in future], we won't be able to take her to KEMRI [referring to SCD clinic, perceived as part of research centre]. That's what she said, and therefore we (pause) we had no choice apart from taking her to KEMRI." (Father, F07)

3.3.1.3 Concerns about KGBC: Rumours and trust

While valuing the opportunity to check their child for SCD, most families I interviewed had concerns about KGBC when the study was first explained. Those who were not concerned included a mother whose child had already been diagnostically tested for SCD and was subsequently recruited into KGBC; two families where an older sibling was known by the family to be affected by SCD (in one of these families, they were pleased with the opportunity for testing); and one family where an influential educated elder was dismissive of negative rumours about the research centre (as being "from those who are not educated") and supportive of SCD screening.

The main issue underpinning concerns at the outset was doubt about the motives and therefore trustworthiness of researchers. This concern reflects a much wider set of misconceptions and

rumours that have been encountered for research in Kilifi in general, stemming from the low familiarity with research and confusion between health research and health care described in the previous paragraphs. In this situation, the community observations of the level of resources available to the centre (that led to positive perceptions of the quality of medical services offered) generated concerns about the source of this 'wealth'. I have already described, in the previous section (3.2.2.1), that traditional beliefs in Mijikenda culture link the existence of wealth with the work of supernatural forces, particularly 'devil worshipping'. For KGBC, some families wondered why researchers were interested in children who were well and had concerns that children's blood samples might be used as part of devil worshipping activities or be sold to generate funds.

These rumours were amplified by the particular and unfamiliar way that blood samples were taken in KGBC, being via the heel[61] rather than the more commonly experienced site on a finger (Marsh, Kamuya et al. 2010). Some families accepted field workers' explanations that heel pricks were less painful and safer, others were highly suspicious. Some staff reported that at times during the early stages of this study mothers would run away from their homes when they heard the field worker's motor bikes approaching. This tension concerning perceived motives for blood taking led to great importance being placed *on getting results back*, not only to provide reassurance about the child's SCD status (where, as in 99% families, this was reported as 'no SCD') but also importantly as providing proof that trust had not been misplaced. As a grandfather pragmatically deduced: "What devils take blood and bring back the results?" (IDI14)

The parents of a child who later turned out to have a positive SCD test described their concerns about the unfamiliarity of the heel prick based on rumours in relation to blood taking. Even after agreeing to participate, these anxieties were still sufficient to lead the father to check with his next door neighbours if the story they had been given by the KGBC field worker matched that given to others:

[61] This site was used because of greater safety and less discomfort in young children than for a finger prick, and the possibility of drawing larger samples than would be possible from a finger prick.

> "I asked where he [the field worker] was going to next … so that I could go and ask if he did same there as he did here. It was a neighbour I know, so I went later to ask and found out that her child had also had blood taken from the heel. So I knew that many children will be done the same." (Father, F12)

Over time all KGBC field workers reported that increased familiarity with the procedures and the perceived aim of the study led to a marked reduction in community concerns about participation, including taking blood from the heel and taking blood in general. This was seen as making their work of recruitment and explaining the study much easier. One way in which this happened was that KGBC field workers repeatedly re-visited some households following the births of new children over time, given that many families live within an extended senior patriarch-led arrangement of smaller 'nuclear' family homes within one geographic compound (Molyneux, Murira et al. 2002).

3.3.2 Being given 'negative' results: Anxiety of waiting and relief of learning

All families and KGBC field workers agreed that families who were given results showing 'not SCD' expressed some or great relief. However, as a result of concerns about the participant child's SCD status and the trustworthiness of the researchers, waiting for results[62] was a particular concern for some of the families interviewed whose children were found 'not to have SCD', as shown by both participant families' and field workers contributions. While waiting, some parents described that they were reassured to have been told that all positive results would be returned within two to three weeks, while others remained concerned that an early positive result might simply not have reached them. The family's understanding and memory of the significance of the timing of feedback was important. In one family (IDI17) for whom I returned results showing 'not SCD' during my interview several months after screening, the parents' relief was evident. The father

[62] Study procedures meant that these families could wait up to four months[62] for results to be returned, that is, with the following round of the research centre's Health and Demographic survey, conducted three times a year.

explained that his "soul was now at peace"; both parents had remembered incorrectly that negative results were the type that would be returned quickly.

In family F21 I was quizzed by a visiting uncle about his nephew's 'not SCD' result and the nature of the disorder. He went on to suggest a correction to the translation of the word used for 'inherited' in the KGBC information leaflet given out to participant families, indicating that the word used sounded more like 'congenital'. He asked if the child's 'negative' result meant that he could not go on to have affected children in future, whoever he married. I was not able to give the family this reassurance, given that I did not know whether the child was in fact a carrier or had normal haemoglobin.

3.3.3 Being given positive results

There was a marked difference in families' reactions to being given information about a positive SCD result depending on whether there was any knowledge of the disorder in the family at the time of screening, and whether or not the child had already begun to experience symptoms of the disorder. The paragraphs below summarise the initial and later reactions to being given information about SCD, including use or non use of the SCD clinic in three sections related to these features.

3.3.3.1 Three families with no family history of SCD and a completely well child/children

The reactions in these families to being told that their apparently completely well child had SCD differed. The strongest reaction occurred in family F07, where both parents described being very shocked and denied the result initially, the husband particularly expressing anger and blame towards the research centre. As explained earlier, the parents had felt pressurised to participate through their interpretation of a possible risk to future access to quality health care, including for SCD if this was found (3.3.1.2). The father was mistrustful of the researchers' motivation in testing their child, and in our first interview hinted strongly at his belief that the KGBC field worker may in fact have injected his daughter with something that caused her to develop SCD, in order that research could later be done on the child:

> "I thought they [researchers] gave my child some chemical which caused some reactions in the child's body so that the child becomes (pause), so that they can test their drugs to know if they drugs work, whether the drugs cure or not. So that thing makes you worry. Because when the child was born, the doctor didn't see or talk of any complications. My child was born at KDH and was given a clinic card and dates, and we attended clinic normally. We also thought when a child is born at a health facility, she is normally examined for any complications, and if there is the doctors explain that this child has this and that, and they didn't tell us that my child is suffering or might be suffering from this or that." (Father, F07)

The parents brought their child for confirmatory testing but did not accept the diagnosis and did not attend clinic again until the child began to develop symptoms after some months. Over time, the parents came to value the services offered at the SCD clinic to some extent, although the father remained concerned about his child's failure to completely recover, saying: "I take her there [clinic] to be cured but if there is no any cure, you find it useless."

The parents in family F12 reported that they had not been given any feedback on the initial SCD test, despite research records that the result had in fact been taken. The father had been at home at the time of the field worker visit and consented to participation. Their young baby who had participated in KGBC was thriving well, but they explained that they were aware of SCD since friends had lost a child through this disorder, and further, they had had health concerns in the past about an older child in their family. They had asked the KGBC field worker if the older child could be screened for SCD as well as the baby but this had not been agreed, given the study protocol. When we visited their home several months later, the parents had not sought any further advice about either child. As a local pastor, the father was opposed to any form of traditional healing. During our visit, their account and my observations made it clear that the older sister was very likely to have SCD. Both children were brought for screening at the district hospital the following day, when the condition was confirmed in both. The parents did not attend the SCD clinic again, but the father informed clinic staff that he intended to take the children to a private clinic.

In family F10, the mother had twin boys who were both recruited into KGBC and found to have SCD. She had agreed for the children to participate, and checked this decision with her husband

afterwards. Her reaction to these results was one of passive acceptance, as shown by her response to a direct question on how she felt when the results were explained:

> "What could I have felt? Because I was told they were sick and I must take them to clinic… then it didn't take long before my child started having swelling, the child by then was starting walking and he stopped I had to carry him and make him sit at one place and when the child became tired you carry him and make him sit in another place. That is the sickness…" (Mother, F10)

She brought her children for confirmatory testing straight away, as asked. However, over time she lost confidence in the type of care provided at the SCD clinic when her boys' condition did not return to normal; and turned to a traditional healer. This accorded with her underlying belief that the boys' condition had been caused by jealousy in a near family member or neighbour, prompted by her luck in having twin boys in her first pregnancy. For this reason, she and her husband had decided she should move back to her maternal home, with financial support, to avoid this perceived malign local influence. She fully intended to move home once the boys' condition had returned to normal. In addition, she explained the difficulty in travelling to the SCD clinic with her two boys when they became too large for her to carry both alone, since she could not afford the fare for public transport. As a result, she chose to sometimes use the SCD clinic and at others visit a traditional healer, causing conflict with her husband who did not approve of the latter.

3.3.3.2 One family with no family history of SCD and a child with relatively mild symptoms before screening in KGBC

In family F13, the mother had deferred to the father's decision about participation in KGBC. She did not express any particular reactions to the SCD test result. She did not attend SCC for confirmation, instead continuing to adopt an exclusively traditional explanation for and way of managing her child's illness, as she had been doing before SCD screening. Her accounts have contributed importantly to section 3.2.2.1 on supernatural beliefs. In this family, the brother-in-law refused to accept researchers into the household, given issues of mistrust, and the mother - while explaining that she had not had enough money to travel to the SCD clinic - refused to accept a fare provided by KGBC. We were unable to follow up on this family after the second visit.

3.3.3.3 Two families with an older child already attending SCD clinic

In family F09, the child had developed symptoms prior to recruitment into KGBC and given that at least one - and perhaps two - of his older siblings also had SCD, the father had been happy about the opportunity for the child to access a SCD test through participation. Given the strong family history, both parents had experienced considerable anxiety in relation to the condition. As has been described in section 3.2.3.1, there was also considerable tension between the parents over the inherited nature of the disorder and conflict over who was to blame for bringing SCD into the family, leading to stigmatisation of the mother and repeated requests for paternal testing. The mother also expressed resentment that her sister-in-law had prevented the KGBC field worker from explaining the result to her, since the father had not been around at that time, ostensibly because the mother could not read:

> "Even if you don't know how to read, can't you be explained to?...You shouldn't ignore someone and say I won't explain to her…you might find that her education is lower than yours but you can still understand depending on your intelligence." (Mother, F09)

The father in Family F09 reported marked difficulties in bringing both children to the clinic since their home was around 20 km away in a rural area of the district, and his normal means of transport a bicycle:

> "I have not missed attending the clinic. When it was just one child I used to cycle to the clinic with her but now they are two, so I can't cycle with them both. So, I walk half way up to X [place on main road to Kilifi] and board a public service vehicle there but I try not to miss the clinic." (Father, F09)

At the same time, he expressed misgiving about the clinic related to his disappointment at the lack of recovery shown by the children:

> "… they give us the same drugs in the same quantity each visit! They don't give different drugs and this disturbs me. If they could be reducing the quantity of drugs, then I could assume the SCD is getting less severe in the children…and that maybe there's a possibility of being told to stop going to the clinic. But they tell us to come back to the clinic again after 3 months and give us the same drugs." (Father, F09)

In family F11, the mother accepted the diagnosis of SCD in her child following participation in KGBC without much surprise, seemingly because she had an older child with the same condition

and the index child developed symptoms within a week of screening. However, it was very difficult to build a good rapport with the mother during these interviews and her account was not always consistent. The child's father was not available for interview. It was not clear who had consented for participation. The mother had been told that the condition was inherited at the SCD clinic but seemed unconvinced given the lack of family history on either the paternal or maternal side, instead repeatedly explaining that she saw the condition as 'a normal childhood illness'. She described puzzlement at why these particular two of her nine children should have been affected. Both of her children were very mildly affected. She tried to attend the SCD clinic regularly but was not always able to find the fares for transport. Asked about her husband's reaction to the children's condition, she replied: "When the problem starts he only says '*bado muende tena Kilifi'* ('you still have to keep going back to Kilifi [district hospital]'). The mother reported that her husband had once taken the older child to a traditional healer, but that she had refused to allow the child be taken again, preferring to use a government clinic.

3.3.3.4 Two families with a child known to have SCD before participation in KGBC

In family F06, the mother had been recruited into KGBC via the SCD clinic rather than at home. Her first child - an older sibling to the index child - had been very seriously affected by SCD, causing great distress. When the diagnosis was established in her first child, she had initially used both biomedical and traditional forms of treatment, as described in section 3.2.2, finally coming to believe that the former was more important. For her second child, she had requested screening at the SCD clinic before any symptoms appeared and believed that much earlier testing and biomedical care for her second child had led to her condition being less serious. She continued to take both children regularly to SCD clinic, made easier by the close proximity of her home to the hospital.

In family F08, the child had experienced severe symptoms and been hospitalised on several occasions before recruitment into KGBC. The mother had sufficient financial resources to attend

clinic regularly and had never visited a traditional healer. She decided to participate in KGBC even though she believed the aim of the study was to provide a community health check for SCD, and already knew that her child had this condition. Her decision, made on her own, seemed to be based on supporting the KGBC field worker who visited her home, who was also an elder within her church, although she explained her main reason as helping the research in case the records were somehow not clear.

> "By then [when the KGBC field worker visited] I had known about my child [having SCD], but …KEMRI [the research centre] had sent people to come to homes and collect samples for testing and carry out research, so he [the field worker] came here for research. I told him its okay but my child has already been tested... He said he was only taking for the sake of research, but he was worried about going back [to the research centre] and saying he had been told such and such, maybe he wouldn't be trusted… And that's why he decided to take the sample. Me, I understood that easily, because he also knew [that the child had been tested] since I showed him the papers from the ward. He checked everything. [Interviewer: Did you know the field worker before he came for your child's sample?] Ah, I know him because he lives there, he comes from this area. He is a person I have knowing [while] growing up. He is my elder, we used to go church together. He is a person I see all the time, and I have known him for a long time before. *But on that day he had come to work*." (Mother, F08) (my italics)

These parents supported one another without blame, and brought their child regularly to SCD. Sadly, the child died in a severe SCD crisis on the way to hospital before the completion of Phase Two of this study.

3.4 Moral concerns arising from community experiences of SCD and KGBC

In the following paragraphs, drawing on the analysis above, I outline the main moral concerns of community members and involved families with regard to SCD and the KGBC research project as shown by the findings in this chapter.

3.4.1 Balancing harms and benefits of information sharing on SCD in Kilifi

In the accounts families gave of their experiences of SCD and testing for this disorder in KGBC, important moral concerns are apparent. Most notable amongst these was their concern about the potential for harm to individual children and their families where the condition is unrecognised, with a particular focus on the relationship between harms for mothers and their children.

Perceptions of harms were based on the potential for unnecessary suffering, and higher risks of death, in children where low awareness of the biomedical nature of the condition led to the use of relatively ineffective (non biomedical) care. The emotional costs to the parents were related to these harms in affected children. Further, structural gendered influences mean that mothers are often at particular risk of being blamed as well as vulnerable to the consequences of family breakup, where this is an outcome. The direct and indirect economic costs to the family of taking care of the child's health were also prominent in these accounts, heightened where a series of relatively ineffective care seeking options were followed over time on a trial-and-error basis. Economic costs were also often gendered in nature, given mothers' roles in caring for the child. These harms seem to be amenable to the introduction of more effective care, through the reduction of physical symptoms in the child, suggesting strongly that sharing information on the biomedical nature of SCD, including during research, has an important moral dimension in addressing physical, economic and emotional harms to the child, her mother and - likely - to her siblings.

Although it is clear that some of these harms - most particularly those related to physical aspects of the condition - cannot be removed by better understanding of the condition alone, there are several ways in which information sharing in KGBC did have the potential to limit harms. Firstly, the data suggest that where screening and referral to services at the SCD clinic ameliorate physical harms for a child, there is likely to be a positive impact on economic and psychological harms for affected children and their families (see section 3.2.5). In addition, *early* diagnosis and care - including where screening is predictive rather than diagnostic, as in KGBC - may be particularly important given its role in improving survival in SCD (Williams, 2010). In relation to the potential benefits of predictive screening, families' stories of the distress and physical suffering over a period of time before a diagnosis was made suggest that early screening may have the benefit of *pre-empting* these harms of 'not knowing'. In any case, the finding that several children in KGBC had *already experienced symptoms* before recruitment *without diagnosis* shows that screening during research may act as a diagnostic rather than a predictive component. This is likely to be true in any setting

where there is low public awareness and government health priority setting around the condition. At the same time, my data illustrate potential harms in predictive SCD screening (e.g. family F07), highlighting the importance of communication about the study, and the nature of the underlying relationship between the community members and researchers.

These accounts have also begun to show that structural gendered influences in some families can mean that in some families sharing genetic information can also give rise to increased blame being directed towards mothers, so that information sharing on the genetic nature of SCD should be seen as a balance between addressing the harms of 'not knowing' about SCD and the risks of generating additional harms in some families when parents have an increased understanding of the inherited nature of the condition. In other words, it is not clear if explaining the roles of both parents in SCD would be likely to make the lives of mothers of children with SCD (and by extension their whole family unit) easier or more difficult. Further, there is a risk that, as a result of this information, fathers may make requests for paternal testing (for example, Family F09).

A further challenge to the expected benefits of information sharing is the low uptake of biomedical services at the SCD clinic. The experiences described above show that reasons for deciding not to seek help from the SCD clinic were importantly influenced by parents' beliefs and perceptions. These included conflicting beliefs about SCD (and illness in general) that led to a preference for traditional forms of treatment[63]; a perceived lack of response of symptoms to biomedical treatment, where there was an expectation of cure, or at least a more obvious improvement; and issues of trust with researchers and the research institution. Other reasons for not using the clinic were the sometimes marked financial and economic challenges experienced by some in accessing the clinic and in waiting in (sometimes) long queues; and where symptoms were perceived as too minor for this investment. Both of these latter apparently practical challenges were also related to parents' understanding of the disorder. For example, mothers who felt that their child was most likely to be suffering from a supernaturally caused disorder would be least likely to seek resources to travel to

[63] I also note that some families talked about nearness to local traditional healers in a way that suggested cultural proximity was linked to geographic nearness in a way that supported used of the local.

Kilifi town, or wait in a queue. Effective communication, and the development of relationships supportive to communication, therefore seems to be central in promoting use of biomedical care.

Overall, the findings identify potential benefits, harms and challenges for SCD information sharing beyond those recognised by researchers at the outset of KGBC. The complexities in understanding how to balance these harms and benefits illustrate recent theories described in Chapter 1 (section 1.6) around the social meanings and impacts of genetic knowledge[64], including the multiple influences on ways in which knowledge may impact families and families interpret genetic information (Chilibeck, Lock et al. 2011; Geelen, Van Hoyweghen et al. 2011; Horstman and Finkler 2011). In line with recognition in this literature of mediators from political culture, the existence of a prominent structural form of gender inequity in the Kilifi setting seems to present particular risks to mothers, while individual influences remain as important here as they have been described in other situations.

As described earlier, the overall plan for the thesis[65] was based on the identification of important moral concerns in a first stage of research (Phase One) that would be taken forwards to a second stage (Phase Two), where the latter was built upon a detailed process of informed deliberation by varied groups of community members. Given the prominence of concerns about harms and benefits, questions about the importance of sharing SCD information during research were identified as the most important unresolved issue emerging from Phase One. Two main questions taken forwards included the specific nature of information that should be shared for *children affected by the disorder* and for *carriers*, respectively, including the contingencies this process might imply, and the ethical arguments behind the approach to information sharing. A third question is about the potential risk that sharing genetic information might lead to *requests for paternal testing* (as for father F09), and the nature of any obligations that this might raise for researchers. These questions are summarised in Figure 3.1 (section 3.5).

[64] See Chapter 1, section 1.6
[65] See methods in Chapter 2

3.4.2 The distraction of SCD screening in diagnostic interpretations of KGBC

Perhaps the most clear potential moral concern about sharing information on SCD in KGBC tracks an issue recognised in the Georgetown workshop on researchers' responsibilities for ancillary care (Georgetown University Workshop 2008), that is, the risk that participants will see the study as being primarily or entirely a community SCD check. As discussed in Chapter 1, such a perception risks undermining voluntary informed participation in research, an important feature of ethical research practice from the perspective of autonomy and respect for persons. An interesting feature of the field workers' interpretation of this phenomenon in KGBC was that in some cases the issue they noted was 'low interest in' rather than 'no understanding of' the main scientific question in this study, and greater interest in screening for SCD. Some participants were said to have been more interested in the directly perceived benefit of SCD screening for the participant family than the theoretical future development of 'drugs and vaccines' that could be useful to others. Greater interest was also seen to lead to participants being more likely to remember this aspect of the study.

Questions about the importance of diagnostic interpretations of research, or distractions from research, acting as an important counterbalance to the proposed value of sharing SCD findings, were taken forwards as an important theme in this thesis through the analysis of findings in Phase Two[66].

3.4.3 Participant-researcher relations: Trust and power

It was clear in my interviews that the relationship between researchers and participants, including the existence and level of trust established between KGBC participants and field workers as well as between participants and the research institution as a whole, played a key role in determining the way that moral concerns arose.

The findings show that trust played an important role in families' decisions about participation, leading largely from a lack of clarity about the purpose of research in general, as described in

[66] See Chapter 7, section 7.2.2.

section 3.3.1.1. Conflation between research and health care led to doubts about researchers' motives in taking 'tests' from healthy children that were also linked to supernatural interpretations of blood use. The father in family F12's action to check up on KGBC field workers by comparing his experiences of recruitment with a next-door neighbour provides a good illustration of mistrust. In relation to researchers' motives behind the study, the reassurance gained by many community members over the first few years of the study was often linked to the feedback of findings on SCD status. The increasing confidence in KGBC and the research centre over time made field workers' jobs of recruitment seem easier but seem likely to have been linked to diagnostic interpretations of KGBC. In other words, the low levels of trust that were reportedly encountered in some cases at the start of the study and the much higher levels that seemed more prevalent later might both have influenced decisions about participation in ways that have been described as inappropriate on the basis of understanding and voluntariness.

Perceptions of the research centre as a high quality hospital influenced some parents' decisions to participate, in an apparent hope of leveraging better access to care or fear of losing it. The role of the relationship between research participants and the institution is illustrated in this case, where an imbalance of power, in terms of access to medical resources is the fundamental influence. Family F07 found this a particularly strong influence given that the disorder being screened for would need the type of lifelong services that only the research centre seemed likely to be able to provide.

In contrast, the role of trust at an interpersonal level found a striking example in family F08, where a longstanding relationship between the mother and KGBC field worker seems very likely to have influenced her decision to participate in order to help the fieldworker in his job, even though she saw no direct benefits for herself, her child or family.

Exploration of the theme of trust in researcher-participant relations and its influence on researchers' responsibilities to share information on SCD and carrier status have been continued throughout this thesis, being explored during the analysis of Phase Two findings and in the conclusions in Chapter 7.

3.4.4 Not sharing information on carrier status: A justifiable 'deception'?

In my interviews for Phase One, low awareness of SCD and its inheritance meant that no families spontaneously raised concerns in relation to sharing carrier findings in KGBC[67]. It seems likely, in the absence of knowing of any other possible outcome of screening, that a result showing 'not SCD' would be interpreted as 'normal'. This would fit with participants' common reaction of relief on being given this feedback.

At the same time, I noted an important incident during my visit to family F21 in section 3.3.2, where a visiting uncle asked questions about future reproductive risks of his participant nephew. Two important implications seem to emerge. The most important is that this request threw the nature of the carrier disclosure policy - as a form of deception - into sharp relief. Clearly, as explained above, such a 'deception' was intended to balance the needs of the community for information against the potential harms assessed for sharing information on carriers given current SCD services and resources, and the costs this would imply for the research overall. A second implication of the reaction of the uncle in family F21 is that the needs of different community members are likely to vary, implying that policy on information sharing should take account of these differences.

As part of the questions on sharing information on SCD status, an important issue emerging from Phase One therefore concerns whether or not researchers should share information on carrier status with families of children screened during KGBC, and if so, how such information should be shared, and what the ethical basis of such a recommendation would be.

[67] In Chapter 1 section 1.4, I describe the change in research policy on disclosure in KGBC, prompted by public pressure. At the outset, only findings for affected children were actively disclosed. Later results were taken to all participants, but no distinction made between carrier and normal haemoglobin. For researchers, perceived limitations on disclosing carrier findings were low community awareness of the disorder, a lack of public access to SCD testing, the lack of a coordinated national or provincial health policy on managing the disorder, and the high resources that would be needed to share the information and ensure validation through re-testing, particularly given that around 15-20% of the children screened were expected to be carriers for this condition.

3.4.5 Accounting for community views on morally important issues: Important gaps and the need for further research

There are several potential ethical concerns and questions raised by researchers in Kilifi that are not directly addressed by the findings in this chapter, and which to some extent mirror the wider literature for genomic epidemiological studies (Collins 1999; Greely 2007; Parker, Bull et al. 2009). The concerns and issues raised in this chapter are *those that emerged as important from researcher and community perspectives* during discussions in Kilifi. Critical reflection on these findings, in the light of my analysis of the literature, for example, suggest three obvious gaps in the issues raised by families and community members: the question of whether carrier information should be shared; whether samples should be archived for future research with the current governance mechanisms in place; and whether or not genetic or genomics research raises particular issues about consent from both parents, where issues of their 'ownership' of the findings may arise as a result of relatedness. As already discussed, the first question about sharing carrier information is embedded in that about disclosure of SCD, and will be taken forwards into Phase Two. The second and third questions will not be addressed in this study for reasons of scope and emphasis, given the aim of enabling sufficiently detailed community-led normative reflection around the most important emerging issues from Phase One[68].

Whilst unsurprising, the existence of gaps in an account of moral concerns stemming from experience of participation in research is illustrative of a more general challenge. A focus on community perspectives means that some ethically important areas that are not clearly understood or articulated within the community are likely to be underrepresented. These gaps act to highlight the importance of developing approaches to community deliberation that allow unrecognised issues to be introduced and explored in a meaningful way. The methods used in Phase Two seem to

[68] This study does offers some insights into questions of parental consent in a genetic study, suggesting that parents' responses in Kilifi were more related to the normal dynamic around decision making within families than the genetic nature of this particular study, much as suggested by the 'post genomics' literature in section 1.6 in Chapter 1.

represents one way in which this might be achieved, as will be discussed further in Chapter 8, section 8.3.

3.5 Conclusions

In this chapter, I report on findings from Phase One of this study, with the aim of exploring direct and indirect moral concerns related to participation in a genomic epidemiological study in Kilifi, including issues around SCD screening and information sharing. The findings give a rich description of experiences and perceptions around the study, and around having a child affected by SCD.

From these findings, I have identified three central, morally important questions to take forwards for deliberation by community members in Phase Two, as described in Chapter 2. These questions concern the way that researchers should act in relation to information generated on SCD during research, including for *affected children*, for children found to be *carriers* and in response to requests for *paternal testing*, as described in Figure 3.1 below.

Developing responses to these questions are important because each seems to present a moral dilemma, and all are central to planning studies in which SCD status of participant children will become known. As described in Chapter 1, these questions are likely to apply to a wide range of studies conducted in malaria endemic settings; and to have increasing prominence against the background of the current epidemiological transition in health burdens towards non communicable diseases. At the same time, other morally important issues in research of this nature that are less visible to community members have not been identified through this phase of research. I have suggested these would require a more actively directed approach, as might be offered by the methods in Phase Two of this study.

From the findings in this chapter, the elements of the dilemma in sharing SCD information during research that require further exploration and deliberation are:

- Complexity in balancing the possible benefits and harms of sharing SCD information in research, but an important potential for better understanding to limit serious physical, emotional and economic harms associated with low awareness of the disorder;
- The influence of structural and individual influences on the relationship between genetic knowledge, family dynamics and the outcome of information sharing, including the role of supernatural health beliefs and gendered blaming;
- The influence of the exact nature of SCD information shared, including the concern that explaining parents' individual genetic roles may lead to requests for paternal testing;
- The potential for SCD information sharing to lead to diagnostic interpretations of research, or reduce interest in the main aim of the study in a way that may undermine informed consent;
- The implication of a form of 'deception' where explicit information sharing on carrier status is avoided;
- The role of trust and power within researcher-participants relations in influencing decisions about participation, and therefore in framing responsibilities for researchers.

The questions identified in this chapter to take forwards to Phase Two are summarised in Figure 3.1. The following Chapters 4, 5 and 6 will separately report on community deliberation around each of these questions. In addition to informing the process of normative analysis begun in this chapter, the findings provide important information to support planning for counselling and communication strategies for the disorder, either in research or health service delivery.

Figure 3.1. A summary of questions from Phase One taken forwards to Phase Two

Chapters 4 and 5: Should researchers feedback results on SCD status for affected children and carriers (respectively) when screening is conducted as part of research? If so, what information should they share? What ethically relevant concerns are at stake, how might these support or be balanced against each other, and what conclusions emerge? How could information sharing activities be seen as the responsibility of researchers? Therefore, in what way should the value of information sharing be seen and justified, and what contingencies and practicalities are raised?

Chapter 6: Where a child has been shown to have SCD, should researchers agree to undertake paternal testing if requested? If so, and if they find a result showing misaligned paternity, should they share this information with either or both of the parents? What ethically relevant concerns are at stake, how might these support or be balanced against each other, and what conclusions emerge? How could information sharing activities be seen as the responsibility of researchers? Therefore, in what way should the value of information sharing be seen and justified, and what contingencies and practicalities are raised?

CHAPTER 4

Phase Two. Discussing information sharing for study findings on sickle cell disorder: Families of affected children

4.1 Introduction: Background, aims and organisation of chapter

This chapter is the first of three linked chapters presenting the findings from discussions with community members in Kilifi as part of Phase Two of this study. The three chapters follow through key questions emerging from Phase One of the research, which explored the moral concerns of community members related to participation in a genomics study in this setting, as described in Chapter 3.

The discussions reported in this chapter address the first of these questions, and concern the importance of disclosing SCD findings where a young child screened during research like the Kilifi Genetics Birth Cohort study (KGBC) has been found to be directly affected by the condition[69]. The discussions reported include the informed views[70] of a range of community members[71], including KEMRI community representatives (KCRs), community leaders, research programme staff, health managers and mothers of children with SCD. The questions for discussion were framed as follows:

Should researchers feedback results from SCD screening in children when conducted as part of research? What ethically relevant concerns are at stake, how might these support or be balanced against each other, and what conclusions emerge? How could information sharing activities be seen as the responsibility of researchers? Therefore, in what way should the value of information sharing be seen and justified, and what contingencies and practicalities are raised?

Section 4.2 provides an account of community members' experiences, arguments and values raised during discussions on the questions above. The section initially focuses on reasons in support of sharing SCD information, followed by issues raised against information sharing, and important emerging contextual issues and values. In section 4.3, I explore in more detail the nature of agreement and divergence in community opinions, based largely on identifying emerging 'facts' and values as an organising concept (Macklin 1999), including the way these are prioritised by different community members, as discussed in Chapter 2 (section 2.4.5). The final section (4.4) presents the conclusions of the chapter, including summary Figure 4.3.

[69] The chapter does not include views on sharing information *on carrier status* for this disorder; these views will be described in Chapter 5.
[70] Community members participated in a series of individual or group discussions, each preceded by information-sharing activities to establish common understanding of the nature of SCD, including its inheritance (see Chapter 2). The question guide, including the scenario used, and a detailed description of the 62 community members who participated in the discussions are given in Annexes C and D.
[71] My use of the term 'community member' is equivalent to 'local resident' of Kilifi district, and includes participants with varying levels of technical knowledge and experience, but who were born and/or have spent long periods of their lives in Kilifi district. All would be eligible for participation in studies at the KEMRI Wellcome Trust Programme in Kilifi.

4.2 Local experiences, arguments and values

4.2.1 Reasons for sharing SCD information: Limiting the 'harms of not knowing'

In Chapter 3, I described low levels of awareness of the nature of SCD within the community in Kilifi, and the extreme distress that many families of affected children experienced in struggling to understand what was happening to their child and how best to help them. A resulting set of 'harms of not knowing' about the biomedical nature of SCD were described in that chapter, including: the exacerbation of ill health and distress for the affected child caused by using relatively ineffective treatment; anxiety and economic costs to the family associated with looking after the child, including care seeking; and forms of blame and conflict within families, particularly affecting mothers and sometimes others accused of 'witchcraft'.

In discussions with community members, the main reasons for sharing SCD information with the parents of affected children concerned the potential to limit these 'harms of not knowing'. In the following I describe the ways in which SCD information sharing was perceived to limit harms, and the types of information thought important to achieve this aim.

4.2.1.1 Increasing the likelihood of effective care for affected children, their families and the wider community

Accessing effective care for children with SCD was seen as a paramount consideration across all groups of community members, given the severity of the health effects for children and its potential for impact on the family. Further, it was widely felt amongst all community members that understanding the nature of SCD would be fundamental to people perceiving the value of and therefore using the types of care seen as most effective, that is, biomedical management. Two elements of understanding were considered necessary to support the use of biomedical care. These were the chronic nature and the cause of the condition. The latter was also seen as critical to

counter commonly existing beliefs and fears that unknown illnesses could be caused by supernatural means[72].

Understanding SCD's lifelong nature was seen as important because of a reportedly common community perception that effective biomedical care should be curative. As shown in Chapter 3, this view of biomedical care potentially undermines perceptions of the effectiveness of care offered in SCD clinic, particularly in the long term. The relationship between understanding the nature of the disorder and using biomedical care is well illustrated by a community member, talking directly about her own experiences of having a child diagnosed with SCD:

> "So the doctor will explain to them…they will easily be able to take the child to the hospital. But if the doctor does not explain to them…in the end they might feel (clicked tongue) 'what are we trying to do, and the child is not getting better? The doctor did not tell us anything except always keep going [to clinic]'. Therefore if they don't know, okay they will continue but they will stop going in the end. They won't come to the hospital again, instead they will do other things [alternative treatments] while the child continues suffering." (KCR02/P2)

In addition, better understanding and care were seen as forms of knowledge that could be passed on to other people within the family or nearby community that might also be affected by the disorder, as illustrated by the following two comments by field workers:

> "Maybe if you give them that information, explain to them, you will be putting them in a position where they know well that in case they have another [affected] child, they will know how to handle that child and deal with the sickness" (IDI04/P1)

> "He [the informed parent] will be I think a good ambassador for us, because if he knows the information, one day, one time, he will share that information with somebody else…[and] that other person will also share that information with somebody else" (IDI04/P2)

4.2.1.2 Countering harmful beliefs about supernatural causes of SCD

The second major, and related, set of reasons why people thought that information on the inherited nature of SCD should be shared with families concerned the commonness of supernatural understandings of health (and other aspects of life's troubles) within the community, confirming

[72] See also section 4.2.1.2.

the perceptions and experiences described by families affected by the condition, as in Chapter 3. During discussions with community members, the importance of taking these issues seriously was repeatedly underlined, illustrated by a comment that fears of unknown illnesses were 'very great in this community' (IDI04). The importance of countering supernatural beliefs for SCD was described as: convincing affected families to use more effective (that is, biomedical) care; and countering harmful effects arising from practices of accusation by traditional healers.

Efforts to convince people to use biomedical rather than traditional sources of care were seen as needing to provide substantial and convincing alternative explanations. The inherited nature of SCD was described as key, based on a view that the notion of causation was fundamental to local understandings of health and illness. Having indicated inheritance as the cause, an explanation of how transmission within the family had occurred would inevitably be needed. As a staff member who ran the SCD clinic pointed out:

> "You can say… okay, it's a condition a child is born with… from where? From who? So in the process of elaborating, you have to mention that it's a condition that the child is born with, but this condition is caused by who?...People will ask 'what kind of a disease [is this] that needs particular attention and we don't know where it's come from?'" (IDI03/P1)

A community representative, who had a daughter with SCD, gave a good illustration of the way that knowledge on the cause of SCD may interact positively with fears about supernatural causes of ill health:

> "When I went to the doctor at KEMRI, Kilifi, the child was tested and in the end both of us were called, my husband and I, and we were told that there was a condition from the father and me that caused the child to get that thing. So when we came back home, other people were saying that it was witchcraft, while others said that…there is someone who is against you, who did this and that to you. But, the two of us, we knew because the doctor had explained to us, and so we were not worried. So when the parents get information, it removes the fear…" (KCR02P2)

4.2.1.3 Addressing gendered blaming

In Chapter 3, I described community experiences and perceptions about the risk of stigmatisation of mothers of children with unexplained health problems. The forms of gendered blaming were

described in detail. Independently, the opinions of community members in Phase Two also pointed to the likelihood of gendered blaming occurring in SCD, underpinned by the phenomenon of paternal denial. For this reason, many community members felt it was very important that affected parents understand the inherited nature of SCD, and particularly the genetic roles of both parents and both sides of the family, over many generations. In other words, gendered blaming and related forms of stigmatisation were seen as amenable to better understanding of the certainty of the father's contribution to the disorder.

4.2.1.4 Removing fears about HIV/AIDS

A further harm perceived by community members as responsive to information sharing on SCD was confusion between the conditions of SCD and HIV/AIDS. Illustrating the reality of this concern, a mother (Family F01) in the first phase of this study had described being driven by her neighbours' rumours to have her children with SCD tested for HIV/AIDS. This was often seen as an important risk, as illustrated by a community representative saying:

> "When they understand [about the nature of SCD], they can be able to remove that fear from their hearts...because some may misunderstand it, they take it [that] this is a disease like HIV/AIDS" (KCR01/P6)

4.2.2 Reasons against SCD information sharing: The risk of generating harm

So far, I have explored the arguments put forwards by the majority of community members in favour of sharing information about SCD. At the same time, others recognised that sharing some areas of information about SCD had the potential to generate harm in some cases, giving rise to debate on the probability of overall benefit or harm in different situations. In the following, I describe community members' *concerns* about the potential for harm through information sharing, and the way these were often countered by proposed benefits in discussions.

4.2.2.1 Generating anxieties about inherited conditions: Hopelessness and reproductive risks

i) General worries about inherited conditions

Some community members argued that better understanding of a condition as inherited would increase anxieties for parents through understanding the *lifelong* nature of the condition. As a community facilitator pointed out:

> "So I think it [sharing information] is important though I have said it's sensitive because some there are thinking, once you are told that, then there is that feeling that we are all sick, because it is inherited, and so we are all sick, we are all going to have sick children, you know, there is that traumatising effect the parent might get from the information" (IDI09P1).

In contrast, others felt that general anxieties about the inherited nature of SCD were not important risks given that some level of worry - likely of a much more severe form - would already exist about the cause and effects of the child's condition, especially where lack of an explanation led to fears about a supernatural cause. Further, it was perceived that explanations of inheritance could be reassuring, as the following views of a community representative illustrate:

> "…if they are not told that this condition runs in the clan, they can be worried, but if they are told that this condition could be inherited from your clan, your grandparents, then the parents won't be worried that their child might become seriously weak/incapacitated…but will be going on with life normally because he inherited it from the clan" (KCR03/P2)

Drawing from the same point, many community members acknowledged that the concept of inheritance was already well understood in the community, and as a consequence would not be 'technically' difficult to introduce for SCD. Illustrating this perception, families in Phase One readily volunteered opinions that illnesses such as epilepsy, diabetes, asthma and leprosy were inherited through families (Marsh, Kamuya et al. 2011b).

Given the more important reasons for explaining the inherited nature of SCD, community members generally saw that some level of worry about the diagnosis was inevitable and should be tolerated. Further, some level of anxiety was seen as positive in supporting regular and long term use of biomedical care. At the same time, all community members talked about the importance of providing adequate counselling support, including to both parents, to address these types of general anxieties and to ensure that a feeling of hopelessness was not engendered, where the latter might be particularly detrimental to the relationship between the child and her family.

ii) Explaining parents' future reproductive risks: Potential for family breakup

Some community members described an important specific concern that explaining parents' individual roles in the inheritance of SCD in their child would lead to recognition of their own future reproductive risks, and the possibility of family breakup. Views on the probability of this outcome were varied. Three mothers of one or more affected children perceived the risk as important, and greater than might arise, for example, from a father's suspicions about paternity. Further illustration comes from a community representative who explained:

> "Okay now, for me, I can say that there will be many separations or that divorce, family misunderstandings, and this will be brought about mostly because maybe they have gone there [to the hospital], and this is their first child, when it's explained to them in detail the way this is inherited, this problem, they can decide we will not get others like those ones, so they can separate *because they will not want to get another child who will be having a problem like that*." (KCR01/P2)(my italics)

This view is supported by findings from Phase One that the *experience* of having several affected children, and the knowledge that other children might be similarly affected in the future, importantly increased anxieties for parents and risks of blaming towards mothers.

In contrast, other community members perceived that sharing information on parents' reproductive risks would not increase the probability of family breakup given that parents may go on to experience having more than one affected child *whether or not they understand the reason* for this. The descriptions of participants in Phase One of mothers of children with SCD being blamed for their child's condition in the absence of understanding its genetic basis seems to support this view. Understanding the fact and nature of future reproductive risks was therefore often seen as a way in which some harmful misconceptions could be addressed. It could also help parents to plan and be more prepared for this eventuality in future, including knowing how to take care of an affected child.

Across these discussions, community members perceived that contextual factors would be an important influence on the risks of family breakup from understanding parental reproductive risks in future. Further, some staff with experience of explaining genetic risks saw separation as unlikely because of the lack of certainty implicit in a concept of chance. In this way, a statistical probability

(1 in 4) of having another affected child was not thought likely to be very meaningful to many people. More fundamentally, common attitudes of fatalism and faith based forms of determinism within the community were seen to counter anxieties about future reproductive risk rates. As a field worker explained:

> "So you'll find that explaining that to them like that they say well as long as it's a chance, you may never know, you'll just give birth but whoever, we'll get." (IDI06/P2)

A further factor pointed out by some field workers militating against separation was that understanding the relative commonness of SCD and the lack of public testing would deter fathers' from taking the risk of marrying another woman who may also turn out to be a carrier.

For a few community members (two field workers and a male community representative, KCR01/P5 and IDI07), family breakup in this situation was seen as opportunity rather than a harm. The possibility of 'starting again' with a new partner, and building a new healthier family, was described as an important alternative to continuing to have children affected by this serious disorder in future:

> "For me, I think it's better if this is explained to them, because even if they are divorced, wherever the other one [the woman] goes she can explain, and if the other one [the man] marries another one here, he will explain to her...we divorced because of this and this, of this and this, now you go and be checked" (KCR1/P5)

> "And one more thing if the worst comes to the worst and the family breaks up, of course let's take an example if they have only maybe one child, if this man decides to marry another wife maybe he can get one who is not a carrier, and the lady can also get a man who is not a carrier, and they start very fresh lives." (IDI07/P4)

4.2.2.2 Generating or strengthening gendered blame, including for misattributed paternity

Some community members pointed out that where SCD had not previously been understood as coming 'from the family line', sharing genetic information would now make this clear. At the same time, the understanding that people can transmit the condition without experiencing any effects themselves - that is, through being healthy carriers - was seen as very counterintuitive. In other words, fathers who themselves felt well would reportedly find it difficult to accept that they were carriers and had passed on the disorder to a child. In contrast a mother's evident biological and less

powerful role in the family would not place her in a similar position to deny being a carrier. The invisibility of carrier status could lead to or support a father's denial of his genetic role.

Two possible interpretations were then thought to be open to the father; to believe or claim that the condition had in fact been passed down from the mother and her family only, or agree that there was another carrier parent, but argue that this was not him[73]. In this way, the risk of raising concerns about misattributed paternity arose from recognition of the same cultural influences of gendered blaming and jealousy that other community members saw as the reason that the information should be shared (section 4.2.1.3). Seeing two 'sides to this coin', several community members (some strongly) believed that relating inherited conditions to the roles of individual parents would become a highly sensitive issue. A preference in this case was for information on the genetic nature to be very general, so that earlier generations and family lines were implicated without identifying a specific locus of responsibility in individual parents. This point of view was made strongly by a community facilitator:

> "I don't think giving good information entails telling them that this has been inherited from the mother and the father, because… I sort of have two types of inheritance in the Mijikenda context. There are those things that you inherit…what you would say is 'inborn', you are born with that thing by virtue of its existing in your family line. Which is different from inheriting from your father and mother, as in its so immediate, so you get it from either or from both of them. That type of inheritance is very sensitive. I think the kind of inheritance that you talk about like someone inherited epilepsy from the family because the forefather had epilepsy or was born handicapped because the forefather was handicapped, that kind of inheritance is quite acceptable and you know there is no blame towards anybody because it's kind of devolved, if you see what I mean, it's kind of, it doesn't have a permanent identity. Whereas this genetic inheritance is one thing that is attributable to the immediate parents, which now the issue of paternity can easily come out." (IDI09/P4)

Similarly a chief pointed out:

> "You know in Mijikenda families, they are extended families, so I think in my opinion we should not say that the disease is coming from the couple alone, it can have come from the extended family, not really the couple themselves, so I think that will minimize the questions and problems as to who will have been the carrier of that." (Chiefs 01/P6)

For the majority of community members who supported explaining the parents' genetic roles, misconceptions of this nature were seen as amenable to more effective communication, particularly

[73] In this thesis I will refer to this situation as misattributed paternity

on the phenomenon of healthy carrier status. An important conclusion of this debate was, therefore, that it was essential that information sharing should include the concept of healthy carrier status, using easily understood language. Conversely, failure to offer an explanation for why fathers could be responsible without experiencing symptoms was seen as unlikely to counter the tendency for maternal blame and consequent problems for the family, including the possibility of break up.

A minority of community members continued to feel that sharing information on the parent's genetic roles carried too much risk of generating concerns about misattributed paternity. This position was supported by recognition that fully understanding genetic roles, including healthy carrier status, would not minimise accusations of unfaithfulness directed at the mother. Instead, doubts about misattributed paternity were linked to understanding the genetic roles of parents well enough to see that another man, outside the family, could contribute his genes in exactly this way. At an extreme, it was considered possible that fathers with these doubts would request testing themselves in order to demonstrate whether or not they were responsible for the condition[74]:

> "So when the child is found with that condition, that's where there is a conflict because this father will want to go for a test there. So if the child is found with [SCD] while I am fine, the child is with [SCD], [the father will ask] where does this come from?" (KCR02/P3)

The possibility that researchers may find themselves testing fathers and showing misattributed paternity in this way is very far removed from the original goals envisaged for information sharing on the inherited basis of SCD, and certainly unintended. There were mixed views on the likelihood of fathers asking for tests, some seeing this as likely - including expressing a view that they themselves would ask for a test in this situation - while others saw this as unlikely, as long as parents had understood inheritance from both parents and the concept of healthy carrier status. As a chief explained:

> "Yes, but I want to add a point. Also we have seen that as a carrier you only have one gene, of which it can't affect your health, you can be surviving very well and yet you have that gene of

[74] This situation arose in family F09 during KGBC as described in Chapter 3 (section 3.2.5).

SCD. So I think with giving him such an information, I don't think he will not understand on how that disease came." (Chiefs 01/P2)

Overall, in a situation where risks of gendered blaming were perceived as existing with or without information sharing on the parent's genetic roles, *most community members reasoned that risks were greater in the absence than in the presence of knowledge*. This view was supported by the four women with children affected by SCD (one community representative and three community members) on the basis of high risks of paternal denial of responsibility. Three of these mothers were in fact unconvinced that describing this role could exacerbate the risk at all. As a community representative also described:

"Because even if you don't tell them, if they are going to divorce they will divorce, without telling them they can divorce, [for example thinking] 'this woman is evil, every time she gives birth it's a sick child, I don't know what is in their family, may be they [family] have evil spirits, so no! You go to your home, you can even take the child, I don't need the child!'But...if they will have been told, they will understand." (KCR01/P5)

4.2.3 The influence of context on potential outcomes of information sharing

Over the course of the discussions described above, it became apparent that those who took part saw the nature and likelihood of benefits and harms of SCD sharing as highly dependent on pre-existing individual circumstances. The types of influences seen as relevant are described in the following paragraphs.

4.2.3.1 Individuals and family relationships

Against a background of strong patrilineal cultural influences in this community, community members agreed that there is considerable variation within families in the way that individuals relate to each other. They felt that this would be a very important influence on the outcome of family disputes. The most commonly drawn upon and important positive concepts were those of 'trust' and 'emotional commitment', with jealousy running counter to these more positive characteristics. In relation to risks of maternal blame, the likelihood of fathers accepting a genetic role for both parents was described as depending on whether these ideas were being introduced into

families where there were already emotional bonds and where both parents trusted each other. Where these conditions existed, paternal denial, gendered blaming and separation were much less likely.

Several community members also discussed the possibility that fathers, faced with knowledge of individual genetic risks, might take a second wife into an established household rather than consider separation from his first wife, assuming he had the financial resources to do this. Again, the emotional bonds within an established family would be an important reason to choose this option. This would be a widely accepted option for families following Islamic and other faiths that allow polygamy, but in practice also occurred in other families[75].

4.2.3.2 Observable patterns of sickle cell disease within families

Community members reported a common response to recognition of an unknown illness would be to scrutinise extended families on both sides over several generations to see whether any patterns could be established that might suggest the origins of the problem. During discussions in Phase Two, I explored community perceptions of the likely influence of these observations of illness patterns on the outcomes of SCD information sharing. There was little agreement on the implications of particular patterns. For example, some community members believed that explaining reproductive risks to young couples with one affected child might increase the risk of family breakup, given that no other children would be involved. Others felt that parents with one affected child would not take a theoretical future reproductive risk very seriously. It seems likely that the patterns themselves may be less important than other contextual features, particularly the attitudes of individual parents and the existence of trust, as described in the previous section.

[75] See for example, family F03, where both parents were practising Catholics

4.2.3.3 Economic circumstances

In Chapter 3, community members' experiences illustrated the potential influence of a family's economic circumstances on risks of separation when a child was affected by SCD, in a setting where poverty was widespread. In that chapter, the effects of poverty in increasing risks were seen to work through an impact on health and an inability to offset the costs of caring for a child with a chronic disorder, contributing to increasing conflict between already anxious parents.

In Phase Two, I explored perceptions of the importance of a family's economic circumstances on the risks of information sharing on SCD. In this case, the independent economic situation of the mother and the related power dynamics within the parent's relationship were seen as key. For example, chiefs described the way that mothers who were able to bring an income into the family were more likely to be able to influence decision making, and less likely to be vulnerable to blaming by their husbands. Women in the urban group of community representatives, particularly individuals with high levels of education, talked positively of their ability to sort out their own problems if separation from their partners were to become inevitable. Other community members believed that, for both genders, possessing economic resources would mean that their partner would be less likely to seek separation as a solution where carrier status was understood. In this way, mothers without an independent livelihood were seen as particularly vulnerable to economic and related health harms from both sharing and not sharing information; this circumstance was seen as constituting a risk in either circumstance. Illustratively, a member of the DHMT spoke of this as a 'pathetic situation'. Throughout these discussions, increasing educational exposure in mothers was seen as importantly reducing their vulnerability to blame and the harms of breakup. Mothers' educational status was also repeatedly described as a marker of development and a counter to harmful traditional attitudes. For example, a community facilitator explained:

> "For one, I think we are moving from the older days…I think to be sincere more people have gone to school, they can understand even the genetic part of this information as opposed to long time ago, and I think people who blame maybe the wife for the genetic makeup - though there's still a gap - but I don't think that's so big" (IDI09/P1).

4.2.4 Emerging values

During the course of these discussions, the reasons given for and against sharing information on SCD illustrated the importance of three main types of values. In community member's discussions of and enthusiasm for disclosure of information, a concept of openness was repeatedly drawn upon. The main recognised benefit of information sharing was seen to be the potential to limit harms for vulnerable children and their families, reflecting a further very important value. These two main themes - openness and protection of families - often constituted the primary form of balancing within discussions on the importance of sharing SCD information. They also tended to underpin perceptions about researchers' responsibilities to share information. Exploration of these three values, and their interrelation, will be continued across the next two discussion-based chapters.

4.2.4.1 Protecting vulnerable children and family stability

The interests of vulnerable children, their mothers and families were paramount in nearly all community members' considerations of the importance of sharing SCD information on affected children. A theme that emerged constantly across all conversations was the alignment of the interests of children and other family members, reflecting a value placed on family stability and harmony. This value will be discussed more in the following chapters, but is raised here to highlight a form of harm, raised by community members, that is centred beyond the individual.

The concept of 'family' is complex. Lindemann and Lindemann (1995) describe two broad meanings: the household or 'aggregate or group of actual members who are closely associated by living arrangement or by commitment, for better or worse'; and the family in abstract, including 'the family line...whose boundaries extend over space and time' (Lindemann Nelson and Lindemann Nelson 1995)(p35). The latter was frequently used by community members in Kilifi in considering the possible source of SCD in a child. In contrast, family stability - and the need for its protection - often referenced structural economic arrangements focused on the parents, affected

child, siblings and other dependent family members. At the same time, community members described intrinsic values of the family as well as a stabilising role in society:

> "What I'm saying is...you cannot make this conflict rise in that home, because by all, either right or wrong…*you have to make them live together*." (Chiefs 02/P2) (my italics)

As a counterpoint, a few community members spoke directly of the greatest value being in '*healthy* families', such that separation of parents where both were carriers might also be seen as a positive choice towards build new healthier families. This was not a common view.

4.2.4.2 Openness and empowerment

During discussions[76] about the potential benefits of SCD information sharing, the concept of openness was often drawn upon to imply that knowledge could act as a form of empowerment, either for individuals or for the wider community or society[77]. Information sharing on SCD was described as a form of empowerment for individual families, allowing them to take more control over their lives, including taking better care of an affected child; reducing blame and associated forms of stigma within families and communities; and for parents to understand their future reproductive risks. As a field worker explained:

> "If the parents are well informed about that and they understand…[they] will be very much aware…and as P4 says 'information is power' and on the other hand people perish for lack of knowledge. So if they get the knowledge, they will continue managing the kids very well, ignoring the others, whatever the other people will be saying." (IDI07/P3)

A further illustration is taken from a health manager's comment on the way that knowledge could empower people to make informed decisions, at the same time pointing to openness as fundamentally trust building:

> "…if you want to get the confidence of the community and whoever you are talking to, give the right information for them to make a right decision" (IDI12).

[76] Most consistently, this included several members of the District Health Management Team, the urban community representatives, and several staff members (an early career scientist, the senior field worker group and a data entry clerk).
[77] This theme will be taken up and illustrated further in Chapters 5 and 6, where it figured more prominently.

It was also seen that better understanding at individual and family levels could contribute to better understanding at a community level, through normal processes of social interaction between families. Drawing on comparison with HIV/AIDS policy, as was common when talking about 'openness', a community facilitator pointed out the effect of spread of information across a community:

> "Ok**,** I'm trying to bring in to my mind a flashback, back in 1984, 1982, when the first AIDs cases were discovered or realised or detected. There was this situation like people who were tested positive were supposed to be quarantined, in some cases it was said they should be isolated from the rest of the people, as people said, the society was like, there was that bigger percentage of stigma. So I think in this same case, ok the condition has been there in the community for several years, but many people don't know or don't understand about it. And I think as little information start getting in, time by time, day by day, it will open up their minds and they will accept. Because right now we are living with HIV patients, and people are caring, it's no longer an issue as it used to be. I think if the clinical officer decides to take a step and explain to the parents, they will have some questions the first time, they come back to the clinic, they are explained. As time goes, they will internalise the information and accept, and when few members of the family accept, and then later a few members of the village accept, and then later on a few members of the location accept..." (IDI07/P3)

As an example of the way that openness in information sharing can work at a societal level, a field worker in Phase One related the story of a mother whose child had been found to have SCD through participation in KGBC, and whose attitude had changed from an initial one of denial and anger to one of positively seeking to influence community knowledge and attitudes by becoming a local advocate for SCD in her village. In this way, many community members saw that openness would mean that parents could take control of their situation and improve their lives now and in the future. These views often drew on ideals of development and the desirability of movement over time towards a situation where the individual and public health costs of SCD would be better controlled.

4.2.4.3 Researchers' responsibilities

Through these discussions on the importance of information sharing on SCD, views on researchers' responsibilities for information sharing were articulated in two main ways. The first was related to the existence of particularly severe needs in children with SCD, and therefore also in their parents, for this type of help. Mothers of affected children reflected on the nature of this responsibility by

drawing on their own sense of needing to take particular care of children who were often in pain and unwell, including the development of particularly close bonds with affected children. Since the main harms that many community members were concerned to prevent were the physical and emotional costs to 'innocent' mothers and to children suffering from SCD, the unfairness and poignancy of situations that might lead to the abandonment of the mother and child, and possibly the child's demise, was a strong emotional force in many arguments. In part this reflected a tendency to perceive responsibilities of researchers as being for the medical care of the child, as part of a wider conflation of research and medical care. In part it seemed to be based on wide recognition of the extremely vulnerable position of many mothers, particularly those who are educationally and economically disempowered. At the same time, the interests of mothers and children were often aligned with those of the family overall, as described earlier (section 4.2.4.1).

A second area discussed by some research staff was the way in which they saw SCD information sharing as a responsibility of researchers based on fairness, and expectations of benefit sharing perceived by the community[78]. As a KGBC field worker explained:

> "Oh yeah some benefits should be there because…these who were sicklers…don't have even fares, I mean bus fares for the children to be brought to the hospital. I think it's also a burden, it's a big burden, okay, they are trying hard to find something to bring the child to the hospital and you'll find that they have defaulted because of what? They have nothing, in fact they don't have even anything to sustain their family [food], so you find it's really difficult and difficult and difficult to the family." (IDI07, Phase One 07)

This comment seems to reflect concepts introduced in Chapter 1 around researchers' responsibilities in relation to background injustices in the setting in which they work (section 1.8.1). Several field workers in Phase One also commented on the difficulty that community members have in distinguishing between different studies, whether these were being conducted at the same time or sequentially, as will be discussed in more detail in Chapter 7.

[78] This issue mainly arose during discussions with some research staff in Phase One, and was not specifically explored with community members

4.3 Reflecting on agreement and difference on sharing SCD information with parents of affected children

In Chapter 2, I argued that agreement and difference in opinions within a community, and between communities, may be made more transparent, and therefore amenable to resolution, by using a pragmatic distinction between perceived 'facts' and values as an organising concept[79] (Macklin 1999). In this way, differences (or agreement) may be seen to occur around peoples' perceptions of 'facts', the values they hold, and/or the way that values are balanced against each other. In what follows, I aim to summarise the findings presented across this chapter using this organising concept, in order to clarify the nature of agreements and differences between community members, illustrated in Figure 4.1.

Overall, the figure usefully illustrates a high level of agreement in perceptions about 'facts' and values, with most of the issues raised being non contentious. It emphasises (and explains) the wide and strong support for SCD findings being shared with families during research, particularly *general*[80] information on its inherited nature.

The main area of contestation during discussions concerned the *probability* that sharing *specific information on parents' genetic roles* would generate more benefit than harm. Although the majority (including all affected mothers) perceived this probability as high, the view was contested. Differences in perceptions about the likely balance of harms and benefits influenced the way that other values were prioritised:

- Where the benefits to families of explaining parents' genetic roles were seen as high, values attached to family interests (agreed values #1 & 2) and openness (agreed value #3) were aligned, and researchers' responsibilities were perceived as promoting these (values #4 & 5). For emphasis, I note that this was the majority position.

[79] Chapter 2, section 2.4.1.2
[80] That is, that the condition is inherited from both sides of the family over many generations, but without highlighting individual parent's specific contributions as carriers

- Where the risks to family stability of explaining individual parents' genetic roles were seen as high, values relating to family interests and openness came into conflict. In this case, family stability was generally seen as more important, and it was felt that researchers should not share information on parents' individual genetic roles in SCD.

In this way, researchers' responsibilities were primarily framed in line with the demands of other types of values, within which the interests of vulnerable children and their families were generally paramount[81]. At the same time, an important part of the explanation for differing views on the probability of beneficial or harmful outcomes of sharing information on parents' genetic roles , as shown by 'fact' #6, was recognition that different families were likely to respond differently. This feature meant that much of the difference described above in balancing values could be resolved, as will be discussed further in the following paragraphs.

Figure 4.1 Agreement and disagreement on 'facts' and values in sharing SCD information

Facts agreed [a]	*Values agreed* [b]
1. There is low awareness of the biomedical nature of SCD; instead it is commonly seen as supernatural.	1. The special interests of vulnerable mothers and children should be protected
2. Biomedical care is often not used routinely over time because its chronic nature and inherited cause are not recognised	2. The interests of family members are related to the interests of the family as a whole; so that family interests are particularly important and should be protected
3. Vulnerable children, mothers and their families are at risk of serious harm through family breakup because gendered blaming and paternal denial of responsibility are common features.	3. Openness and truth are important intrinsically and instrumentally, with emphasis on empowerment through knowledge, and progress as societal good.
4. Traditional care is usually sought for supernatural disorders; individuals blamed may be harmed.	4. Researchers have a responsibility not to harm/to prevent or minimise harm to participants.
5. Explaining inherited basis of SCD in general terms will promote uptake of more effective care and reduce conflict in families.	5. Researchers have a responsibility to share benefits of research with participants, and to engage with issues
6. Families' responses to information on the genetic roles of parents of an affected child will vary, depending on individual and contextual influences.	

[81] At the same time, this assessment may be influenced by the specific circumstances of these discussions; primarily that these responsibilities are bring framed for researchers in relation to genetic information which may already prioritise concepts linked to families.

7.	Counselling processes can increase the acceptability of sensitive information and minimise risks of harm.	of wealth disparity[c].
	Facts contested	*Values contested*
1.	Probability that sharing information *on parents' genetic roles* in SCD will promote interests of family given challenges of : paternal denial of responsibility; parental awareness of future reproductive risks; and potential for requests for paternal testing (showing misattributed paternity).	1. *Balancing* of values #1, 2 & 4 against value #3, depending on position on contested fact #1.

Notes:

a. All agreed 'facts' were considered important and widely supported.
b. Values are presented in an order that reflects their prioritisation, although 1 to 4 were all strongly supported.
c. Raised by some research staff, but not widely discussed in this study.

Heterogeneity between families

During discussions, two interrelated factors were widely seen as the main influences on outcomes of information sharing on SCD in general; levels of *understanding* and *contextual* features. In relation to understanding, communicating about the biomedical nature of SCD, as for genetic information in general, was recognised as challenging for many community members, given the complexity of concepts around inheritance mechanisms, risk and the meaning and validity of tests. Differences in understanding (between researchers and participants) were therefore widely seen as

ways in which sharing SCD information might generate more harm than benefit. At the same time, the risk of harm through 'misunderstanding' was seen as amenable - at least to important degrees - to skilled communication efforts. Understanding, as an influence, was therefore framed as a *challenge* rather than as importantly undermining the strongly perceived benefits of disclosure.

The influence of context took on profound significance in these discussions. Community members' disagreements about the likely consequences of information sharing almost always reflected agreement that different families were likely to accept and react in different ways to the same information. Although understanding would be an influence, aspects of the family's context were seen as key. Important contextual influences raised were the nature of individual personalities and relationships within families, patterns of SCD observed in the family (dependent on chance), the influence of religion and the family's (particularly the mother's) socioeconomic situation. These influences were seen as crucial to responses to information about SCD, and of particular importance for the risks associated with explaining parents' individual genetic roles in the disorder. Other authors have also commented on the significance of pre-existing relationships in shaping family responses to genetic information in other settings, so that families 'carry their patterns [of relationships] into the arena of the disease' (Geelen, Van Hoyweghen et al. 2011). Similarly, newly acquired genetic information has been seen as reinforcing pre-existing family dynamics (Chilibeck, Lock et al. 2011). Arguably, a major point of difference between these effects in more developed countries and the issues raised in Kilifi concern the seriousness of the impact of *reinforcing* family dynamics that are *already structurally inequitable*.

In this way, micro and macro level features of context are brought together. Community members in Kilifi highlighted the role of gender relations, poverty and traditional beliefs around supernatural causes of health and illness as important macro level influences. For example, low recognition of biomedical concepts in general and underlying structural influences of gendered power were seen to work together against an expectation of benefit from sharing SCD information. Similar challenges for genetic testing have been reported in other settings with strong patrilineal traditions

(Naveed, Phadke et al. 1992; Wang and Marsh 1992; NCOB 2006). The fact that both macro and micro level features were seen as important predictors of outcome emphasises the need for communication on genetic roles in families to routinely take both structural and individual influences into account in order to avoid generating more harm than good, where these harms may be very serious for some families.

4.4 Conclusions

This chapter begins the process of exploring findings from Phase Two of this study, describing community views around the first set of morally important questions identified in Phase One. The questions for this chapter concerned the dilemma of whether or not information on SCD should be shared with families of *children found to be affected by the disorder* during participation in research, given the uncertain balance of harms and benefits apparent from Phase One. Many findings in this chapter confirm those of Phase One (Chapter 3), particularly concerning the potential for explanations about the inherited nature of SCD to limit harms related to low awareness about this condition; the possibility of harms linked to information sharing; and the influence of micro and macro level features of context in shaping likely family responses to SCD information. The main outcomes of deliberations with community members in this chapter include:

- Based on the importance of harm limitation, there was strong agreement that findings on SCD should be shared with families of children found to be affected during participation in research, and that this information should always concern its nature as an inherited condition from both sides of the family over many generations.
- Explaining *parents' individual genetic roles* in SCD has the potential to generate both benefit and harm, depending on *the way information is communicated* and *individual families respond*:
    - An important potential positive outcome is countering paternal denial and maternal blame; conversely, possible negative outcomes include increased gendered

stigmatisation, charges of misattributed paternity, parental anxiety through recognition of future reproductive risks, and threatened family stability.

    - It was agreed that most families would benefit from understanding this information; however uncertainty over the benefit/harm balance led to recommendations that this type of information should be shared through family counselling, within which families most vulnerable to harm could be identified and supported separately.

- The discussions have produced rich descriptions of experiences and contextual influences that would support the development of counselling processes for families affected by SCD in this and similar settings.
- Key values emerging from these discussions concern the interests of vulnerable children, linked to family stability, and the value of openness as a form of empowerment at individual and societal levels. Perceptions of researchers' responsibilities for sharing SCD information mirrored the way that these values were balanced in different situations.

The conclusions on SCD information sharing are consistent with recommendations for genetic counselling in general in the literature and guidelines (Naveed, Phadke et al. 1992; Wang and Marsh 1992; Atkin, Ahmad et al. 1998; NCOB 2006), and provide ways of taking account of the diversity seen in this context. They highlight that this type of activity may be seen as a responsibility of researchers who undertake SCD screening as part of studies, unless such services are already available (Ravitsky and Wilfond 2006).

As introduced in Chapter 1, researchers' responsibilities for disclosure of SCD information may stand slightly separate to the importance perceived for this action by community members. Given the overall emerging importance of seeing 'disclosure' as a process potentially requiring a relatively complex form of family counselling as well as validation of testing and provision of care, one component of formulating responsibilities for researchers will need to take into account the investment of resources necessary to achieve the perceived benefits of information sharing.

Alongside the findings from the following two chapters, an analysis of the way these findings can be seen to influence researchers' responsibilities will be undertaken in Chapter 7.

Figure 4.2 Summary of community views on sharing SCD information in studies

<table>
<tr><th>Reasons for sharing information</th><th>Reasons against sharing information</th></tr>
<tr><td>Increased likelihood of using effective care, with positive physical, emotional and economic impacts on child and family, based on:
Explaining lifelong nature to increase long term uptake effective care
Explaining inherited basis as counter to supernatural explanations, seen to have serious harmful effects (blaming of mother and others in family; use of less effective care; fear of supernatural and HIV/AIDS)
Address common tendency for paternal denial of responsibility, particularly where more than one child affected:
Gendered blaming towards mother as ‘cause’
Suspicions of marital infidelity causing supernatural illness (chirwa) or involving misattributed paternity
Acts as counter to concerns about parental conflict through understanding future reproductive risks (see reasons against sharing information)
Ability to pass information to others affected in family/wider community

Addressing challenges: Since other inherited diseases known, will help in explaining, and often seen as less serious than non-inherited diseases (e.g. asthma is seen as inherited and a disorder children ‘live with’)</td><td>Explaining parents individual genetic roles may lead to greater harms through:
Denial by father and greater blaming of mother (as inherited from her)
Increased suspicions of misattributed paternity (especially since father feels well)
Increased awareness of future reproductive risks may increase conflict between parents and risk separation, with greater impact on mothers and affected children.
Causing worry about nature as lifelong incurable disease – risking hopelessness
Challenges:
Underlying causes very difficult to address, particularly structural gendered blaming and supernatural beliefs
Technically difficult to explain (genes, healthy carriers etc)</td></tr>
<tr><td colspan="2">Main emerging values:

Protecting interests of child and family: Seen as fundamental, includes limiting harms associated with low awareness of SCD. Disclosure mainly seen to support this value but also some risks, based on paternal denial; and also challenges in explaining. But <u>on balance</u> information sharing seen to offer greatest likelihood of protecting interests of child and family; risks of gendered blame seen as greater without information on genetic roles of parents than with this understanding.

Openness: Important to empower individual families and wider community to limit harms and promote benefit through better understanding/care of SCD, including reducing stigma. Works to support a value of protecting family interests.

Researchers’ responsibilities: Given convergence of above values, to respond to particular needs of sick/vulnerable children and their families <u>and</u> to support openness in the interests of affected families, including limiting harms to which they are exposed; to act fairly in relation to inequities in access to resources between researchers and many affected families.</td></tr>
<tr><td colspan="2">Main influences:
Micro: Observable patterns SCD in family; severity of child’s condition; individual personalities and relationships; cultural influences e.g. supernatural beliefs, taking second wife, seeking family reconciliation; economic circumstances (including formal education/independent economic status of mother) and related access to health care
Macro: Gendered power imbalances, economic hardship, supernatural paradigms for health beliefs</td></tr>
<tr><td colspan="2">Important contingencies:
Communication & counselling: Requires skilled personnel, explain over time to both parents; give information on healthy carrier status; balance realism and hope; explain genetic roles of both parents with caution. Importance of addressing increased risk of harm to some families of explaining both parents’</td></tr>
</table>

CHAPTER 5

Phase Two. Discussing information sharing for study findings on sickle cell disease: Families of children with carrier status

5.1 Introduction and background

This chapter parallels the approach taken in Chapter 4 of taking forward a set of morally important questions from Phase One through a process of deliberation with community members in Kilifi, as part of Phase Two. In contrast to Chapter 4, which explores the first set of questions raised in Phase One on sharing information on SCD with families of affected children, this chapter turns to the second emerging theme, on the importance of sharing information *on carrier status* for sickle cell disease (SCD) in children who are screened during research. As before, the informed views of a varied group of community members engaged in a deliberative process across a series of individual and group discussions form the basis for assessing the importance of disclosing carrier information.

5.1.1 Carrier status for SCD and screening in the Kilifi Genetic Birth Cohort study

There are key differences for health between having SCD and being a carrier for the condition (from here, referred to as being a carrier without specifying for SCD), as explained in Chapter 1 (section 1.5 and 1.7.1). Carrier status, sometimes also referred to as 'sickle cell trait' is generally considered a relatively benign condition (Tsaras, Owusu-Ansah et al. 2009), with its main importance centred in the potential for affected children to be born to parents who are both carriers[82], and the protection conferred against malaria (Williams, Mwangi et al. 2005). In the Kilifi Genetic Birth Cohort (KGBC), the percentage of children found to be carriers was around 18%, in contrast to 1% of children affected by the condition.

[82] Where both parents are carriers, there is a 1 in 4 risk that any future child will have SCD and a 1 in 2 chance of carrier status (see section Chapter 1, 1.5)

During KGBC, young children were screened at home for SCD status in addition to sampling for wider genomic analysis (see Chapter 1, section 1.4). All families were actively given some information on findings from SCD screening, but carrier results were not differentiated from those for children with normal haemoglobin. Families of children with normal haemoglobin and those with carrier status were informed that their child 'did not have SCD', while those found with SCD were referred to the specialist clinic, run collaboratively between researchers and Kilifi district general hospital, for confirmatory testing, counselling and care. In Chapter 1, I described that this approach to feedback of carrier findings was recognised by researchers as a compromise, the nature of which this study aims, in part, to explore.

Against this background, the discussions reported in this chapter explore the views of 58 community members[83] on what they consider to be the most appropriate approach to feeding back carrier findings in young children where this information is generated during research, including:

Should researchers feedback carrier results from screening for SCD in children when conducted as part of research? If so, what information should they share? What ethically relevant concerns are at stake, how might these support or be balanced against each other, and what conclusions emerge? How could information sharing activities be seen as the responsibility of researchers? Therefore, in what way should the value of information sharing be seen and justified, and what contingencies and practicalities are raised?

5.1.2 Organisation of chapter

The main section of this chapter (5.2) presents community members' views on sharing carrier findings. The section first presents reasons in support of sharing carrier information, followed by those against, before addressing contextual influences and emerging values. In section 5.3, I explore agreement and difference within these opinions, summarised in Figure 5.1, drawing on a

[83] This question was not discussed by one group of chiefs (KCR02) due to time constraints.

pragmatic distinction between 'facts' and values as an organising concept. The chapter conclusions are presented in section 5.4, followed by a figure (Figure 5.2) summarising the main findings.

5.2 Local experiences, arguments and values

A majority of community members felt that carrier information generated on a child screened as part of research should be shared with their parents, but this was not a unanimous position and a range of influences were recognised that coloured the emerging views. Additionally, some community members changed their minds over the course of the deliberations. The sections below describe the reasons given in support of or against the majority view that carrier information should be shared with families, as well as contextual influences on and emerging issues from these views.

5.2.1 Reasons for sharing carrier information

For the majority of community members who felt that information on carrier status generated during studies should be shared with families, their views primarily drew upon perceived benefits of disclosure for the child, their family and the wider community, both in the short and the longer term, as described in the following paragraphs.

5.2.1.1 Knowledge can reduce the child's future reproductive risks

The main reason community members felt that carrier results should be returned to families was that this knowledge was seen as benefitting the participant child, and indirectly their family, through the child being able to use this information to make reproductive choices in the future. This benefit was based on an assumption that an individual shown to be a carrier in early childhood would later be able to check the sickle cell status of a future potential marriage/life partner. If testing were available, all community members believed that the sharing carrier findings in children would be important.

Acknowledging that screening for SCD status of a future partner could not be assured, some community members still felt the anticipated benefits of reproductive choice could be achieved.

Community members raised the possibility that services might in fact become available when the children participating in this study reached adulthood, perhaps 15 years or more hence. Many agreed with this idea about the unpredictability of the future, particularly for medical services. One suggestion was that the size of the genomics study in Kilifi itself generated the possibility of a wider local understanding of risks in future:

> "There is, maybe in this place where you are doing the research, many children, many children will be tested, maybe in future a person can come to meet with a person and ask each other about their situation, maybe he comes to find maybe this one was also tested [in the research] and their condition is this or this…and they can come to meet a person who is not a sickler or a carrier." (KCR01/P2)

5.2.1.2 Knowledge can help to prepare for and minimise harms in future

Whether or not future testing of a marriage partner was taken as a possibility, a majority of community members believed that carrier findings shown during research should be shared, based on a perceived benefit of knowledge as a form of individual empowerment, with the potential to act against a range of possible harms in the future. In this way, parents who understood that there was a risk of a child being affected by SCD in a subsequent generation would *be prepared* for this eventuality and be able to prepare their child for the same possible outcome, even if they could not avoid the risk. This was seen as important because low awareness of SCD within the community was associated with a high likelihood of ineffective care seeking as well as harmful forms of blaming, as described in Chapter 4. The quote below illustrates this point using a slang phrase of '*amejiset'* that graphically combines Kiswahili and English, literally meaning 's/he has *set* her/himself':

> "At least, at least when s/he comes to experience, s/he will be prepared [*Kiswahili: amejiset*]. At the time s/he comes to marry and if s/he has a child who turns out, that is, s/he has a child like this, s/he will remember 'eeh I was told, I was told, in the past I was told'" (KCR01/P4)

Another community member was clear about this value of carrier information and the way that he, as a parent, would pass the information on to benefit his child:

> "He [researcher] should tell me the full information, so that, whatever he tells me, I will also be able to tell him [child] when he grows old, I will also tell him [child] so that he can know his condition, how he can live with his wife and [when s/he] starts getting children, so that he does

not marry and experience a condition that he would never have known about if he was not given the information, but since he was given the information, he may remember this and the effects in the future." (KCR02/P1)

Similarly, a field worker described the benefits of knowledge, even without testing, as generating better understanding of the condition and its origins:

"If it [the SCD testing service] is not there then I think it's also helpful, because knowing that status the future generations won't be surprised, because the parent would have gained that knowledge of knowing at least that in future I might have some kind of, children with some illness. Though the other parent was not tested but at least the father knows probably the mum is a carrier, and that's why we're having these kids. Though the test itself is not there, but at least he will have a clue that I was told. So it does make a difference, it does." (IDI06/P2)

More explicitly showing the concerns that information sharing can help parents to understand the nature of SCD and ensure any affected children receive effective care in future, a community member explained that:

"They should be given [information], because even if there is no policy, but the parents have that idea and the doctor, that there is this and this, even though they will have married each other, they will have their grandchildren. So immediately they see that condition, they'll know this is what the doctor told us, immediately, there will be no need of taking the child to the witch doctor to be given traditional medicine but will immediately be taken to the hospital to be seen." (KCR02/P3)

The strength of feeling that some community members described for a need to share carrier information is well illustrated by the following comment by a community representative:

"He [the child] is fine, yes, he is fine, but because we want to know the reason, we want to give first priority on health, it's mandatory, we have to enlighten the parents that this problem may manifest itself in future and maybe we shall not have, maybe they will not have the information because it is not yet known. So, if this information is not yet known and the condition ends up affecting those who will be born in future, who are not aware, how will this problem be solved? So I think they [parents] need to be given the full information." (KCR 02/P4)

In any case, some community members saw a possibility of using carrier information to reduce future reproductive risks by relying on family stories around health to predict the likelihood of a future partner also being affected. However, this approach was seen as controversial given the inability to distinguish between carriers and those with normal haemoglobin through observation:

"No, the thing will be with that information with the family history, I mean if you understand the sickle cell disease information well, you will realize that even if your sibling is a sickler you also have a chance that you are normal so you can't just look at them and say well, the fact that

> they come from a family with sickle cell, it's possible that they, I mean you may end up forfeiting the very person who is normal and you love." (IDI09/P4)

One mother of a child with SCD raised a specific benefit to the parents of the carrier child that the knowledge would act as a warning that they themselves could have a child with SCD in the future. Again, in this eventuality, they would be prepared in a way that would address doubts about the cause:

> "It is good to explain to them because there is a possibility... the same parents might get another child who might have the condition... So it will be good since you will somehow be alerting them. You tell them that in future there might be this and that and so you shouldn't have any problem because even if you give birth to a child with this, it's caused by you, parents... if you have a child with this condition, you shouldn't worry why it's carrying on." (Mot1/P1)

Further, since this knowledge was perceived as likely to be passed on within the family, and within the neighbourhood, the benefits of knowing about carrier status were seen as positively impacting the health and welfare of other family members and the wider community.

It was recognised that sharing carrier information would inevitably give rise to some anxieties. The acceptability of causing worry seemed dependent on the extent to which the information was seen as useful or - conversely - unhelpful. For those who saw knowing carrier status as valuable, the generation of some anxiety was seen as an acceptable and inevitable part of gaining the benefits of knowledge. As a field worker graphically put it:

> "Ok (pause), ok to me I think its ok for them to be worried, because if you are worried it means you still want to live!" (IDI07/P3)

More subtly, a chief described the way he balanced the negative impact of anxiety:

> "So me I think one is getting, one can get disturbed in his mind when he or she has no knowledge of something, but if he is aware of something, he is psychologically prepared to tackle or to cope with such a problem when it happens. So I think it's better these parents be told, so that they can prepare their child in future to have no worry about it, and to know how he will cope with the disease when it rises in his life." (Chiefs 01/P2)

5.2.1.3 Knowledge can create a welcome demand for improved services

In addition to a better understanding of SCD within the wider community being an expected consequence of disclosing carrier status during research, some community members - including a

health manager - suggested that wider awareness at community level of these issues could lead in time to grass roots pressure for better services to be provided, either through public advocacy for health policy change or pressure for more information to be provided to individual patients empowered through knowledge to ask questions, as illustrated by the following quotations from a chief and a district health manager:

> "You have also said that currently there is no policy in Kenya for testing sicklers, but the information will assist the parents, if they are many I can say it will assist them to lobby for a policy, so that testing will also be done even in schools…" (Chiefs 01/P6)

> But I think if they [parents] get more knowledge about this probably they would see the importance of also inquiring from the clinician to know, and to be explained more...and [if] the results are positive, what does that mean? So they will create that demand and clinicians probably will have time to explain, because usually that does not happen. Yeah the results come, the clinician just prescribes treatment, explains, the patient goes. But probably if the community… are sensitized about this then I think demand will be created and they will now be asking the clinician what, what, what is this all about? And that way more information will be disseminated." (IDI13)

5.2.1.4 Knowledge can help in maintaining the health of a carrier child

A unique but important idea was put forward by one participant who was a carrier herself, and had lost two brothers to the disorder as well as being the parent of a child who was a carrier. Her son was screened for SCD at the age of 7 years, given the family history. She described a value of knowing her child's carrier status so that she could take particular care over his health. In part these views illustrated a realistic concern, since some health risks - although unlikely - are recognised for carrier status, as described in Chapter 1 (section 1.5). This participant's assessment also shows the potential for information sharing to raise less well bio medically founded concerns about health:

> "A carrier should also always be healthy, and then like nowadays you can be told to volunteer to donate blood so when donating you should know that you are a carrier and there is a limitation, because you know if you overdo it and the sickle cells in your blood, you are not a sickler but if they'll be a little bit more than the red blood cells, you'll also go down with the sickle cell [crisis]. And then until they pick up that is when you will come back to normal…So it's important. [In response to a further question about how she would have felt to be given this information on her son's carrier status earlier] I would be very happy, because I know now my son is a carrier, I will feed him well and make sure that he plays well, and all that, yeah." (IDI08)

5.2.2 Reasons against sharing carrier information

The benefits of information sharing described so far were continuously being balanced against harms that might also be generated. In what follows, I describe perceptions of potential harms and the ways in which these views were often countered, as part of the process of deliberation.

5.2.2.1 Living with worries about uncertain future risks

As described, the main reason that some community members (a minority) felt carrier findings should not be shared was the child's likely inability to use this information in future, given current lack of public access to SCD testing. In this context, knowledge of carrier status was seen as generating anxiety about uncertain future risks for 'no good reason'. Several community members felt that in this situation they would prefer not to be given the information *themselves*, in three cases[84] including statements (as below, from a community facilitator) seemingly reflecting an assertion of a right not to know:

> "I just don't think it's worth knowing, yah, because I am trying to think in any decision I have to weigh the benefits and the cost, okay so if you look at the cost, well I know I'm just anxious, you know, and the only benefit I can get from it is if I can ever use it to sort of protect myself. But if there is no structure in place to say test the other person that I want to get married to, for instance, so that I can know at least I have used the information to my benefit, then the cost is higher than the benefits. So for me I would rather just stay the way I am [not be told]." (IDI09/P4)

Similarly another community facilitator explained:

> "I think it's a tricky situation and I would think if the children are told or if the parents are told about their children being carriers, they will know in advance but it won't be helpful to them in any way. And if they know, I think I'm strongly on the side that they are *not* told, not told, yeah. They can be told, they will be aware of that, but unless they, like if the child is still very young, as they grow up that could sort of stigmatise their behaviour or the way they are growing up or the way they interact with other people, they will feel like I have this condition in me. And yet it doesn't harm, you know, they're normal, it's normal, it doesn't harm them in any way. So I think not telling them is most ideal." (IDI02)

Often views were expressed in a more circumspect way, illustrated by the following quote from a community representative:

> "I mean you will explain as well as you can, but later you will leave that particular person worried. When he [the child] comes to marry later, he will say that it would have been better if I

[84] 02, KCR2/P6 and 09/P4 – not systematically asked in every case though.

didn't agree for him to be tested. This person was living without any problem, and now that he [the child] has been tested, [has] the fear that when my child grows up and gets married it's possible that he'll have children affected [by SCD]. So you will have accomplished your goal but the parent will be left in suspense." (KCR02/P6)

At a wider level, a district health manager – while preferring that carrier results should be shared – accepted that the current lack of services for screening or supporting public awareness of SCD meant that sharing carrier findings could lead to widespread discontent and frustration:

"Yeah I think for this particular moment, in our set up, probably it might not be prudent to give that information out, because people will be interested, yes, I've gotten this information that I'm a carrier, what next? I will want to know more, and what can I do about this? And then it will come to a point where one requires the next service, but it won't be there, so that's where we would have created a barrier and we'll then create frustration, and yeah and emotion in the families, so I think at this particular moment …" (MoH01)

A few (two out of 58) community members strongly expressed a similar doubt about the value of sharing carrier information in the absence of a wider programme of medical support for the condition, as one described:

"After the research, are there going to be strategies to address the condition? Maybe at that time, once such a strategy has been identified, obviously, there will be a process of informing the community that some time back, there was this type of research that was done and now there is a drug that has seen discovered, so when you notice these symptoms you can get help from this place. At this point, the parents will be explained the importance of the research that was done and the results that were realised so that if they experience this condition… they will go the doctor for assistance, this is when they will be getting benefits." (KCR02/P6)

In some cases, opposition to the idea of generating anxiety about a child from a young age was accompanied by suggestions about alternative screening measures, such as premarital screening:

"I think these parents should not be told anything. But maybe as the kid grows, maybe when he is in his adolescent stage, maybe the youth now can be told more on the sickle cell, so that maybe when they are about to marry maybe they do some test so that maybe you can get your spouse, who is maybe normal or maybe sickle cell gene, something of the sort, yea. But the parents should not be told because after all they will be getting worried for nothing, and yet the baby will keep on living a good life." (Chiefs 01/P3)

Also illustrative of the value placed on avoiding unnecessary anxieties for the child, an older community representative was keen that information sharing should be done in an abstract way that did not directly implicate the child him/herself:

"I should be informed that there is this and this and this and this, like a story we read in books, we get this information and we get to know a lot of things because of this information. So we

> see its better if he is given the information like a story…*let him not to be told he is this and this*, but let him be told that there is an illness called sickle cell disease…" (KCR03/P4) (my italics)

Although many community members supported sharing carrier information regardless of access to public screening services, the effect *on the child* of worrying about uncertain future risks was particularly concerning to some community members. As above, views were influenced strongly by the extent to which the information was seen as useful to the child. However very serious adverse consequences were considered by some as at least possible, as described by two community representatives below:

> "You know children take a lot from their fathers and if you tell a child something it sticks … now that can affect him psychologically, that now there is some weakness here… and him, that condition won't affect him personally. Maybe it will affect his child. Now my opinion is we should let him be free of that curse so that he is free of that one and should continue to grow." (KCR03/P6)

> "When you tell him directly that you have this then he will be thinking a lot, and even consider committing suicide because even when I get married things won't be good." (KCR03/P4)

More often, community members felt that the information could be passed on in a way that would provide more benefit than harm through anxiety, although some disagreement continued on the most appropriate age for a child to be given this information. At a younger age the child might be more accepting but understand less, while at an older age (at adolescence, for example) the young person would be likely to understand more, but have greater sensitivity to personal information affecting their long term future and relationships. One participant was able to draw on direct experience of explaining carrier status to her child at the age of seven years, who accepted and understood carrier information well. A key point raised by one community representative (KCR01/P6) was that if carrier results were to be returned *it would be critical to ensure test results were reliable*.

5.2.2.2 Misinterpreting sickle cell trait as adversely affecting health

Community members were divided in their perceptions about the risk that parents would misinterpret carrier status as a form of ill health[85]. This phenomenon has been described as 'the vulnerable child syndrome'[86], in which children are inappropriately seen as vulnerable to illness on the basis of past events (Green and Solnit 1964; Miller, Robert et al. 2009); and as a form of 'medicalisation', defined as 'drawing into the biomedical domain of physical aspects and behaviours such as antisocial conduct that could be understood alternatively as a sin, a crime, a moral fault, or a disease' (p175) (Finkler 2005). In Kilifi, a majority felt medicalisation of a normal childhood could be an outcome of sharing carrier information in some cases, given low familiarity with the concept of genetic carrier status. The manner in which a staff member (IDI 08, section 5.2.1.4) described the 'advantages' of knowing about her child's SCD status also points to this risk. The potential for medicalisation was seen as occurring either immediately or in response to future illnesses experienced by the child:

> "I still support my point of [parents] not being told because you know once you explain to these parents that this kid has such and such a problem, you know their psychology will be fixed on your advice as the doctor, oh, the doctor said because my kid is not actually 100% right, well, so anything that will be coming across the kid's life the parent will relate it to the sickle cell, any small thing will be related therefore to sickle cell, so actually the parent will not be, will somehow be psychologically disturbed as he or she brings up this child, yeah" (Chiefs 01/P3)

An example of the way this form of retrospective medicalisation might work was given during a discussion by community facilitators (IDI09) on how and why health beliefs in general can be drawn upon over time:

> "P3: Yeah ...there is a... myth or what, that ... the navel from a young child [umbilical cord], it takes some few days for it to drop off, yeah, after the tie, so the *mkunga* [Kiswahili: traditional birth attendant] ... in those few days when they are taking care of that child, if it's a boy, they will take maximum precautions to make sure that it drops off clear from the genitals. But if it drops on the genitals, then they are saying, it's a belief that the boy will be barren in life, he will be impotent, he won't be able to sire, so it's something that they will carry [remember] yeah, later in life."
> P4: "But... [if] he... marries and the wife goes complaining and then this will be now shared 'oh, actually we remember this and this happened!' And without that, if the child grows up to be okay, then people will obviously forget. So that is why I'm saying ...later on if the child doesn't

[85] However the extent to which medicalisation of carriers might be seen as due or undue is not straightforward, both medically and bio socially, as discussed in Chapter 1, section 1.4.
[86] Green and Solnit (1964) describe the vulnerable child syndrome (VCS) as a set of clinical features in which unfounded parental anxiety about the health of a child resulted in disturbances of the parent-child interaction.

> show any symptoms, they are likely to forget and 20 years later it might be just a vague you know idea, somehow someone came along to tell us some information but we can't remember, whereas if there were some signs they would obviously remember this information for a very long time."

However, many community members felt the potential for medicalisation would be countered in the majority of families by a number of factors, including: the lack of symptoms experienced, such that families would either accept the condition as non harmful or forget about the finding in time; parents' reflection on their own health, since at least one parent must also be a carrier; and the much greater priority likely to be given to challenges and hardships confronting many community members on a daily basis. According to some, difficulties in explaining carrier status [87] might also be lessened by relating SCD to other conditions recognised as running in families with 'skipped' generations, such as albinism.

5.2.2.3 Increasing stigma for affected children and families

The potential for children affected by SCD or their families to experience stigma has been discussed in Chapter 3. This study was not set up to examine experiences of stigma in adults affected by SCD, but it was generally acknowledged by community members that individuals affected by the disorder would be likely to experience difficulty in finding marriage partners related to the way that symptoms would undermine their ability to contribute to physical tasks important to a typical family's livelihood:

> "Because even at home there is…a lady where they say that, a person having that problem of *mifupa* (Kiswahili: bones) cannot do difficult jobs, you hear. You cannot pound maize, dig, they say you cannot do because…that's why its really possible to be discriminated in getting married because they say that when you marry that person, she cannot work, mmmm, that [stigma] is possible." (Mot01/P2)

Given this potential for stigmatisation of affected individuals, some community members thought carriers might experience difficulty in seeking marriage partners because of their future reproductive risks. More commonly, this was argued against for several reasons. Firstly, the

[87] However, narratives from families in Phase 2, who commonly described their confusion and distress in trying to understand the pattern of SCD that unfolded within some families, suggested the sporadic nature of SCD inheritance was both puzzling and a cause of particular tensions about cause and blame (section 3.2.4).

influence of individual level factors on blaming and stigma was seen as more important, including the socioeconomic status of the family and the nature of relationships already formed[88]:

> "When they are in love, they are already in love, even if the parent likes (pause) there are other people who used to say that I am a Giriama[89] and so I don't want my girl to be married by a Chonyi, or I am a Kauma and I don't want my girl to be married by a Giriama. Also I think if the man and the woman love each other, it's a must they will marry whether the parents love or hate her, or whether the family like it or not, they will still marry. So I don't think there is a problem for her to be discriminated against [by the family], maybe if the husband or the wife have not come to an agreement but I don't think there is the issue of discrimination." (Mot01/P2)

In support of this view, some community members also felt that the point of premarital screening for both partners was to give the couple a choice; knowledge of carrier findings was not necessarily seen as likely to dissuade a couple from marriage where an emotional bond had already been formed.

Other points raised against a potential risk of stigmatisation in sharing carrier information included: children and families had the option to keep carrier information confidential; carrier knowledge could reduce pre-existing stigma based on an assumption of higher risk across all members of an affected family; and greater openness about the disorder could work towards reducing stigmatisation, a point made particularly strongly by all the mothers of affected children. As a community facilitator said:

> "And maybe if we go back to the stigma... I think if you borrow a leaf from what HIV/AIDS campaign have been doing, the kind of education which is being provided is like fighting that stigma because it's about that stigma which it makes this problem has, yeah, it has spread very much, easily yeah" (IDI09/P3)

Additionally, in relation to risks of gendered forms of blame and stigma in affected families[90], knowledge and acceptance of carrier status (for one or both partners) *before marriage* were seen to support mutual trust and help parents take care of an affected child in the future, if this were an outcome.

5.2.2.4 Creating suspicions of misattributed paternity

[88] As for SCD information in general, as discussed in Chapter 4 (section 4.3)
[89] Giriama, Chonyi and Kauma are ethnic subgroups within the overarching Mijikenda group
[90] See Chapter 3, section 3.2.4.1

In the same way that doubts about paternity were raised by information sharing on the genetic roles of parents in Chapter 4, some community members saw that a similar concern could emerge from explaining a child's carrier status:

> "Maybe to add on that point, that point, when a child is born and she is found that she is a carrier that means she has only one gene, now if you tell the parents, the two parents would want to know where that gene is coming from? Is it coming from me or from who? That is another conflict!" (Chiefs 01/P4)

Doubts about misattributed paternity were also seen to be potentially caused by a common confusion between genetically inherited conditions and sexually transmitted infections, as described in the previous chapter. In the case of children with carrier status for SCD, there were suggestions made that some parents may wonder if the condition had been transmitted sexually, rather than by inheritance. However, this set of potential concerns around misattributed paternity was generally considered minor in relation to others raised during discussions. Questions around researchers' responsibilities towards fathers who request screening themselves for this reason are addressed in the next chapter (Chapter 6).

5.2.2.5 Questioning the reliability of parents in passing carrier information to their children

A fundamental assumption underlying expectations of benefit from sharing carrier information with parents of young affected children was that this information would be passed on in a reliable way to the affected child at some point in the future. A few community members strongly argued against disclosing carrier information through parents on the basis of the unreliability of this approach, while continuing to believe that it was better for children to have this knowledge about themselves. Although this argument was not raised commonly, it was influential in the situations where it was discussed[91].

This judgement was largely based on views that many parents would be unlikely to remember and accurately recall this unfamiliar information over the period of time needed before findings could

[91] This argument was put forwards by a small number of individuals including a well respected older community representative with local leadership roles, an experienced community facilitator and an administrative chief)

be understood and acted upon by the child, that is, up to adolescence or adulthood. Firstly, and most commonly, these community members considered that parents would simply forget the information over time, given the lack of symptoms experienced by the child, as has already been considered in section 5.2.2.2. Secondly, the risk of misinterpretation of carrier findings, already recognised as possible at the time of information sharing, was thought to be even more likely over time; the knowledge transferred to the child in future might therefore be inaccurate in important ways. Thirdly, parents might deliberately decide not to tell the child, a point raised by a chief basing his view on experience that parents often do not share information on HIV status with their affected children, preferring to avoid this upset for the child and family, as illustrated below:

> "Me I still disagree, they should not be told, because most parents, like we have a programme in my area on HIV/AIDs infected children. So we ask some of the parents, when a child reaches seven or eight years you are supposed to disclose the news to the child, but there is no parent who discloses the news to the child, unless the child grows and gets to know for himself or herself. So it will be the same thing with sickle cell, even if you tell the parent, he or she might not disclose the news to the child, yeah, so its better the child himself is told about the disease so that he can help himself or herself." (Chiefs 01/P3)

For these reasons, some community members felt carrier status should only be explained directly to the child by an 'expert' - not the parents - to avoid misunderstandings and ensure the knowledge could be appropriately acted upon in future. Some therefore preferred that children be tested for carrier status at an older age. One suggestion was to leave a comprehensible written record of the information with the family to give to the child later in life. Another - attracting immediate challenges for confidentiality from a colleague - was that carrier status be recorded on a child's health card.

Somewhat ironically, a community facilitator pointed out that the likelihood of parents remembering the information would be linked to their interpretation of the findings. In this way, *parents who saw carrier status as a concern for their child's health would be more likely to remember this information* than those who (more accurately) interpreted the finding as having little importance for the child's health. Memory would be prompted and influenced by concerns about the child's health:

> "…to some extent I think they would remember, but what will make them remember is that they feel … the child is sick…so they will remember all through that my child is sick, not my child has a condition which needs a person to make a good decision during marriage, but they will just remember my child is sick, my child is sick…that thing cannot move out of their mind" (IDI09/P1)

Other community members felt parents were the most appropriate channel for passing information to a child, particularly where testing in later life or storage and later dissemination of results were not feasible. As an early career researcher explained:

> "Obviously we can argue that the right time is later on in their lives, when they can understand and when they have a normal childhood and when now you know it matters. But given our situation in the third world, where we already don't have enough money, we have already spent money testing them and finding them carriers, and then we are going to wait until they are 18 and then recall them so we can counsel them about marriage? That's even a waste, more waste of money! If we are thinking about them, that is. Unless we are saying we can ignore them, and hope that the problem will go away, and hope that the 17% carriers will not marry each other and double the prevalence of sickle cell, which will double the cost of treating this same disease 20 years from now." (IDI05)

In addition, parents were considered by many as best positioned to offer a sensitive explanation based on their close understanding of the child, and ability to judge when and how to pass on the information in the most acceptable way, including over time, as illustrated by the following quotes from a community facilitator and chief:

> "They can help to prepare and explain this to the child over time. They can also explain that being a carrier is not an illness, particularly since they themselves are also carriers." (IDI01)

> "I want, I just want to add a point, by telling the parents I think psychologically...you will be preparing them, and the approach of them telling their child he is having such a gene it will be also very easy for a child to understand, [rather] than waiting up to his youth stage then getting tested in school. That one maybe will even be disturbing his mind ...yeah, but if the parents are very much aware of it, they will know how to ... inform their child before, and maybe their approach of telling their child will be better off than waiting till he is tested in school." (Chiefs 01/P2)

5.2.2.6 The influence of HIV/AIDS control policy on understanding of carrier status for SCD

One form of medicalisation of carrier status in SCD arose from a mistaken comparison with the common use of the term 'carrier' in HIV/AIDS transmission. Since it was widely known that 'carriers' of HIV/AIDS would later develop the illness AIDS, use of the term 'carrier' in relation to SCD was considered likely to have similar future connotations:

"In fact some people think that if you are a carrier [for SCD] it will reach a time that this thing will explode and now you will become sick." (IDI04/P1)

Additionally, shared use of the term 'carrier' implied a link between the conditions that was seen to strengthen potential for stigmatisation in SCD:

P: …"if I go out and tell somebody 'I am a carrier', people will go, like what, of HIV, or what? You see? But you have to be definite enough, like me, I use the word sickle cell *trait*...because the word 'carrier' it was used so much with HIV. So I prefer when somebody asks you, 'are you having normal blood and all that?' I would say 'No I am a sickle cell trait,' that is the word I like using most.
Interviewer: Mmhh, so you don't like the use of the word 'carrier'?
P: The carrier, I also use it but I use trait before I come to it. [When they ask] 'What does that mean?' I tell them I am a sickle cell carrier." (IDI08)

5.2.2.7 Supernatural beliefs and carrier status

Several community members described a set of anxieties that may follow sharing carrier information linked to the prevalence of supernatural beliefs in the community[92]. Where a child was perceived as suffering from an abnormality, even if this was not currently visible, it was felt likely that some parents may seek the help of traditional healers to 'treat' or remove the abnormality, as the following exchange between the interviewer and a community facilitator (IDI09/P3) illustrates:

"P3: That's the take home message, directly, the take home message will be 'I am sick, yeah I am sick'
Interviewer: I am sick? Ok
P3: and if at that particular point it won't be handled [well] maybe it won't be handled adequately, then they might even go beyond, looking for other ways of maybe removing or exorcising, because it's like you have something, so I can explore other ways, not medically yeah if it's not handled well …
Interviewer: Imagining what kind of ways people might choose to try and do this?
P3: Yeah because if we are saying okay how is it caused? or how did it start? yea and we are saying we don't know, maybe it's because this place had so many mosquitoes and think this gene has generated, so it's like ok if it's that gene then yea if the hospitals maybe have failed to identify the cure, maybe our herbalist or traditionalist healers might …"

There was widespread acknowledgement that the way in which information was communicated would centrally influence parents' ability to understand and explain carrier status to their child.

5.2.3 Individual and contextual influences in interpreting carrier status

[92] As also described in Chapters 3, 4 and 6

The previous two sections have explored arguments in support of and against sharing information on carrier status in children. Throughout these discussions, community members often saw that the differences being raised around parents' interpretation and ability or inclination to accurately share carrier information represented actual differences between families and individuals within families. As one community representative put it:

> "When he has been told that...when you leave you don't know how he will react back there, because of the way the heart is... they [people] are not the same…" (KCR02/P6)

In other words, some parents might undertake this task more effectively than others, although the drivers for this 'effectiveness' were to some extent controversial. Reflecting similar emerging issues in Chapter 4, community members recognised that a number of factors operating at an individual or family level, including the relationships between individuals in a family, could therefore influence the extent to which carrier information would be likely to generate positive or adverse outcomes. The main reasons discussed by community members in relation to this knowledge were individual personalities, the patterns of SCD visible within families and socioeconomic factors, with particularly heated discussions over the role of formal education:

> "You know not all people are equal, even those who are learned, who have their education even them they are not equal, that one you will see he is like this, that one you will see he is like that. But if those who are not educated at all, when you tell them something is this and this, he will think in his mind, because God has given him a brain to think, he will think to say ah! Then these things can be true so let me go and check for myself." (KCR01/P5)

This quotation already suggests a perceived unimportance of *formal* education, a perspective that was controversial, as shown by the following comments:

> "They [the parents] may accept that my child is a sickler, because he or she may see the signs, and maybe after some time the child becomes sick. But here is somebody here who is not sick, and yet to tell him that your child is a *carrier*. Then I think it will take time for those who are not learned to understand." (IDI06/P3)

> "The highly educated people are the most difficult to explain to, but the ordinary people will listen and think, because education is different to intelligence." (KCR01/P6)

The way in which these existing understandings of health and illness - whether related to education or not - would interact with information sharing is illustrated by a field worker:

> "If you tell that parent 'ok, your son is ok but he is a carrier, I just wanted you to know.' [He will say] 'If my son is ok, oh thank God, I was hoping and praying for this. Now I think you are doing a useful job and its ok' … He or she was told that he is a carrier, but she or he basically wanted to know if the child was ok or not. That's the two things that they are asking, is my child ok or suffering?" (IDI 06/P2)

5.2.4 Revisiting key values: Protecting families, openness and researchers' responsibilities

In Chapter 4, I described that community members' reasons for sharing information on SCD in *affected children* often invoked values around three main themes: the interests of vulnerable children and families; openness as empowerment for individuals and wider society; and the responsibilities of researchers. In the following paragraphs, I will continue to explore the way that these themes emerge and work to both reflect and shape views *around sharing carrier information*, highlighting any differences seen between sharing information on the condition and for carrier status.

5.2.4.1 Protecting vulnerable children and family stability

Throughout this chapter, the interests of vulnerable children, their mothers and families tended to be aligned with each other and seen as fundamentally important in discussions about sharing carrier information, in a very similar way that that seen in the previous chapter. At the same time, the degree of vulnerability for a child found to be a carrier was seen as very different to that of a child directly affected by the disorder. Harms that sharing information on carrier status might limit were related to future and uncertain events rather than a serious existing situation. These potential harms and longer term perspective for carrier findings were, however, still considered important to the family at the current time.

A difference in the normative functions of 'the family' also emerged in these discussions, arising from the challenges perceived for some parents in passing on information about carrier status accurately and sensitively to their children in the future. This suggests that there may be practical limitations to the extent that parents can represent their children's interests, focused here on their ability to act as custodians of information about the child. At the same time, this limitation did not

seem to detract from normative guardianship functions in parent-child relations. Interestingly, a similar 'joint interests' role in parent-child dynamics is supported in two sets of guidelines from the UK on disclosure of carrier findings during genetic testing in minors, described in Chapter 1. These specifically mention that the interests of a child cannot be considered separately from those of his parents and other family members (Borry, Fryns et al. 2006). However, other similar guidelines from developed countries reviewed by Borry (2006) describe these interests as more separate, drawing on principles that emphasise the particular and separate moral status of a child which were notably different to most community members' views in Kilifi.

A further insight into values associated with childhood was offered by these discussions through the importance placed on protecting children from unnecessary anxieties, including those that might be passed on by parents who were not clear about the meaning of carrier status. From both these perspectives, views emerged about the importance of children being able to grow up free of unnecessary worries as far as possible, including that this was seen as an important part of future adult health and wellbeing.

5.2.4.2 Importance of openness as empowerment for individuals and society

In Chapter 4, describing community views on sharing SCD information with families of affected children, I described the way that openness emerged as an important value in discussions about information sharing on SCD at individual and wider community levels. Openness was particularly strongly articulated at an individual level, linked to perceptions of the importance of disclosure in working against serious and immediate harms of 'not knowing'. At community, or societal levels, openness in sharing SCD information was seen to support wider understanding of the disorder, with an important influence on stigmatisation, and to build trust in the KGBC as a study and in the research programme as a whole.

In the case of carrier information, openness was again seen as a very important value, although arguments made at individual and community level often seemed similar in their strength. At an

individual level, sharing carrier information was seen to empower affected families through the possibility of avoiding future reproductive risks, should wider SCD testing become available in future, and through preparedness, where wider testing was not available. Empowerment at an individual level is well captured by a mother of a child with SCD saying:

> "It's better to tell them so that if they want to get married they can go ahead and marry, and if they don't want to then they shouldn't be married, *but when you tell them is when they will know*." (Mot01/P2) (my italics)

Views sometimes reflected greater intrinsic, rather than obviously instrumental, values of knowledge for carrier information, perhaps linked to potential instrumental effects being less marked than for SCD information. For example, a community facilitator commented:

> "Yeah the knowledge is very important, yeah, even if I can use that information or that knowledge now to make an informed decision later in life, instead of maybe that information has been hidden from me yeah and I'm living as a normal person without knowing…" (IDI09/P3)

Similarly, but hinting at associations with HIV/AIDS policy, a community representative commented that "we are living in a time when everyone should know about themselves." (KCR03/P3)

At a community level, the main perceived utility was in empowering people through better knowledge to take control of risk, and impacts on the public health burden of this disorder. One example of the latter position is given by a research staff member quoted earlier (IDI05; section 5.2.2.5) who felt strongly that failing to share carrier information would be like "hoping the problem would go away" and not "double the cost of treating this same disease 20 years from now". Another staff member spoke of carrier information sharing as 'moving forwards', saying: "We should be moving forwards, because if we don't tell them the truth, then we will remain in the same state of fear." (IDI01)

A more direct example of knowledge as empowerment at a community level raised by a few people concerned the possibility that carrier knowledge could empower affected families/communities to lobby for better SCD services in the future. At the scale of a specific project, this may be difficult

to imagine, but patient advocacy and public pressure have in fact been the driving force for SCD programmes in many parts of Africa where these exist, in the form of charitable foundations (Ghana Sickle Cell Foundation 2010; Nigeria Sickle Cell Foundation 2011).

Some tension was perceived between a value of openness about carrier status at community level and one of protecting the interests of individuals found to be carriers in relation to the risks of stigma, well illustrated by a community facilitator's comment that:

> "As bad as it sounds, isn't that [that people base their marriage choices on genetic information] what we want? Because if we are going to look, I mean in the future, and say you want to have a sickle cell disease free generation in the future then that isn't that exactly what we aim to do? So that if you are getting married I mean the rest could be empowered to say, what is your sickle cell status and say 'well you are a carrier, sorry, I can't get married to you because I am a carrier as well' (IDI09/P2)

Finally, a negative impact of *a lack* of openness about carrier information was described. This was perceived as a potential for loss of individual or community trust towards researchers (and the research institution) if it became known that carrier information had been withheld. Examples of ways this might happen included future SCD screening with a conflicting result, or the birth of an affected child in a future generation of the family:

> "Maybe we may just say that we don't want to displease the parents, but then after that, yes, the kid will be a carrier and nothing will change that...when he or she marries another carrier, ok, 'we were told that the kid was negative, how did this come about? Or was there something which was being hidden as to..?' Ok, it will bring some question marks." (IDI06/P1)

> "Let's take for example I have a child and I'm told the child is negative... so I will believe that, and in the real sense he's a carrier. So when they grow up, the other child comes to ask 'what was I tested?' 'Yeah you were tested and we were told that you don't have sickle cell, you are negative' and later on maybe my child gets married and gets a child who has sickle cell... So it will be as if we are cheating them! We have to tell them the reality, they have to be told the reality from the beginning so that they can plan themselves." (IDI07/P2)

5.2.4.3 Researchers' responsibilities

A number of points were raised about responsibilities for sharing carrier information that might be seen as specific to researchers or relate to perceived rights of research participants. Two groups of community representatives described a responsibility for researchers as equivalent to that of a 'visitor' to the home, linked to the approach taken to recruitment in this study. The family had not

invited or sought help from researchers or medical providers, but acted as *hosts* who had welcomed researchers into their home. Certain expectations or norms for such a visitor to return hospitality, or at least take an interest in the welfare of the family, seemed to be extended to researchers working in this way, as illustrated by the three quotes below (within which I have inserted my italics to highlight specific areas):

> "We are saying that they should be told because you have had *visitors*, they have done research, *they have come, they have asked for permission* to do their research, why should you not even know the results? We are saying they should be told, or I'm saying they should be told." (KCR01/P5)

> "Again, the researcher *will visit you in your own home*, so he is *supposed* to tell you all the information about the research that is to be done to your child." (KCR02/P4)

Other community members pointed in a similar way to the relationship between researchers and participants as a form of agreement wherein parents might reasonably ask for results of SCD screening, including carrier status, and researchers support such a request:

> " I'm sure, I'm sure the parents, they'll demand to know, after they have been with the, I mean the doctor *who goes to their homes*, and maybe … they find that the child is affected then *they must have to know*…and if they demand, they will have their own rights to be told" (Chiefs 01/P5)

The language of rights, with implications for the responsibilities of researchers, was also used by a health manager early in an interview in talking about returning carrier results (although he later changed his mind about the most appropriate action, seeing that a wider programme of SCD information and service activities would be needed to support the return of carrier findings in an particular study):

> "I think they have the right to get that information yeah, and that the benefits, the parents will make decisions early enough for treatment, and I think they need to get this information, although the policy has not, doesn't define that clearly, but my view is that it is important, that it is better if the parents know about this as early as possible." (IDI11)

5.3 Reflecting on agreement and difference on sharing SCD carrier information

In this section, as in the previous chapter, I explore the nature of agreement and difference in community views by summarising the findings in this chapter under categories of agreement or difference in perceptions about 'facts' or values, as an organising concept (Macklin 1999). Figure 5.1 shows this process of mapping. In contrast to the equivalent figure in the previous chapter (Figure 4.1), which explored views on information sharing for children affected by SCD, Figure 5.1 highlights the emergence of greater controversy in perceived facts and in the balancing of values in discussions about disclosure of carrier findings. The controversy around facts emerges specifically in the context where there is no certainty of access to SCD screening later in life – the situation that pertains in Kenya at present.

In Figure 5.1, the contested facts relate to differing views on the probability of overall benefit or harm resulting from sharing carrier information. In part, as in Chapter 4, these relate to recognition of the influence of *diversity* within the community, so that some families are seen as more likely to be able to utilise this information in a way that brings benefit than other families, particularly where information is communicated effectively (for example, avoiding risks of medicalisation).

Additionally, for those who felt that the harms would outweigh benefits of information sharing, a value was raised around a *right 'not to know'*. This feature contrasts with values emerging in discussions around sharing SCD information for affected children. The underlying conflict in values occurs between the importance seen for children to grow up free of unnecessary worries (agreed value #4) and that of *openness* in empowering families to be prepared for possible adverse events in future (agreed value #5). The value of openness is *reinforced* by perceptions that information sharing with families might lead over time to greater awareness of SCD and its inheritance in the community, and to public health gains for society.

Further new values in this figure concern perceptions of researchers' responsibilities linked to: social norms, where researchers visit the homes of prospective participants; the act of agreeing to participate in research (as described in section 5.2.4.3); and professional norms of transparency (avoiding deception). These values were largely agreed.

Overall, as in Chapter 4, the main basis of divergence of opinion in these discussions can be seen as a function of diversity within the community but against a background of less obvious benefits of information sharing on carrier status than for affected children (primarily, not including limitation of existing harms). The potential for benefit is further reduced by uncertainty that parents will be able to effectively pass this information on to children in future. Given this lesser importance and urgency of disclosing carrier findings, the acceptability of harms is also lower. This is reflected in the greater tendency to claim a right 'not to know' about carrier status. An important implication of this articulation of a right 'not to know', albeit described by a minority, is to highlight the importance of including an explanation of carrier status in the informed consent process for SCD testing (Borry, Fryns et al. 2006; Knoppers, Joly et al. 2006)

Figure 5.1 Agreement and disagreement in facts and values on sharing SCD carrier information

Facts agreed

1. Where SCD screening is available on demand, sharing carrier information will help individuals, families and communities to reduce future reproductive risks and/or minimise harms associated with having an affected child in future.
2. Biomedical understanding of SCD is uncommon in the community; common supernatural and lay explanations are often associated with harmful forms of blaming and/or less effective care.
3. Better understanding of a health condition can allow individuals and communities to ask/lobby for more information and better services.
4. Knowledge of carrier status has some individual clinical utility[a].
5. SCD constitutes an important public health risk, comparable to HIV/AIDS
6. Families' responses to information on carrier status, depending on individual and contextual influences.
7. Counselling processes can increase the acceptability of sensitive information and minimise risks of harm

Values agreed

1. People should be able to control future reproductive health risks, if wanted.
2. The interests of the most vulnerable in society should be prioritized; the interests of children are aligned with those of their families.
3. Children who may be born with SCD in future (and their families) have interests that should be represented now.
4. Well children should be able to grow up free of unnecessary worries.
5. Openness is important in empowering individuals to act in their own interests; and in empowering communities/ societies to advance health and welfare.
6. Researchers have a responsibility to be open, and not to conceal findings; including as part of agreements around research participation.
7. Researchers who approach families at home as visitors have a responsibility to respect hospitality shown, as a cultural norm.
8. Researchers should support the interests of participants as part of the 'agreements' in research
9. Parents are well placed to judge and act in a child's best interests (are good guardians of their child's interests).

Facts contested

1. Where screening not widely available, probability of benefit to family, given risks of 'medicalisation', stigmatisation and emotional distress in parents and child.
2. Probability that parents will be able to remember/explain information about carrier status in the future (act as 'custodians' of information).
3. Probability of SCD testing being available in future generations.
4. Probability that understanding carrier status in the absence of wider SCD services will lead to frustration due to unmet demands for services.

Values contested

1. Where a child is found to be a carrier, the parents/child have a right to know/not to know this information; researchers should/should not share information.
2. *Balancing* of values #4 and #5, influenced by position on contested fact #1

Note: a - Raised by one participant with child who is a carrier (IDI05)

.

5.4 Conclusions

This chapter describes the second set of findings from Phase Two of this study, reflecting the views of community members in Kilifi on the importance of sharing information on *carrier status* for SCD when this finding is generated during research. These questions were identified as important during Phase One, and from dilemmas experienced by researchers on sharing carrier findings during a genomics study. Key findings from community deliberation in this chapter include:

- A lesser perceived importance and urgency of sharing carrier information in comparison to children directly affected by the disorder, including lesser probability and severity of harm and greater communication challenges (through parents, in the future);
- High perceived importance attached to better public awareness and access to SCD services, including people's rights to control future reproductive risks for this disorder;
- Confirmation of the importance of diversity within the community in influencing the probability of benefit or harm resulting from sharing *carrier information* for SCD in children during research, as described in earlier chapters;
- The need for communication and counselling about carrier findings to account for diversity within communities, including identifying families with high potential to experience *benefit or harm* through sharing carrier information, and taking account of a right 'not to know';
- Continuing predominance of a value attached to protecting the interests of vulnerable children and their families (including in the future), and strong support for greater openness on SCD in general, including carrier testing services as above;
- A new feature of researchers' responsibilities included the existence of social norms and expectations for researchers who visit community members *at home* as part of study recruitment; and of the role of trust, in relation to the current practice of not distinguishing between carrier and normal SCD screening results.

These findings will be taken forwards to Chapter 7 in developing an overall analysis of researchers' responsibilities for sharing carrier information when SCD screening occurs during research.

Figure 5.2 Summary of community reasons for sharing carrier information during studies

Reasons for sharing information	*Reasons against sharing information*
In presence of wider screening: child can use information later to support future reproductive choices for benefit at individual and wider family levels *In absence of wider screening:* Services for screening may become available later, before child reaches adulthood Child and family will be prepared and better able to take care of any children affected in future; including limiting harms of 'not knowing' Parents of carrier child will understand own potential risks of having affected child in future; and be in better position to look after child (limit 'harms of not knowing') Less conflict between parents where have child with SCD against background of understanding risk Family can pass information on to others to give wider benefits to community; knowledge as empowerment for individual and society Anxieties associated with understanding risk to future generations are real and should be accepted as part of gaining benefits of knowledge; at least one of parents is also a carrier so should be able to accept as benign condition; asymptomatic nature will limit anxieties Wider awareness of SCD will generate demand for better services, including better individual care and public screening; and reduce stigma towards condition, as has occurred for HIV/AIDS May rely on family stories to assess risk, but unreliable; In Kilifi DHSS area, children who know their carrier status may be able to use this if marry locally? Able to take particular care of health of carrier child?	Creating anxiety about future risks when no certain way of avoiding (unnecessary anxiety) Children and adolescents particularly susceptible to anxieties; concept of good childhood free of worries; emotional lability in adolescents Generating frustration at lack of services, given implications of carrier status; should not share this information until services are more widely available Carrier status likely to be seen as an illness (medicalisation), now or in the future, including seeking care based on supernatural beliefs remembrance linked to misinterpretation as illness; link to use of word 'carrier' in HIV/AIDS High likelihood that parents will not understand, will not remember well enough, or will not be able/willing to pass information on in future to child Stigma for carriers at time of marriage; if information known widely (but option of confidentiality if family choose) Paternal denial of responsibility could generate doubts about marital fidelity
Main emerging values: *Protecting interests of child and family:* Seen as important in lieu of future affected generations; includes idea of not causing unnecessary worry (by sharing information that cannot be used) as well as enabling child to take better care of future interests; tension between paternalism seen as appropriate or over-reaching. *Openness:* Strong value of openness expressed for society to understand carrier status and be able control individual risks and public health burden; in tension with value of protecting interests of child where seen to generate stigmatization of carrier (particularly in relation to marriage). Also tension between value of knowledge for its own sake and others claim of a right not to know, unless public screening available to support use of knowledge. *Researchers' responsibilities:* Openness important for carrier findings in order to maintain trust of community in researchers/research; nature of KGBC in recruiting healthy children at home generates responsibilities to protect families' interests, like a visitor.	
Main influences *Micro:* individual/relationships in relation to stigma; educational/economic status of carrier in relation to understanding and stigma *Macro:* Education/economic status particularly for women to counter stigma in marriage	
Important contingencies: Availability of resources to validate and provide counselling on findings	

Phase Two: Discussing requests for paternal testing for sickle cell status arising during research

6.1 Introduction to the chapter

6.1.1 Background and aims

In Chapters 3, 4 and 5 I described community members' experiences and concerns that sharing information on the genetic roles of parents of a child found to have sickle cell disease (SCD) during research could generate or increase doubts about faithfulness between marital or life partners. The primary concern in this situation was that family breakup would be the outcome, with a vulnerable child and mother being sent back to the maternal home, and where economic and sometimes social support might not be available. At the same time, sharing information on the genetic roles of parents has been seen as central in working against structural gendered harms in SCD, which were viewed as common and serious in the community. In this chapter, I will describe and explore community reasoning on the way researchers should respond to requests for parental testing in a paradigmatically difficult case where these two commitments - to the family and to openness - were seen to be in tension with each other, as well as less straightforward as values.

In Chapter 3, it became apparent that doubts about paternity could emerge in families with an affected child[93] where the parents were given information on its inheritance *but the father denied his genetic role*. It was agreed that some fathers in this situation may ask for testing themselves, as occurred in family F09[94]. Undertaking paternal testing would inevitably incur the possibility of showing misattributed paternity. On this basis, there is a potential for the following sequence of events to occur: sharing of information on parents' genetic roles, generating doubts about paternity, prompting requests for paternal testing, revealing misattributed paternity, and breakup of families. The sequence also illustrates the potential for real tension to emerge between commitments to

[93] Paternal denial of responsibility was also raised as a possibility for children shown to be carriers, although with lower likelihood

[94] See Chapter 3, section 3.2.5

values community members have seen as particularly important; that is, *openness* and the *stability of the family*. This tension could in some cases take the form of a genuine dilemma, reflected in varied views on researchers' responsibilities for sharing information.

The nature of this dilemma could be described as based on a 'slippery slope' argument, premised on the extent of *inevitability* that acceptable actions will lead to unacceptable ones (Hope 2004). By this account, if fathers are likely to request testing, researchers are seen to have a responsibility to respond positively to their requests, and information sharing on misattributed paternity is perceived as both inevitable and unacceptable as an outcome of paternal testing, then a slippery slope argument would work against sharing information on parents' genetic roles. Conversely, if researchers are not seen to have this responsibility, or if disclosing misattributed paternity is seen as either non-inevitable or acceptable, then information sharing on SCD inheritance becomes less of a dilemma for researchers.

Four questions seem likely to inform the extent to which this dilemma might act as a 'slippery slope': How inevitable or likely is it that fathers will request paternal testing? If paternal testing is requested, does this present an obligation[95] for researchers to agree? If paternal testing is undertaken and misattributed paternity shown, do researchers similarly have an obligation to share this result with the parents? Finally, if misattributed paternity is shared with the parents, what impact would this action have on the initial assessments of researchers' responsibilities to explain the genetic roles of parents, given that this was premised - as shown in Chapters 4 and 5 - on the ability to limit harm?

To focus discussion around researchers' responsibilities towards parental testing and disclosure of results, community members were asked to consider the situation of family F09, bearing in mind that testing was not at that time routinely offered to parents in Kilifi, either as part of clinical or research services. The questions for discussion included:

[95] See Chapter 1, section 1.7 (footnote 23) for a description of my use of the terms 'responsibility' and 'obligation' in the thesis

Should researchers agree to undertake parental testing if requested? If so, and if they find a result showing misattributed paternity, should they share this information with either, or both, of the parents? How could these information sharing activities be seen as the responsibility of researchers? What ethically relevant concerns are at stake, how might these be weighed up against each other and what conclusions emerge? In what way should the value of information sharing be seen and justified, and what contingencies and practicalities are raised?

Through an analysis of these discussions, I aim to explore the way that community members considered questions about researchers' responsibilities for testing and sharing results with parents of an affected child, including their perceptions of the likelihood of this request being made, the nature of moral dilemmas that might be created, ways in which these could be weighed up and possible solutions.

6.1.2 Organisation of chapter

To a large extent this chapter will build on the findings and analysis of earlier chapters. Section 6.2 describes community members' perceptions, experiences and values in relation to the questions posed for this chapter. The section begins with reasons given 'against' (6.2.1) and 'for' (6.2.2) paternal testing and disclosure of findings, although the nature of the debates means both sections contain some counter-opinion. In section 6.2.3, I describe the way in which three main values indentified earlier (openness, the stability of families and the responsibilities of researchers) emerged during discussions in this chapter. Data on the influence of contextual issues are included within these sections, with the exception of the influence of HIV/AIDS awareness on attitudes to SCD, discussed separately in section 6.2.4. In section 6.3 I explore the nature of agreement and disagreement within discussions, including the extent to which these are based on perceptions around 'facts' or values, and the conflicts emerging from differences in values, drawing on relevant areas of literature. I also consider the likelihood that fathers will request testing at the end of this section. In Section 6.4, I draw conclusions from this chapter, which ends with Figure 6.2, providing a summary of the main findings.

6.2 Local experiences, values and reasoning on requests for paternal testing

6.2.1 Reasons against sharing misattributed paternity information: Risks of harm to children, their mothers and families

There was widespread shared concern across all groups that testing parents for SCD had the potential to show that the social father of the child was not a biological parent, potentially leading to very serious harm. The risks of harm were seen as heightened where mothers, even without testing, were already vulnerable to such suspicions, as described in some detail in Chapter 3. In fact, the reason that some fathers might request testing was perceived as related to pre-existing doubts about paternity, or at least their responsibility for their child's condition. Whether or not doubts existed previously, the risks of demonstrating misattributed paternity were almost always seen as potentially harmful to mothers, their affected children and their families.

The potential outcomes of such a situation were described differently by different community members, with individual circumstances having a major influence. Perhaps the least extreme version of harm was illustrated by a community member's description that, following disclosure "there will be no peace in that house" (KCR02/P4). But all community members agreed that the harms could include separation of the family, causing potentially very serious hardship to mothers and children who would (as described in Chapter 3) be likely to be sent back to the maternal home. A typical description of this risk came from an assistant chief from an urban area, saying:

> "Definitely if the results are out and the man is not a carrier, and if we give out the results, that home is broken already."(Chiefs 02/P1)

The harms of misattributed paternity were seen as compounded by the fact that the child at the centre of controversy was chronically unwell. As a mother of two affected children pointed out:

> "…he [the social father] won't agree to live with a sick child, he doesn't know where it came from, he won't also agree and that's why he will have to tell you, okay, *where you got this one from, okay, take him back*." (Mot/P2) (my italics)

Even if the mother were not sent away, she and her child could still be adversely affected in important ways. As another mother of a child with SCD explained:

> "I think some might agree for you to stay there but he [the affected child] won't be given the same care as the others...because he [the father] will obviously know that this child is not mine. And since the child will sometimes fall sick, you [the mother] are the one who will be encountering many problems…even sometimes he [the child] might be admitted and paying of the bills are mine" (Mot/P1).

For mothers without independent resources, meeting such costs would be very difficult and almost inevitably compromise the child's care and chances of healthy survival. At the extreme end of reported harms, some community members perceived risks of serious physical assault to the mother. A community facilitator described misattributed paternity as a "*panga* case", where *panga* is the Kiswahili word for machete. Several community members in one community group discussion (KCR03) were adamant that this type of information could not be disclosed, given risks that the mother might be attacked and maybe even murdered. In this group, the prominence of jealousy as a feature of some marriages was linked to "little chance of forgiveness" (KCR03/P3). These very serious harms might also affect a suspected biological father, as suggested by a member of the DHMT saying:

> "It is a very big worry because if circumstantially it does not prove to be that both of them are carriers, then it would probably lead to breakages of families, yeah, and even witch hunting, because you know separating, divorcing somebody is very easy but again in the society they would want to know who the father to this child is." (IDI13)

6.2.2 Reasons for paternal testing and sharing misattributed paternity results

6.2.2.1 The importance of testing fathers to counter paternal denial of responsibility

By contrast with the perspectives outlined above in which some community members argued for the importance of refusing paternal testing or withholding information on misattributed paternity *in order to protect the interests of a sick child, mother and family*, others argued that it was important to agree to these requests *for the same reasons*. Testing fathers, where requested, was seen as beneficial because paternal denial of genetic responsibility was viewed as common, risking serious outcomes which might be minimised by parental testing, even if this might create difficulties in the

minority of families where misattributed paternity was found. In any case, some considered that finding misattributed paternity might not necessarily imply an obligation to share this particular piece of information, whereas those fathers shown to be carriers could be informed and would then 'settle down' and accept their responsibilities. One basic assumption for this point of view was that unfaithfulness was not seen as a common feature of marriage. As a mother of an affected child explained:

> "There, it's important for all fathers to be tested for it to be known, because those who get children outside their marriage are very few. You might get for example in a group of twenty people, maybe it's only one person who went out of the marriage, and shouldn't the others be told?" (Mot/P1)

In fact, as noted in Chapter 4, one of the most strongly expressed concerns of affected mothers included in these discussions was the risk of paternal denial of responsibility, including chronic conflict and breakup. Where separation did not occur after the birth of a first affected child, this was seen as increasingly likely over time if further affected children were born to the couple. Agreeing to test parents, thereby demonstrating the father's carrier status, was therefore considered an important strategy to prevent family breakup, particularly since the father's request for testing was perceived as likely to be underpinned by denial of this role.

In addition to a numerical weighting given to 'faithful' mothers, who were considered by some likely to outnumber their 'less faithful' counterparts, community members not uncommonly drew on their views on marital infidelity as a moral issue. On this account, the interests of a faithful mother should in any case outweigh those of an unfaithful one. For example, a field worker expressed the following view:

> "I also agree that the test should be done, basing it in this way. When it's a couple, I think they are one thing, and if the lady went out and got another child then it means their relationship is not good, and when the man asks for the test, then you can just do the test and this will also show the man that 'my wife is not faithful', and it will assist in other ways, like when this lady is not faithful, then she can bring other types of illnesses in the famil ... Maybe she will get AIDS and brings this (laughs) to the family? So I think the best way is to test and they will know what to do" (IDI07/P1).

As this quotation shows, this judgment of unfaithfulness as morally wrong was often described in relation to the risks of bringing other illnesses into the family, such as HIV/AIDS. Less often, community members described unfaithfulness as an intrinsic moral wrong, for example, by drawing on faith based beliefs and attitudes. These moral views about faithfulness and unfaithfulness are described further in the next section (6.2.2.2).

Thus, overall, the likelihood of harm to the mother, child and family through paternal denial of responsibility was seen by several community members to outweigh those of finding misattributed paternity, supported in some cases by judgments about marital infidelity and the possibility of withholding controversial results, should the need arise. Importantly, this view was expressed by all four affected mothers involved in these discussions, as well as being the predominant view in two community representative groups (KCR 02 and 3) and one group of chiefs (Chiefs 03) and being raised at times by other types of community members. However, amongst those who felt that fathers should be tested where asked, marked controversy remained over whether or not misattributed paternity results should be shared, as I will continue to describe throughout section 6.2.

6.2.2.2 Severe harm may not result: Possibilities of forgiveness and alternative resources for mothers

Apart from the strongly perceived need to address paternal denial of responsibility for SCD described above, community members varied in their assessments of the nature and likelihood of harms that might result if misattributed paternity results were found and shared. Key issues in this discussion were the possibility that an unfaithful mother might be forgiven; and the ability of mothers to manage an independent life adequately if separation did occur. The exact nature of the outcome was seen as dependent on particular aspects of the family's situation, working at individual (micro) and structural (macro) levels, much as in Chapters 4 and 5. These contextual issues are clearly overlapping, but will be discussed under separate headings in the following paragraphs.

i) Love and trust as an influence on forgiveness

As in Chapter 4, many community members continued to describe the particular nature of different relationships, especially the existence of trust, as key in predicting the likelihood that fathers would request SCD testing as well as parents' responses to emergent knowledge. Typically, a community representative expressed the idea that*:* "…maybe they love each other so much they may not divorce...?"(KCR01/P5). Perhaps tellingly, a community representative with an affected child described support for openness in testing fathers and sharing results as far as possible, but when her colleague made a strong point about this, added quietly and seriously: "...only if they trust each other" (KCR02/P2). At another time, she told her own story of having parental roles explained, including the possibility that SCD could be brought into a family through misattributed paternity. Through this experience, she again emphasised the need for trust as the basis for acceptance of this information:

> "The child was tested very well and was found to have sickle cell…it was explained very well to us by the doctor. Even these issues concerning… 'going out' were also explained to us, it was among what was explained to us…but if you do not have trust in the house, this issue [conflict over misattributed paternity] will come up, [because of] lack of trust." (KCR02/P2)

In her family, the father had accepted his genetic role without requesting a test; it is interesting to note however that many relatives on the paternal side were known to have been affected by SCD, but none on her side of the family. A field worker gave a further good illustration of this variation within families, saying:

> "There are so many reasons which can cause that [breakup]. So if it is a family that is well, I mean well tied up, when the relationship is that rich and strong, I think even if they realise that the man is negative and the wife is positive, I mean I'm trying to imply that even if the man learns that this could not be his child, but still if the relationship is a good one, they might discuss and resolve." (IDI07/P3)

ii) Traditional attitudes to forgiveness for marital infidelity

During the course of discussions on the acceptability of unfaithfulness, several community members discussed general attitudes towards this phenomenon within the community, with mixed views on how acceptable it might be. Several community members expressed very non judgmental attitudes themselves, for example that "it's normal for people to be lost" (KCR03/P5) as a reason for being prepared to forgive. Similarly, a local saying about the importance of both remorse and forgiveness for mistakes made was quoted in this context: "kufanya kosa si kosa, na kuregelea, kosa ndio kosa" (*Kiswahili*) (KCR02/P2), meaning, 'a mistake is only a mistake when its repeated.'

Some health managers and chiefs gave a clear indication that unfaithfulness in marriage might traditionally have been seen as more acceptable than is often currently the case:

> "…of course nowadays things are changing, but there was the, not really a leeway, but I think it was like very obvious when a woman moved out with other men, and it was also very normal for a man to move out with other woman… that's why the man can [easily] blame the woman...because *to them that thing has been happening.*" (IDI14) (my italics)

Some of the reasoning that might underlie a traditionally less judgmental attitude towards marital infidelity was also hinted at, as shown in the following three quotations taken from discussion during the refreshment break of a chiefs' meeting. A participant pointed to the very high value placed on children within a family, and the possibility that the arrival of a new child might outweigh other considerations of jealousy or biological relatedness:

> "… but in Giriama there is a saying, a child is a child even if he has been brought from any other place, but he is the one you call a goat[96] or we could say he is the cock. Where is the profit? It is here [in the social father's home]…it's not a big deal!" (Chiefs 03/P2)

His colleague agreed, clarifying that this would apply to children conceived outside marriage as well as the existing children of a woman who is later married to another man:

> "…and then here she has given birth to my first child, the second, and then this third one maybe…she has moved outside wedlock and conceived, still that difference cannot be noticed at all, even the others [those born out of wedlock] are mine." (Chiefs 03/P1)

[96] Meaning that the presence of a child within a family represents a substantial gain to that family, irrespective of parentage

The traditional nature of this attitude towards misattributed paternity was suggested by a chief who linked its prevalence to more rural ways of life and less exposure to education:

> "… it's [misattributed paternity] not a difficult condition as such…I don't know why you [referring to chiefs from the more urban areas] take it very hard, you who are in [town], I don't know you who are educated here in [town] but those who come from our areas, [rural area] they are. . ."(unfinished sentence) (Chiefs 03/P4)

These views are interesting in showing that the concept of fatherhood according to traditional values in Kilifi often encompasses a wider social rather than a purely biological meaning, as has similarly been described from the perspectives of men in western settings (Turney 2005; Ives, Draper et al. 2008). It is possible that the traditional values being spoken of here would be more prominent amongst older fathers; although controversial, older age was often seen to increase the likelihood of men accepting misattributed paternity through these traditional attitudes, as shown by a field worker saying:

> "I'm trying to imagine a situation where it's an old man who has several wives, they wouldn't bother about kids being from another parent, I don't think. Because I know of situations where an old man has maybe four wives and maybe the fourth wife is very young, to an extent that there doesn't seem to be a close relationship, and the young lady gives birth for several years, people will even gauge that these kids do not belong to the old man, and the man still accepts that they belong to him." (IDI07/P3)

Similarly, a group of chiefs (Chiefs 03) described this phenomenon, using the common slang phrase 'dot coms' to refer to younger men:

> Interviewer 2: "…so what you are saying is that really we shouldn't be worrying about this [misattributed paternity], although we may find sometimes that fathers are not the real fathers of their children, that people will be able to accept that information, even if we tell them…?'
> P5: (quietly): "yeah"
> Interviewer: "People will accept the information? (pause) Yeah? How do others see it?"
> P4: "that is a very delicate situation!"
> Interviewer: "it's a delicate situation!" (Laughter)
> P4: "to tell someone that aah, my friend I'm sorry…"
> P2: (interrupting) "…but with old age"
> P4: (interrupting) "… it's okay"
> P2: "it's okay, but with the (pause)"
> P4: (interrupting)… "dot coms!"

A further influence of tradition was described in Phase One and raised by some staff groups in Phase Two. Where, as is commonly the case in more traditional homes in the community, marriage had been planned between the maternal and paternal clans, this wider circle of people

would be drawn into resolving serious issues arising between husband and wife, particularly if this seemed likely to lead to separation. Examples were also given of ways in which the mother's family might provide her with a temporary home at times of conflict, but over time actively seek to resolve underlying issues. In these cases, separation was said to be 'not just a case of what the parents want to do'.

There are two arguments against a view that some traditional values would increase the acceptability of misattributed paternity. Firstly, fathers may have particular difficulty accepting a chronically unwell 'outside' child, whose 'value' may be seen differently to that of a healthy child (see section 6.2.1). Secondly, current awareness of HIV/AIDS seems to generate a more negative attitude towards misattributed paternity, as will be discussed in section 6.2.4. In contrast, and acting in support of 'traditional' tendencies for forgiveness and reconciliation described in this section, many community members described an influence of Christianity in generating a duty to forgive, as described in the next section.

iii) Christian attitudes to forgiveness

In many community groups, the Christian requirement for forgiveness was one of the most strongly emerging themes proposed as a safety net for the potential harms of information sharing. Many community members in this study were actively practising Christians; some therefore felt that fathers would have a Christian duty to at least try to forgive their unfaithful wives, particularly where the interests of a sick child and other children were at stake. A common pattern for this form of reasoning was for it to be raised by a participant at a fairly late stage in the discussion, at which point many community members would recognise and support these beliefs, continuing to explore the relevance of stories from the Bible, sometimes in great detail. A typical such Biblical reference concerned a woman condemned for marital infidelity:

> "He [Jesus] asked the woman, 'where are your complainants?' she replied, 'I don't know'. Then Jesus told her that 'you have been forgiven, your faith has saved you and so go in faith and don't do it again'. Therefore all people have sinned, it's not that only this woman has

committed adultery. All of us should forgive each other. No one is righteous on this earth". (KCR2/P3)

Forgiveness of unfaithfulness as a religious duty was less discussed in other (non community) participant groups, although some Chiefs were inclined to raise this as important. For example, one Chief supported his arguments on the importance of truth telling with a comment of: "But will it be right before God?" (Chiefs 02/P5). Community members who saw the possibility of forgiveness as a tenet of Christian faith always assumed the existence of true remorse by the mother, often in ways that were linked to religious beliefs. As a 53 year old urban community representative explained:

> "I can know very well that that child is not mine but...I will sit with that mother and explain to her...I know that you been cruel to me, you see, and I do not want you to be cruel to me again. As for me now, I have known that this child is not mine...you got this child with another person you yourself know... I have accepted you as my wife to stay... here at home, and I will raise this child you have brought me but...I want to tell you that this should be the last one...you will not go outside and bring me [another child] to educate... just to take my money from my pockets, I won't accept, do you hear? So I have forgiven you...but I don't ever want to hear about another child here, from now ...we are only bringing up this one!" (laughter from the group at this last comment) (KCR1/P4)

The negotiation of forgiveness between husband and wife on this basis was described in detail as a formal process within his church by this community representative, where special meetings of the wider church community could be used to give a wife an opportunity to openly confess and express remorse, in this way helping her husband to agree to reconciliation.

iv) Mothers' resources and social networks

Where forgiveness was not sought or forthcoming, some community members were concerned to point out that not all mothers would necessarily experience serious hardship, and that this would depend on the existence and adequacy of alternative independent livelihoods or forms of support. In this situation, the special responsibilities of mothers towards their sick children were described as providing an important motivation for mothers to seek such alternative support. As one community representative explained: "I will be fighting to protect the health of my child." (KCR02/P5)

Positive views about the ability of some mothers to look after their own interests were largely voiced by community representatives and staff members who were women, relatively well educated and (often) employed. One exception was the community representative quoted above, who went on to explain the presence of other types of social networks that might be available*:*

> "If I see my partner has become angry such that I suspect he might beat me or say some other words, I won't keep quiet, I will go anywhere to look for assistance… I will make sure that I have got advice from the doctor to explain to me what has happened … so the child can continue living well… [and] there are ways of sorting out these things in our homes, and so we shouldn't be hiding anything... So, talking about the situation, we should be free to talk about any situation that will come up at any time, and a solution will be found…" (KCR2/P5)

6.2.3 Revisiting key values: Protecting families, openness and researchers' responsibilities

In earlier chapters, I have described the way that three key values - openness, the stability of families and the responsibilities of researchers - emerged across discussions. During debates on paternal SCD testing, these values were again invoked, as I will describe in the following paragraphs. The greater levels of tension occurring during these discussions is reflected in much more detailed dissection of specific aspects of these values in comparison to earlier chapters; and to a different way of balancing values overall, as will be addressed in the discussion section of this chapter.

6.2.3.1 The importance of truth/openness

In the discussions reported in the current chapter, information sharing on misattributed paternity generated much more controversy than that emerging around dislcosure of findings on SCD or carrier status in earlier chapters. As a result, values of openness were more challenged[97]. The following paragraphs describe the ways in which openness was seen as important.

[97] For example, see strongly felt reasons against openness in sharing information on misattributed paternity in section 6.2.1.

i) Lying might be found out and lead to more serious outcomes

The strongest argument made in support of openness was the potential for later discovery of information that had been initially concealed, and the belief that this might often lead to more serious consequences than truthfulness. Strong statements of this nature came from every type of participant group, although was not necessarily the predominant view in each group. This possibility of parents seeking testing elsewhere as a 'check' on an earlier result was articulated well by an affected mother:

> "Haven't you seen a person going from one hospital to the other to be tested? He is not sure…let me seek advice from other people. He was tested here and found not to be sick but it has been hidden when he came here for clinic. He goes to another hospital, and is found that he isn't sick at all while here you have been told that you both contribute, you must both have the *chembe chembe* [sickle cell genes] but at the other place he is told that he doesn't have the *chembe chembe* at all, and so how will it be? So I think it's important for him to be told, as this is only one child [in the case study] and you will organise yourself" (Mot/P3)

During one of the chiefs' discussions, this point was also made in relation to their own work with the community:

> "My point is we have to come to the truth, because we are with them for quite a number of years, we are not just there to solve [problems], just by not telling them the truth, and then afterwards when they go and get the fact finding from the others, they get to know that you have not told them the truth, they come to you looking at you in a different face, because we have them, they are our people." (Chiefs 02/P2)

The strength of arguments based on concealed truths being later found out was seen in the fact that it tended to be a 'show stopper'; those arguing for concealing truth could generally find no way to deny this potential eventuality and the risk of more serious outcomes as a result. Those who remained opposed to openness, in spite of the risk of deceit being discovered later, generally drew on arguments concerning the greater short term risks of sharing misattributed paternity findings. The risk of deception being later uncovered was described by some field workers as being particularly serious for researchers, including loss of trust at both individual (family) and wider community levels. At the same time, other field workers thought that refusing to test the father when this was requested would also look as though researchers had 'something to hide'.

ii) Withholding information will encourage unfaithfulness and suggest complicity of researchers

Separate to the dangers of concealing 'unfaithfulness' for trust in researchers[98], a different type of long term danger was seen for family stability. According to this view, put forwards by some community members in all groups, concelament could act as an encourgament to continued unfaithfulness, which might lead to greater family health problems. As a community representative described:

> "...that mother will continue doing that, first she has... brought children who may not be of that place...now she gives him [the husband] burdens from outside, and ... maybe they will have other diseases. (KCR1/P4)

The 'other diseases' being considered were either a reference to increasing rates of SCD or to HIV/AIDS. For example, a chief explained that:

> "Me I'm of the opinion that…the truth should be said, to safeguard the community. In fact if they are told the truth, people now will stop that habit of going out [being unfaithful], probably, and maybe it will assist the government, especially maybe having so many diseases like the HIV/AIDs and some more, so they will be worried, it will be a sort of worry, if once my kid and myself are tested positive with this gene, and the father is negative, then I will be known, that eeh surely something has gone wrong, so they will stop, and the families will come up with no diseases, and they will be safe." (Chiefs 03/P2)

Others agreed that being open about individual cases of marital infidelity could have this (literally) chastening effect on others in the community:

> "Eeh, now you, it shows this child is not mine, maybe I can chase her to go to her home and then when she reaches there, the neighbours may say why is mother so and so at his home, aah she was chased, her husband said the child is not his because of this and this, now the other women will take an example from her!" (laughter from the group)(KCR1/P3)

That researchers might be seen as complicit through hiding unfaithful practices was illustrated by a mother of two affected children:

> "...yes, she will continue with her (pause) [unfaithfulness], many children will be born with such conditions and it will now be a problem…I think it's better for him to be told because the moment you hide [from] him, the mother will know that the doctor blinded him, and so let me do my things here" (Mot/P1)

In this way, some community members saw an important reponsibility for researchers in maintaining openness in relation to findings. In section 6.2.2.2, I described the way that marital

[98] As in earlier chapters, community members at times found it difficult to consider the responsibilities of health researchers and providers separately. This conflation has a bearing on how responsibilities for information sharing might be configured for researchers, and will be discussed in Chapter 7 (section 7.3).

infidelity may have been more acceptable in the past according to traditional values than in recent times. The more judgmental attitudes towards unfaithfulness described in this section seem to stand in contrast to 'traditional' attitudes of acceptance described earlier.

iii) Openness can empower families and the wider community

Some community members perceived a value in being open about misattributed paternity as empowering family members through knowledge, towards individual and societal benefits, although this was a much less common or strongly held value than for information sharing on SCD or carrier status. As a health manager described:

> "… if we deny them the results [of paternal testing], then they will be living in darkness, and it would be as good as not having initiated the process of testing the child." (IDI13)

As in earlier chapters, discussions about the advisability of paternal testing and sharing misattributed paternity results in SCD often prompted reflection on HIV/AIDS control policies, specifically concerning Voluntary Counselling and Testing (VCT) and discordant couple counselling as measures that might be applicable to SCD. Most community members were very positive about the effects of HIV/AIDS control activities; openness in relation to testing and disclosing HIV status were seen to have been instrumental in reducing stigmatisation towards that disorder:

> "…there is VCT [for HIV] going on at the moment, and everywhere people have been singing about it, that both the father and the mother should go for test. We are now living; some of us have HIV/ AIDS… I mean some of us have the virus, and we are living with the virus, and life is moving on, isn't it? Also in that [testing for SCD] situation…if it's the father who is insisting on being tested, he should be tested. Also the mother should be tested. If you are found in that situation, the doctor will explain to you, and if you understand, like the way the HIV/AIDS doctors or counselors explain to us, if you understand after it's explained, you will continue living. Let it be like that situation, so as for us not to be confused, to avoid shutting down the child's future" (KCR2/P5)

Openness in current HIV/AIDS policy was not universally persuasive when applied to SCD. Notably, the group of community representatives who perceived greatest risks of serious harm to mothers through showing misattributed paternity (KCR03) did not consider 'openness' a good way forwards. One member of that group briefly raised the importance of openness in sharing

misattributed paternity results by reference to perceived successes of HIV/AIDS programmes; she was unsupported by other members, and later apparently changed her mind.

iv) An intrinsic value of openness

An intrinsic value of a concept of openness was often hinted at, rather than directly expressed. Probing on reasons for the importance of openness was more likely to lead to expressions of an instrumental nature to this value, as described in the previous sections. The most common way in which an intrinsic importance was described was as a general discomfort with the idea of 'hiding things':

> "if we say that other things should be hidden, will be kept under the mattress, that will be…meaningless, and so the doctor should explain, and the father should be tested if he requests, and the mother should be tested too if she requests. There should be freedom." (KCR2/P5)

Intrinsic support for openness was also occasionally expressed as a faith based value:

> "There is no need of...hiding...what are you hiding? And when we come to religion, we will have gone deeper..." (KCR01/P4)

v) The acceptability/unacceptability of some types of lies

One way in which a value of openness was opposed was through differentiating between different types of lies and their acceptability. Views in support of the acceptability of some types of lies were particularly invoked where the risks of sharing information on misattributed paternity were seen as high, but paternal testing was seen as an important way of helping many families where this would not turn out to be the case:

> "Okay me I'm saying we can't avoid testing to the parents, but maybe for the one who will be coming out negatively we can…tell him that as a carrier sometimes you can be tested and come out negative while you have…some genes. But…we shall be…carrying on checking until we see the genes… because as a carrier, sometimes also you don't notice any symptoms, that's why maybe you feel you are very okay, but…you must be having such a gene in you." (Chiefs 01/P2)

Some chiefs explained that this form of avoiding truth telling was sometimes needed as part of their professional roles in charge of community security. A good illustration of the way that this

could be perceived as 'more than the truth' rather than a straightforward deception is given by a chief:

> "Yeah they would have known the reality that these diseases, this disease comes from the parents as carriers in order to have that sick child, eeh, but *additionally you can say something to add on top of that*." (Chiefs 01/P6) (my italics)

Most community members agreed that concealing or distorting the truth might be justified in some circumstances, although they did not always agree about the exact nature of the circumstances. For some community members, the risk of harms to sick children, their mothers and families of sharing misattributed paternity was exactly the sort of condition that would justify a disortion of the truth. However there was agreement that one absolute precondition for an acceptable lie was that it should be used to help the person being misled:

> "It [lying] will be fine because lying to someone is not cutting them. Lying, you know there is a kind of lie you can protect with, so that you get something to help, and another kind [of lie] where if you follow it, you will go to hell, now this one is not that of hell (all laughing), this one is for saving someone!" (KCR03/P4)

This idea of a 'good lie' being underpinned by the nature of its intended consequences was well illustrated by a health manager:

> "If I think about the outcome, if you think I'm giving the right information but this information is going to destabilize this person, the family, it would cause a lot of issues, then you better know how to, even if you are not going to tell the truth, but something that will assist this person. That is my thinking…when you come to this testing of the husband and all that, and telling the results, I think there we have a lee way. We want to guard against the divorce and other subsequent (pause), yeah" (IDI12)

A community member described this type of 'white lie' as sometimes being a professional responsibility for doctors to support their patients' interests:

> "I want to add something there. Doctors do tell lies, there was a dispensary in X, so when people go there they tell the doctor, 'Doctor I have come, I want you to inject me', you see? When they have a bit of fever, they want an injection, even when doctor examines them and finds it's not an illness that needs an injection, but someone wants to be injected. So he used to use distilled water…Yes, he says 'you want an injection?' when he gives them tablets, they claim the tablets don't cure them, only injections. So he used to give them distilled water also, yes and they get better!" [All Ps laugh]

6.2.3.2 Protecting vulnerable children and family stability

As in earlier chapters, the importance of protecting the interests of vulnerable children and family stability were central to much reasoning around paternal testing. At the same time, counter arguments were most strongly invoked in the discussions reported in this chapter, in the form of a father's rights to information on his own SCD status. Two main versions of values attached to fathers' rights were expressed; the responsibility of doctors/researchers to respect the rights of fathers to knowledge about their genetic status, and the unfairness of fathers being 'tricked' into financial support for children that were not theirs:

> "This man is just...educating children that are not his, ooh my dear, he should be told...for me, I'm saying this, that he should be told, we shouldn't melt [English] this man (others laughing) ...we should not melt him, he should be told... truly, me I see it's better he is told!" (KCR1/P4)

However, fathers' rights to SCD information were also opposed by some community members who explained that they would prefer *not to know the truth* in this situation:

> "If he tells me the truth that will be very good, but things will go crazy when we go home, and when things turn sour, I'm sure my partner won't stay at home, she will go. But because there's that will to keep the family together… even though we are not saying it should be like that, let him lie to me so that I continue to stay with my wife." (KCR03/P3)

As above, a discussion of father's rights (and interests) could easily collapse back into one about the stability of the family of which he was a part. In any case, individual rights were often not a clearly defined area of reasoning since overlaps occurred in talking about individual's rights, their 'interests' and outcomes. In this situation, the greater importance often attached to anticipated outcomes (than individual rights) was illustrated by a health manager:

> "Obviously… [if] the outcome of the results will dictate that either they stay together or they don't stay together…then probably we would be reluctant in subjecting the couple into the genetic investigations…[and] disclosing the results to this couple…I would borrow heavily from the VCT counselling protocol, that if telling the results to your wife…will lead to either separation or probably cutting down some of the support that used to direct to such a person, then I would better not tell you the results." (IDI 13)

6.2.3.3 Researchers' responsibilities

As above and in earlier chapters, perceptions of the way researchers should handle information sharing were strongly influenced by the assessment made of the foreseeable benefits and harms of disclosure. Given the prominence of concerns about the welfare of vulnerable children with SCD and their mothers, many community members perceived that researchers should aim to protect these interests. As the earlier sections have shown, these views prioritising the interests of families were embedded in a wider debate on the nature and likelihood of harmful outcomes, the value of openness and truth, the rights of individuals and the influence of individual circumstances. As I will describe later in this section, there was also variation in views on the domain to which researchers' responsibilities should apply.

Researchers' responsibilities to protect the interests of vulnerable children and their families were perceived as particularly marked where community members also saw the role of researchers as essentially being the same as those of health care providers[99]. Given the seriousness of the outcomes of disclosing misattributed paternity (as perceived by some), this responsibility was seen even at the cost of infringing other moral values that would otherwise be seen as important, such as openness. These community members expressed an opinion therefore that doctors should refuse requests for parental testing, or conceal harmful information where testing was agreed, in order to protect mothers and their sick children. For example, a community representative described, with emotion:

> "The aim of the doctor is not to break up the family….that's not the doctor's aim! His aim is for the family to live in peace!" (KCR02/P6)

In a more measured way, another community representative made the same type of suggestion:

> "My opinion is that if the man insists on being tested, let him be tested but they should come for their results next month perhaps or next week. So before that, you doctors should sit, because you know when you say the truth, you will have caused a fight in the home, so you should look for a way of hiding the truth so that at least there be peace at home, and continue caring for the child." (KCR03/P6)

[99] In Chapter 7 (section 7.3) I discuss the implications of conflation occurring between perceived roles of researchers and health providers.

Most community members saw it as important that, at least, researchers/doctors should not directly raise questions about misattributed paternity (for example, by offering parental testing) and, in explaining the genetic roles of parents, try to avoid generating requests for parental testing as far as possible.

As a counter to the view that researchers have a responsibility to avoid paternal testing or sharing misattributed paternity results, some community members discounted issues around marital relations as being within the sphere to which such responsibilities might apply. This view tended to be held, often strongly, by those who also supported a policy of openness. The issue was raised across the groups of community representatives - where it remained contentious - and by some field workers, but was less discussed by other types of community members. Illustrations are given below:

> " Let him be tested...you have done your part, the results that come out belong to them...your part is to educate him, and he has been educated...the results are not your problem, now that's for him and his family, they will decide own their own the way they will live...(separate) you, you will have done your duty." (KCR1/P2)

> "So I cannot say that the doctor made a mistake by explaining [the truth] to my husband. I am the one who went out [had an affair with another man]. The doctor never told me to go out! Therefore, I shouldn't blame the doctor (laughter from the group). The doctor should be free to explain to me, explain to everyone." (KCR2/P5)

> "…after that I think the parents are the ones who to decide if they want to be tested or not tested, and if it comes to them one is positive and the other one is negative, I think that is not the question for the researchers, that's the question for themselves at home there, you are just here to treat the child." (Chiefs 01/P5)

6.2.4 The influence of HIV/AIDS on perceptions of SCD as 'an epidemic'

The topic of HIV/AIDS seemed to be considered particularly relevant to the discussions on SCD in this chapter because of shared background issues of marital infidelity, and community members' awareness of strategies such as couple counseling used to counter potential harms for HIV discordant couples. During the discussions reported in this chapter, some voiced strong support for the importance of openness based on perceptions of SCD as *an epidemic*, needing control measures

at least as strong as HIV/AIDS in order to prevent future 'catastrophes'. This was often linked to seeing SCD as a new disease, as well as one increasing at a very fast rate:

> "…they should be told [about misattributed paternity results], because this is a new era, and there are new diseases arising." (Chiefs 01/P3)

> "I was just thinking…this condition, it's…becoming common nowadays… maybe it was there before but maybe in small traces. But because people were not informed, the carriers kept on marrying each other, now there are more people who are born positive with this condition. So if the test is done and information is given to these people, and maybe it could be brought on board like other illnesses, where health talks are done in the dispensaries…if they become fully aware, they may be able to go for testing before marriage, and this will stop or rather discourage that situation where carriers marry each other, more and more, more and more." (IDI07/P3)

Chiefs similarly took HIV/AIDS programmes as a positive model for SCD. An experienced chief from an urban area took an extreme line in making this comparison, suggesting that SCD programmes should be implemented in a more aggressive way than was done at the outset of the HIV/AIDS[100] control activities:

> "I want to cite on HIV and AIDs, where the first case was identified sometime back, but everybody was closing his eyes until a time when we thought 'aah now this one should be declared a national disaster', when very many already had been affected, and infected. Then sometime again over the years, they have said now it's a total war…where I feel like when I go there it is to do or die! And I'm not given any weapon to go and fight that… enemy, but I'm given…a set of condoms. Can you please go and fight? The other bit of me to fight is VCT, [but] then they said it is voluntary. I either go or I don't go, who will say what, because it is voluntary…To me I would have felt that if the government is serious on this issue it would have changed this from VCT to CC &T, compulsory counselling and testing. So it's the same case here…Now I'm linking that one with sickle cell, that it should not be voluntary but compulsory testing and counselling, these people should be tested so that we identify earlier cases of sickle cell disease, and management can be done at those levels, and I'm sure we will be out of it!" (Chiefs 03/P1)

6.3 Reflecting on agreement and difference on paternal testing and sharing misattributed paternity results in SCD

In this section I explore the way findings described in this chapter can be mapped out as perceptions about 'facts' or values, to clarify the nature of difference and agreement within the discussions (Macklin 1999). This process of mapping is shown in Figure 6.1, which illustrates the much greater degree of divergence of opinion on whether researchers should agree to paternal

[100] Current HIV/AIDS testing policy in Kenya supports 'opt out' testing to all users of government health facilities as well as offering VCT services.

requests for testing compared to sharing information on either SCD or carrier status. Three overall options emerged for responding to requests for paternal testing, in contrast to the two broad positions adopted in earlier chapters ('for' and 'against' disclosure). These three positions were: refusing to test (position #1); agreeing to test and sharing all results, including those indicating misattributed paternity (position #2); and agreeing to test and sharing true paternity outcomes but withholding misattributed paternity results (position #3).

As in earlier chapters, particular attention will be paid to the views of an important minority, that is, mothers of children with SCD to ensure that the interests of this particularly vulnerable group are represented in processes of balancing different viewpoints, a recognised challenge for deliberative forms of empirical ethics (Parker 2007b). The diversity of opinion was not notably representative of group type within these discussions, as shown in Figure 6.2 in Appendix E5[101].

From Figure 6.1, many of the agreed values emerging from community discussions in this chapter reflect the high perceived importance of outcomes as a value, as described in Chapters 4 and 5. Unlike earlier chapters, where conflicts in values were largely underpinned by perceptions of the probability of different outcomes, conflicts in values (#1, 2 & 3) in these discussions additionally concerned fundamentally diverging views around issues of sexual fidelity in marriage, the importance of forgiveness and the boundaries of researchers' responsibilities. Further, there was conflict in the way that researchers' responsibilities towards truthfulness and openness were seen in situations where misattributed paternity was found. One implication of these conflicts is that previously strongly supported values concerning the *interests of vulnerable children and family stability became challenged*, such that this privileging was sometimes only directed towards families where the mother could be shown to have been 'faithful' through paternal testing.

[101] This figure was developed to represent the findings from each group in more detail, given that different types of groups (staff, health managers, community leaders and community members) were purposively selected in this study to explore anticipated diversity of opinion.

Figure 6.1 Agreement and disagreement on facts and values on paternal testing

Facts agreed [a]

1. Vulnerable children (affected by SCD), mothers and families are at risk of serious harm because gendered blaming and paternal denial of responsibility are common features.
2. Testing parents and sharing results can undermine paternal denial of responsibility, if misattributed paternity not shown, and will benefit some families.
3. Testing parents and sharing results can lead to severely harmful outcomes for the mother, child and family if misattributed paternity shown.
4. Counselling processes can increase the acceptability of sensitive information and minimise risks of harm.
5. [For some] The rate of increase of SCD in community is comparable to HIV/AIDS epidemic.

Values agreed [b]

1. The interests of sick children and vulnerable mothers are particularly important and should be protected.
2. The interests of family members are related to the interests of the family as a whole; family interests are particularly important and should be protected.
3. Openness and truth are important both intrinsically and instrumentally, with particular emphasis on empowerment through knowledge, and progress as societal good. Living in darkness is not good.
4. Doctors/researchers have a responsibility not to harm/to prevent harm to their patients/participants.
5. Doctors/researchers should not become complicit in deception.
6. Individuals have a right to information generated by medical tests about their health.

Facts contested

1. Whether there is a low or high likelihood of severe harm resulting from testing fathers and sharing misattributed paternity results [c.]
2. Where there is a low or high likelihood of reducing paternal denial of a genetic role through sharing information on true paternity.
3. Whether or not fathers who request a SCD test are likely to have doubts/rejected their genetic role.
4. The actual rate of marital infidelity

Values contested

1. Marital infidelity is/is not intrinsically wrong (most emphasis on instrumental values; influence of tradition and religion).
2. Forgiveness is/is not a virtue; supporting traditional/religious values vs. fostering unworthy/unhealthy relationships.
3. Doctors'/researchers' responsibilities do/do not include domestic issues such as misattributed paternity.
4. *Balancing values* # 4 & 5: Distorting/withholding the truth is/is not acceptable when not primarily done to protect the person implicated.

Notes:

a -The 'facts agreed' were widely held across all groups, apart from fact #5, relating to SCD being seen as an 'epidemic'. This perception was not disputed, but neither was it universally raised.
b - Values #1 to 3 were all widely and strongly held, and as the findings show, were also at times in conflict. 'Agreed' values #4, 5 and 6 were also commonly held, but less strongly.
c- Depending on a number of factors, including nature of relationships, extent of gendered attitudes (many circumstantial issues feed into this, including age, patterns of SCD observed, no children), economic circumstances, ability to forgive (personal, traditional and faith based value), traditional mechanisms (polygamy, involvement of wider family).

Across these agreed and diverging views, I describe below the ways in which perceived facts and associated values underpinned the three overall positions outlined at the start of this section: refusing testing, testing and sharing all results, and testing and withholding misattributed paternity results. Community members did not comprehensively or inflexibly support particular positions; individuals contributed some - not all - of the points listed under each option, and some moved between positions over the course of discussions.

Position #1: Those who would refuse to test fathers on request:

- Assessed a high risk of severe harm in the short term for some families where misattributed paternity was shown, and considered this more important than the potential benefits for other families of showing fathers' (true) genetic roles; supported by a concern that misattributed paternity may be quite common;
- Additionally considered that dishonesty would be unacceptable given the likelihood of researchers being 'found out' and subsequent loss of trust;
- Placed a high value on family stability; and doctors' and researchers' responsibility to prevent or minimise harms to patients or participants as far as possible. In addition, gave some – but limited – support to values of openness, but only in as much as these supported the interests of the family.

Position #2: Those who would agree to test fathers and share all results:

- Prioritised the benefits of demonstrating paternal responsibility in families with 'true' paternity over the harms of showing misattributed paternity in other families; and in any case, assessed a low risk of severe harms for families where misattributed paternity was shown, including that risks are short term and could be mitigated through effective counselling and communication. Also held that misattributed paternity is less common than true paternity;
- Placed a high value on values of transparency and openness, with a particular focus on researcher-participant trust and knowledge as empowerment towards societal progress. Supported by seeing that doctors' and researchers' responsibilities not to harm do not extend to

social issues, such as unfaithfulness in families; that women who are unfaithful should not be considered as important as those who are faithful[102], given risks to their families from HIV/AIDS; and (less often) fathers' rights to information about their health;

- Perceived that all results, including misattributed paternity should be shared because lack of transparency is likely to be found out and lead to serious levels of mistrust between participants and researchers, even if done with good intentions.

Position #3: Those who would agree to test fathers but only share findings showing true paternity (withholding misattributed paternity results):

- Sought a compromise between positions #1 and #2;
- Agreed with position #1 on high risks of severe/ immediate harm to vulnerable children, mothers and their families through sharing information on misattributed paternity; and with position #2 on the importance of paternal testing to families with 'true' paternity in showing fathers' genetic roles;
- Also agreed with position #2 that withholding information on misattributed paternity risked loss of trust but this was outweighed by values related to the interests of the family;
- Therefore agreed with position #1, that researchers should uphold the responsibility to prevent harm to families, outweighing their responsibilities to transparency in cases where harm was a likely outcome.

6.3.1 The work of facts and values: Supportive and conflicting

Community members were aware of and challenged by understanding that either testing parents or refusing to test parents could lead to severe harms for vulnerable children, mothers and families, particularly where misattributed paternity was shown. As in earlier chapters, disagreement on the probability of outcomes of testing fathers and sharing the results often reflects agreement that these

[102] By 'faithful' I refer to mothers who were perceived as having been faithful through paternal testing by community members; the converse applies for mothers described as 'unfaithful'.

would vary between individuals, depending on personal or family characteristics and the wider context[103].

An important emerging new perceived 'fact', which was stated strongly by some and not generally opposed by others, concerned the nature of SCD as a dangerous and new epidemic. This misconception led to a prioritisation of openness in approaches to 'controlling' the disorder as a public health requirement. Greater awareness of the true nature of this genetic condition would be important to rebalance this particular assessment.

Additionally, there were important differences in the way that agreed values were balanced against each other, which appear to be underpinned by fundamental differences in the types of values supported. The main conflicts in *balancing agreed values* occurred between prioritising *family stability* or *openness* (as a societal good); and in researchers' responsibilities to families to *avoid deception* or to *prevent harm.* The former dilemma was shown, for example, by a community member's opinion that although some women may lose out through paternal testing showing misattributed paternity, and subsequently being sent back to their maternal homes, others in her home would realise from this 'lesson' that they should not be unfaithful, leading to stronger healthier families.

Two sets of values, around which there was fundamental disagreement, seem to underpin these conflicts in balancing. These concern views about the unacceptable nature of marital infidelity (such that the interests of such mothers would be impacted, for example); and about the boundaries of researchers' responsibilities between medical and domestic domains. The former distinction plays an important role in balancing family stability against the benefits of openness; while the latter seems key in weighing up researchers' responsibilities to avoid deception and maintain transparency.

[103] For example, the possibility of forgiveness for marital infidelity emerged as an important influence, through an intrinsic quality of some individuals and relationships, a Christian virtue and a feature of traditional attitudes to fatherhood in Mijikenda culture[103]. At the same time, jealousy was posed as a directly conflicting attitude to forgiveness, and also described as 'typical' of some marriages in Chapter 4.

6.3.1.1 Balancing the interests of 'faithful' and 'unfaithful' mothers

The perceived importance of openness in countering paternal denial of responsibility and promoting public health interests meant that some community members believed requests for paternal testing should be met, even at the risk of showing misattributed paternity in some cases. The central judgement made was that the interests of 'faithful' mothers - thought to be a majority - should outweigh the minority where 'true paternity' could not be confirmed. The values expressed underlined the predominance of the interests of a presumed majority of mothers; and a view that unfaithfulness was in any case to be discouraged given the risks of associated infections like HIV/AIDS. This attitude contrasts with that expressed by other community members about norms of forgiveness for marital infidelity, drawing on religious or traditional beliefs.

Overall the negative attitudes towards sexual infidelity in marriage in Kilifi seemed to represent a strongly instrumental nature to this judgement that may differ from more implicit notions of wrongdoing described elsewhere for misattributed paternity. In Australia, for example, the 'demonization' of implicated women (particularly in the media) has been described and critiqued by articulating a need to "...abandon a view of accidental pregnancy and paternal confusion that sees them in moralistic, judgemental, and politicised terms and instead consider them to be unintended outcomes of the human condition; that is, human mistakes made by the actions of women and men in the context of intimate relationships." (p245) (Turney 2005)

The complex assessments of misattributed paternity made by community members in Kilifi[104] certainly seem to draw on judgemental and politicised versions of misattributed paternity, but the moralistic element seems much less prominent. I note, illustratively, the opinion of a community member that 'a mistake is only a mistake if it is repeated'.

[104] Drawing on background cultural conditions of jealousy, gender inequity and the high value placed on children; more contemporary influences of Christianity on forgiveness and epidemics of sexually transmitted infections on fear; and on recognition of the role of individual characteristics and relations,

Instead, where judgemental views were expressed, these seemed to fit most closely with a utilitarian approach of weighing up benefits and harm to all concerned, and importantly taking into account the public health benefits of discouraging marital infidelity. This included the assessment that misattributed paternity was 'not very common'. From a utilitarian perspective, it would in fact be very difficult to make an assessment of the balance of harms and benefits of paternal testing where very severe harms (including death) may be a possible outcome. The rate of misattributed paternity in this community is not known. Studies on rates of misattributed paternity have shown widely differing rates within different communities, so that extrapolation would be difficult (Turney 2005; Young, Kim et al. 2009). As others have noted, there is little empirical evidence on the outcomes of disclosing misattributed paternity to couples, where this arises incidentally during research or medical care (also see next section) (Ross 1996; Lucassen and Parker 2001; Young, Kim et al. 2009). However, it seems reasonable to accept the consensus of community members on the existence of severe risk following disclosure.

On this basis, it seems that although the main form of reasoning drawn upon to support prioritisation of the interests of 'faithful' over 'unfaithful' mothers is based on a utilitarian approach (linked to HIV/AIDS prevalence), it is not at all clear that risking harm to the latter group of mothers would support the types of gains anticipated for openness. However, this unresolved difference in the way that values of openness and the stability of particularly vulnerable families were balanced remains a feature of the community discussions in this chapter. In Chapter 7, I will go on to consider the implications of this divergence in formulating responsibilities for researchers to agree to paternal testing and share misattributed paternity results.

6.3.1.2 Withholding misattributed paternity results to protect the interests of 'unfaithful' mothers and their children

Amongst the differences in values described in the previous section, some community members strongly felt that the best option to avoid potential harms to affected children and their families would be to test fathers where requested but withhold information on misattributed paternity. Some

community members were very creative in thinking of ways in which feedback of results could preserve truth as far as possible while avoiding disclosure of misattributed paternity. With few exceptions, there was general acceptance that 'good lies' of this nature, used to help the person deceived, would be acceptable. The main counter to withholding results was the risk of later discovery of the truth, in turn leading to loss of trust between the family and the researcher[105]. In highlighting the notion of trust, community members are pointing to a key concept in ethical relations between researchers and research participants[106] which will form a main focus of interest in Chapter 7. At this point, I note that withholding misattributed paternity results could work to undermine trust at the individual research-participant relationship level, and also - through the spread of information across communities described as common - could come to break down trust between communities and researchers in general.

In general, much of the biomedical ethics literature on misattributed paternity concerns the incidental generation of this finding during medical care, genetic testing or research. The situation discussed in this chapter - of a father whose child is a research participant requesting genetic testing - is clearly different to that of generating incidental findings about a participant in any of these other circumstances. The main researcher-participant relationship in the latter situation is with the parent, while in the Kilifi case it is with the child. The findings of misattributed paternity have already been generated in the latter situation, whereas in the Kilifi case, testing to show this result is being requested. There is a much smaller body of published work considering paternity testing on demand, largely through the commercial sector, which parallels some aspects of father's requesting testing for SCD in Kilifi, as will be discussed later in this section. However the commercial setting seems also to present a major difference in terms of the way responsibilities to undertake testing might be understood, for example, as described by the partial entrustment model for researcher-participant relations in Chapter 1 (1.8.3).

[105] In the short term and locally, this seems an unlikely outcome since testing is not currently widely available in Kenya. However, progress in health services for SCD - after all, a goal for researchers working on the disorder - may bring changes locally, and these limitations are in any case particular to this setting.
[106] Also see Chapter 3, section 3.4.3.

Where misattributed paternity is an incidental finding in genetic testing, the consensus of guidelines, according to Lucassen and Parker (2001), is that husbands should not be directly told the result on the basis that 'genetic testing should not be used in ways that disrupt families' (Lucassen and Parker 2001). Two large scale international surveys of attitudes to disclosing misattributed paternity findings in genetic testing showed that a great majority of professionals believe that such results should not be disclosed, although the later survey also indicated that a majority of women surveyed in the USA in fact preferred that the social father be told (Wertz, Fletcher et al. 1990; Wertz and Fletcher 2004). Lucassen and Parker (2001) describe the case against disclosure of misattributed paternity based on breaching confidentiality of mothers; dissuading others from testing where this outcome may be a result; and the pressure that misattributed paternity knowledge has for couples, including the possibility of violence and breakup, leading to obligations of non disclosure based on duties of non maleficence.

Reasons for disclosure are described as avoiding deception and associated loss of trust; ensuring that accurately informed choices can be made in future; and challenges to the autonomy of the parents, leading to charges of undue paternalism on the part of the doctor/researcher if this information is withheld. Ross (1996) highlights similar issues in the dilemma of misattributed paternity as an incidental finding in genetic testing, and like Lucassen and Parker, breaks the trend amongst guidelines by making a strong case for disclosure (Ross 1996). The controversy in Kilifi is well reflected in aspects of this analysis of the case for and against disclosure of misattributed paternity findings in genetic testing. Of particular relevance to arguments in Kilifi are the concerns that sharing information may generate conflicts, violence and family breakup; and that non disclosure involves deception and may lead to loss of trust between participants and researchers in future.

In relation to the literature on paternity testing on demand, earlier described as showing some parallels with requests for paternal SCD testing in KGBC, Davies' (2007) review of the 'Changing Face of Misattributed Paternity' points to the rise of such demands through the private sector, with

strong media driven public interest in Europe and the USA (Davis 2007). Commercial paternity testing is in fact likely to be a much more widespread international phenomenon; I note that the private company referenced in Chapter 5 offering SCD testing in the Middle East also offers paternity testing, 'without the need for maternal samples' (Eastern Biotech and Life Sciences 2011). Turney (2005) also indentified the role of the media in generating unjustifiable forms of 'moral panic' about misattributed paternity that fuel demands for testing (Turney 2005). Her concluding comments are helpful in considering what might need to be done in the face of such demands: "What is needed is a careful rethink about how we deliberately and meaningfully deal with biological knowledge about paternity when it does not match social arrangements. To that end, initiatives that break down barriers between feuding couples, rather than building them up and strengthening them, are urgently needed. Finding non judgemental pathways to honest, open access to testing, coupled with support for all parties, and a broader definition of paternity should be a priority." (p245)

Whether or not responding to such demands are seen as a responsibility of researchers, where paternal demands for SCD testing arise, the response to such requests might usefully draw on these concepts of strengthening relationships, openness and providing support for all parties. The HIV/AIDS approach to couple counselling may act as an important model for such a process in developing countries. As in the previous section, however, the issue of withholding misattributed paternity results (after testing) remained an unresolved moral dilemma for community members across these debates that will be taken forwards to Chapter 7.

6.3.2 Is paternal testing likely to be commonly requested?

In my introduction to this chapter, the question of researchers' responsibilities for paternal testing and information sharing was linked to a potential slippery slope argument about the inevitability that acceptable actions (such as information sharing on genetic roles of parents) would lead to unacceptable ones (such as being obligated to test fathers and sometimes disclose misattributed

paternity). To a large extent, this question hinges on the extent to which the actions of agreeing to test and sharing information are seen as having some form of moral imperative. However, a potential slippery slope is also a less convincing concern if fathers are unlikely to request testing in the first place, and analysis of the discussions provides an indirect commentary on this possibility.

When asked directly, community members' views on the likelihood of fathers requesting a test were very mixed, suggesting at least the possibility of a 'slippery slope'. However, militating against this outcome is the fact that across many discussions, particularly those involving community members, understanding that the genetic roles of parents *could* lead to increased paternal suspicion of unfaithfulness was often quite a vexed issue, characterised by confusion and contradiction. The strongest understanding of the likely outcome of explaining the genetic roles of parents was that this would *remove* fathers' doubts about their paternity, not that this information might increase suspicion. It was only after prolonged discussion and explanation, and sometimes not then, that many community members recognised the genetic risk of 'outside fathers'. Amongst the group who had difficulty in understanding how unfaithfulness could be linked to a genetic risk, it was instead intuitively assumed that where fathers of sick children had doubts about their wives' faithfulness, this would centre on the possibility of a sexually transmitted infection, particularly HIV/AIDS, or the traditional condition of *chirwa* (see Chapter 3). Accordingly, explaining that SCD was neither of these conditions, but was in fact inherited through the family line across earlier generations was seen as a powerful way of countering paternal doubts about misattributed paternity.

6.4 Conclusions

In this chapter, I describe the third set of findings from Phase Two of this study, exploring the views of community members in Kilifi on the way that researchers' should respond to *requests for paternal testing*, when they arise following disclosure of SCD in a child participating in research. The possibility of such requests being made was raised in Chapter 4 and 5, during discussions on

sharing information on the disorder and carrier status in children with their families. Key chapter findings include:

- There was greater divergence of opinion than around sharing information on SCD or carrier status, across all types of community members, with difference mainly arising from varying assessments of the likely outcomes of information sharing, the principles or values associated with the predicted outcome and balancing of these values against each other;
- Some, but not all, divergence of views can be understood through community heterogeneity and the interplay between characteristics of individuals, relationships (particularly the role of forgiveness) and the wider structural context within which individuals operate and relationships are built and sustained;
- As in earlier chapters, relatively low importance was attributed to individual rights, in this case, a father's rights to information, particularly where this conflicted with the interests of the family; and researchers' responsibilities tended to be aligned with reasoning about the importance of a duty of care towards vulnerable children and their families, or towards openness;
- Unresolved issues in balancing values concerned:
    - Prioritising the interests of mothers whose children with 'true' paternity (given risks of paternal denial of responsibility) and wider public health interests of 'faithfulness' (given the HIV/AIDS pandemic) over those where misattributed paternity was shown; as supported by all mothers with children with SCD.
    - The acceptability of testing fathers and withholding information on misattributed paternity where this was shown, countered by concerns that deceit may lead to later loss of trust.
- For some community members, this moral dilemma (in deciding whether to protect the interests of an perceived majority of families - where true paternity is shown - or those where

paternity is shown to be misattributed) began to break down the very high value consistently placed on family stability in earlier discussions.

- Misconceptions about SCD as a new epidemic and positive views on the supportive nature of couple counselling, both drawing on current HIV/AIDS policies, illustrate that genetic screening activities can both be challenged and supported by parallels drawn with this infectious pandemic.

In conclusion, where a child has been found to have SCD during screening in a study, ethical reflection on community reasoning in this chapter shows that although views on how researchers should respond to requests for paternal testing were mixed, arguments in support of agreeing to test and *either sharing or withholding* misattributed paternity results were importantly undermined by opposing positions. Combined with reasoning about the low likelihood that requests for paternal testing will follow information sharing, a 'slippery slope' argument on the relationship between information sharing on parents' genetic roles and harm generated by disclosing misattributed paternity therefore *seems unlikely to be relevant for current research practice*.

However, there are outstanding differences of opinion within the community that cannot be resolved at this stage. Further analysis of the two main unresolved conflicts in values, and the role of resources seen as important to paternal testing, will be taken forwards to Chapter 7 in framing responsibilities for researchers. An overarching conclusion, however, is that studies that include genetic testing in settings like Kilifi should be aware of the possibility of paternal denial of genetic responsibility, and support strategies to work against this potentially harmful influence where possible.

Figure 6.2 Summary of community reasoning about paternal requests for testing

Refuse paternal testing	*Agree to test and share all results*	*Agree to test/ withhold misattributed paternity*
Stability of family *where misattributed paternity* shown is likely to be harmed Risk of sometimes severe harm to mother/accused biological father where misattributed paternity; influence of traditional attitudes of jealousy Openness/truth important: If testing done, researchers must share all results because lying might be later found out and lead to more serious outcomes – loss of trust in researchers/ research	Stability of family *where true paternity shown* will be protected against risks of paternal denial of responsibility Stability of family *where misattributed paternity shown* will not always be seriously harmed: • Possibility of forgiveness, based on: personal qualities of individuals/relationships; faith (Christian); traditional attitudes towards children/marital fidelity • Role of wider family to support couple • Role of 'couple counselling' (HIV/AIDS model) key Openness/truth important: • Lying might be found out and lead to more serious outcomes – loss of trust • Withholding information suggests complicity of researchers – loss of trust • Withholding information will encourage unfaithfulness, increase rates of HIV/AIDS • Being open will act as 'lesson' to be faithful • Intrinsic/faith-based value of truth • Form of empowerment of individuals (moving out of darkness) as well as society Misattributed paternity is not common so that more families will be protected by testing and sharing results than by refusing to test Family break up gives opportunity to build new healthier families?	Stability of family *where true paternity shown* will be protected against risks of paternal denial of responsibility Stability of family *where misattributed paternity shown* will be protected Risk of sometimes severe harm to mother/accused biological father *where misattributed paternity* shown likely to be removed. Although lying should generally be avoided, lies that aim to protect the person lied to are acceptable; currently seen as part of professional role (appropriate paternalism) Many fathers would not want to know.
Main emerging values: *Protecting the interests of child and family:* Some disagreement on this value when applied to families where misattributed paternity shown, given greater value placed on outcomes for other families, underpinned by strong concerns about the impact of paternal denial of responsibility and perceived public health importance of 'faithfulness' in marriage from HIV/AIDS programmes. Concept of family breaks down into 'faithful' and 'unfaithful'. *Openness:* Conflict between values of openness as empowering 'faithful' families/wider society (with main focus on prevention of HIV/AIDS at societal level) and interests of children/families with misattributed paternity. Importance of protecting families also raises conflict over value of openness as transparency, with disagreement over whether acceptable to withhold results showing misattributed paternity after paternal testing, including a right not to know. *Researchers' responsibilities:* Not agreed, given conflicts within and between values of protecting families and openness, as above.		
Main influences: *Micro:* Individuals/relationships; traditional and faith-based influences on attitudes to fatherhood, jealousy and forgiveness; formal education/economic independence and social networks of mothers; observed family patterns SCD *Macro*: structural gendered influences; economic hardship; supernatural paradigms for health beliefs		
Important contingencies: Communication & counselling: As before, with particular importance given severity of risks in some		

CHAPTER 7

Framing responsibilities of researchers towards information sharing on sickle cell status during the Kilifi Genetic Birth Cohort study

7.1 Background and aims of chapter

In Chapter 1, against the background of a dilemma around sharing sickle cell disease (SCD) information in a genomics study[107] in Kilifi, I described the broad aim of this thesis to develop appropriate responses to important ethical questions about disclosure of these findings, including the responsibilities of researchers. Included in this aim was an exploration of particular empirical methods in normative reflection, as a potential approach to community consultation.

My analysis of findings from Phase One of this study - reported in Chapter 3 - identified three central and morally important questions for studies that generate sickle cell disease findings in malaria endemic settings. In Chapters 4 to 6, I described community views and reasoning from Phase Two about these questions. In this chapter, I reflect on the findings from both phases of study and relevant areas of literature to describe morally important features of this dilemma, and to set out my reasoning and conclusions on the nature of researchers' responsibilities for disclosing SCD findings in this situation. This chapter therefore addresses the second, and first part of the third, specific objective of the thesis (Chapter1, section 1.5) as follows:

- To build a 'bottom up' normative analysis of the moral concerns identified, including the ways in which the perceived value of sharing SCD findings in research can be taken to inform responsibilities for researchers;

[107] The Kilifi Genetic Birth Cohort study

- To propose specific recommendations on sharing SCD information (for affected children and those found to be carriers) for the genomics study, including their ethical basis; and key considerations in planning for SCD information sharing in other types of studies and different malaria endemic settings[108].

In the move between describing community perspectives on the importance of sharing SCD information and formulating responsibilities for researchers to undertake this activity, I describe my reasoning in a series of arguments. The first argument is presented in section 7.2, a major component of overall reasoning, where I propose two main ways in which these responsibilities can be seen as specific to researchers. The first way relates to the literature in Chapter 1 on researchers' responsibilities for ancillary care (section 1.8). The second follows through the emerging theme of trust in researcher-participant relations.

The next stage in my reasoning (section 7.3) explores the influence of a common tendency for community members to *conflate* the roles of researchers and health providers. In section 7.4, I describe three main challenges to assumptions that importantly underpin community perceptions of benefit in sharing SCD information. These are: the feasibility of effective communication; the (unresolved) risk of harm from generating requests for paternal testing[109]; and the core issue of the availability of resources needed to achieve expected benefits. In the final step of my argument, I propose an influence on researchers' responsibilities from my analysis of important values emerging from the discussions (section 7.5). Finally, in section 7.6, I bring together key elements of this argument overall to describe my conclusions about these responsibilities in relation to affected children and carriers, including normative conditions.

7.2 How can perceived benefits of sharing SCD information generate responsibilities specific to researchers?

[108] The second part of this third objective will be addressed in Chapter 8.
[109] See Chapter 6.

During Phase Two, the predominant argument for sharing SCD information with families of affected children - and to a lesser extent those of carriers - was a perceived form of benefit that importantly included *limitation of harm*. Community members and researchers recognised that sharing SCD information can only achieve such benefits where certain conditions are met: validation of testing, counselling on the findings and providing at least basic clinical services. Disclosure of SCD information for affected children and carriers is therefore seen as a form of ancillary service, whose extent depends on the nature of alternative existing services.

The literature on researchers' responsibilities for ancillary care in Chapter 1 underlines that the existence of a perceived benefit - even where extremely important - may not necessarily implicate researchers, unless responsibility can be seen as specific to them (Georgetown University Workshop 2008). Researchers have unique obligations not to cause harm to study participants, under a principle of 'non maleficence' and to maximise benefits of participation (Beauchamp and Childress 2001; CIOMS 2002). But there are a number of reasons that these principles do not seem to inform responsibilities for researchers to share SCD information generated during studies.

Firstly, the harms associated with low awareness of SCD were not generated through participation in research. Rather, these were linked to the situation in which research was being conducted. Secondly, while helping families affected by SCD would act as a benefit for those particular families, it is difficult to see information sharing as an obligation towards maximising benefits for participants overall, given that less than 1% of study participants had a child with this condition. Even for carrier status, any putative benefit of disclosure would only accrue, at best, to a minority (18%) of participants. If sharing SCD status findings were seen as a form of benefit sharing, more than four fifths of participants would not experience benefit in this way. Thirdly, there are other ways in which benefits could be planned; these might also take account of the interests of a larger proportion of families in the study.

However, two main ways in which these information sharing responsibilities, as a form of ancillary service, can be seen as specific to researchers are described in the following sections.

7.2.1 A professional (and personal) responsibility to assist

For community members in Kilifi, the reason why SCD information sharing was seen as a specific responsibility for researchers stemmed from the perceived seriousness and urgency of the child's situation. Less clearly articulated, being responsible for generating the findings meant researchers were the only party in a position to assist. Since the SCD clinic in Kilifi was already to a large extent supported by the research centre, researchers could not, for example, pass this responsibility on to another agency, such as the public health sector (Ravitsky and Wilfond 2006). On the basis of a common obligation to assist[110], these features of the situation suggest a strong responsibility for researchers to share SCD findings for affected children. According to Singer's principle (Singer 2011), only an equally important reason not to assist would prevent this need for assistance from becoming an obligation to morally responsible people. This type of common morality could not account in the same way for an obligation to share information on carrier status, given the lack of immediate and certain harm.

Given the nature of SCD information sharing as an ancillary service, professional responsibilities are likely to have greater traction for researchers than those related to a common obligation to assist. The partial entrustment model (Belsky and Richardson 2004; Richardson and Belsky 2004), and its derivations (Georgetown University Workshop 2008), are a common way in which researchers' professional responsibilities for ancillary services have been analysed, as described in Chapter 1[111]. These responsibilities are defined by three main features of research: the scope (in relation to findings); the nature and strength of the engagement between researchers and participants (including vulnerability and dependency of participants); and the costs of providing support, given issues of resource prioritisation (Georgetown University Workshop 2008). If, for

[110] See Chapter 1, section 1.8.2
[111] See Chapter 1, section 1.8.3

example, analysis of a study showed that SCD findings were within scope, participants were likely to experience high vulnerability and dependency in relation to the findings, the study methods involved high levels of engagement between researchers and participants, and providing services would have low cost, there would be a strong argument that researchers have a specific moral responsibility to provide ancillary services, that is, validate testing, counsel on the findings and provide basic care.

Based on features of scope, vulnerability and dependency in this model, community members seem right to find sharing SCD findings an important responsibility of researchers in KGBC towards affected families[112]. For carrier status, the major difference is in levels of vulnerability which - like benefit - was seen as considerably less than for affected children. The harms seen as potentially addressed through sharing carrier information were similar to those for SCD (harms from low awareness), but in the case of carriers these were uncertain and related to circumstances occurring in the future. In fact, the risks for a carrier child of having an affected child in future can be roughly estimated at 0.75/100[113], occurring perhaps 15 years in future. Lower probabilities for carrier risks, as well as their occurrence in the future, influence vulnerability in a way that seems to provide a much less convincing reason for researchers *to have a responsibility* to share carrier information with participant families.

Intensity of engagement in studies like KGBC seems more complex, and researchers and participants viewed this aspect of the study differently. From either perspective, the most apparent engagement was through two home visits, one to recruit the child and take a capillary blood sample, and another to return the result of SCD screening. However, from a research perspective, the study passively followed up each participant's future inpatient records throughout childhood. This component of the study was less visible to - and was considered to have no practical implications for - participants. Even without taking account of this longer term component, study

[112] As in Chapter 4

[113] 18% children in KGBC were found to be carriers. If we assume marriage is a random choice, they have an 18% x 18% chance of marrying another carrier and a 25% chance for each child that they will be affected by the disorder.

field workers described the way that many participants had difficulty in distinguishing between different studies being conducted simultaneously and over time in Kilifi, such that their *perceptions* of intensity of engagement for the research centre *as a whole* was high, including for individual studies with low interactivity. One implication of this difference in perceptions is that it may raise contradictions - at a minimum, for the community - to consider researchers' responsibilities around individual studies and from single perspectives, particularly in busy longstanding centres like Kilifi.

Overall, based on features of scope[114] and aspects of engagement informed by vulnerability and dependency, community members' perceptions of the importance of sharing SCD information generated during research seem to present a much more convincing case for researchers having a positive responsibility for disclosure than in the case of carrier status, particularly where no public services are available for wider screening. An outstanding issue impacting responsibilities in the model is cost; this element will be discussed in section 7.4.3.

7.2.2 Responsibility from trust in researcher-participant relations

The concept of trust has a central role in the partial entrustment model discussed above. During Phases One and Two, the concept of trust within research-participant relationships emerged in a further way that can be seen to influence researchers' responsibilities for SCD, discussed in this section. Community members described a dynamic form of trust between researchers and participants, the nature of which was built and maintained over time, within and between different studies.

Concepts of trust and power take a central role in the social science literature on ethical implications of researcher-participant relations, where research is seen as fundamentally a social activity. By illustration, research participation has been seen as 'joint action or coordination *with*

[114] Scope seems less important for the questions being asked here, since SCD findings are always 'in scope' for studies that use this information as part of analysis. There are other arguments against scope being an important feature of researchers' responsibilities for ancillary care, as described in Chapter 1, section 1.8.3 (Dickert, 2007)

other parties' as opposed to 'an individual's involvement in a matter of events' (Dixon-Woods, Suokas et al. 2009) (p2216). Trust is defined as a relational notion concerning voluntary actions that involves 'an element of risk derived from one individual's uncertainty regarding the motives, intentions and future actions of another on whom they depend' (Gilson 2003) (p1454). Of relevance to this study, trust can be generated at interpersonal and institutional levels, since individuals with whom a relationship of trust might be built can also have a particular role identity. They may, for example, belong to the 'institution' of health care professionals, or carry a role that is institutionally located (Gilson 2003; Dixon-Woods, Suokas et al. 2009). Trust can be influenced by affective conditions, such as emotion and empathy, and cognitive understandings. Trust-building behaviours include technical competence, openness, concern and reliability (Gilson 2005).

The role of trust has been particularly highlighted in informed consent processes (Kass, Sugarman et al. 1996; de Melo-Martin and Ho 2008; Dixon-Woods and Tarrant 2009). For example, ethically important shortcomings in informed consent have been seen through failing to take account of the social nature of researcher-participant interactions in which this process is embedded. As Bosk (2002) proposes: 'Improved disclosure is not a matter of content. Rather, it is one of creating a social context that reinforces content' (Bosk 2002) (pV68). Studies have shown that research participants are strongly influenced by the relationship in which they stand to researchers; the informed consent process can become a secondary consideration in decisions about participation. The role of power imbalances in these relations is particularly important in clinical research, especially that involving seriously ill participants (Kim, 2008). As Miller and Weijer (2006) describe, 'the patient-subject cannot reasonably be expected to act as a shrewd, independent party to a contract, enjoying an arms-length relationship with the doctor-researcher in which both fend for their own interests' (Miller and Weijer 2006) (p545).

Such imbalances of power mean that - in deciding voluntarily to participate in research - participants are to varying extents obliged to adopt an attitude of trust towards researchers, for example concerning their motivations, professionalism and veracity (Bosk 2002; Gilson 2003;

Molyneux and Gikonyo 2005). As a result, researchers have been seen to have a matching obligation to take account of the trust placed in them (Georgetown University Workshop 2008). Most commonly, as above, the role of *too much trust* has been pointed out as impacting voluntariness and understanding of research. In addition, the converse - *mistrust* - presents ethical challenges in research, including for beneficence, justice and the public good represented by research (Molyneux and Gikonyo 2005; Marsh, Kamuya et al. 2011a). The ease with which sometimes hard-gained trust can be lost, leading to mistrust, is well described (Gilson, 2003). Mistrust generates ethical concerns where scientific validity and sustainability of research are undermined; conflict is created within communities around research; and individual access to study benefits is undermined, sometimes with a disproportionate effect within communities. Adapting Bosk's earlier comment, these understandings imply that researchers should create a social context - including due levels of trust - that supports ethical research practice. This influence of trust can be seen to generate moral responsibilities for researchers related to sharing SCD information, as will be described in the following sections.

7.2.2.1 Trust and perceptions of study aims

From Phase One, it is clear that sharing SCD findings was a largely popular activity in KGBC, even though the condition was little known prior to the study. In response to this low awareness, field workers often spent considerable time[115] explaining the condition as part of informed consent processes. In Chapter 3, I described that this focus on SCD screening reportedly distracted participants from, or sometimes replaced, the fundamental scientific question in this study. A diagnostic interpretation, or a view that SCD screening was the *main* purpose of the study, was frequently reported.

Undertaking screening and bringing back the results to each household was often a trust-building activity. Relief about negative results, which occurred in the majority of families, strengthened perceptions of the value of the study. Some parents also experienced relief where the disclosure

[115] In Chapter 3, some field workers described taking 2 hours for such an explanation.

policy dispelled worries about rumours[116]. These positive reactions, based on diagnostic interpretations, or emphases, seem to describe a form of *undue trust* in researchers. It contrasts with field workers early perceptions of *low levels of trust* in this study. Neither form of trust could be described as 'due' in relation to informed voluntary participation. Both illustrate how difficult it can be to achieve valid consent in settings where research is unfamiliar and health needs often unmet.

While the risk of diagnostic interpretations of research provide an important moral challenge to sharing SCD information, where stronger reasons support this policy, this risk may be better seen as an important *perverse outcome* that researchers have an additional responsibility to address, through communication strategies and other aspects of research planning.

7.2.2.2 Trust and expectations of due concern and reciprocity

Gilson's (2005) description of trust includes its nature as 'a voluntary action based on *expectations* of how others will behave in relation to yourself in future' (my emphasis) (Gilson 2005)(p1454). For community members in Kilifi, expectations of due concern and reciprocity seem to underpin elements of trust in this way. The perceived importance of sharing SCD information in affected children is linked to an expectation of disclosure as a form of due concern and reciprocity. Failure to undertake this action implies a lack of reasonable interest in the welfare of research participants.

In relation to reciprocity, agreeing to blood samples being taken from *healthy children during home visits*, for example, led some to expect that researchers would - if needed - help to limit any serious harms shown by the blood tests for the child and family, as a fair exchange. Expectations of reciprocity were also influenced by field workers' roles as members of the local community. Pre-existing relationships, depending on their nature, could strengthen or undermine trust in field

[116] As explained in Chapter 3, section 3.3.1.3, such doubts are fuelled when low awareness of research, much greater familiarity with medical care, and common supernatural explanations for misfortune come together in explaining unknown research activities.

workers (Molyneux, 2010). In family F08, for example, the mother expressed high levels of trust and willingness to comply with a field worker whom she held in high esteem.

Expectations of due concern or reciprocity do not generate responsibilities for researchers to share information, but - from a trust perspective - suggest that researchers should take account of and respond appropriately to these perceptions, for example, by addressing causes of undue expectations. Seeking agreement between researchers and communities on ways in which expectations should be met during research would be maximally trust building, given the transparency and due concern implicit in the process[117].

In KGBC, community expectations of SCD information sharing can be argued as reasonable for affected children, but seem less so for carriers. Notably, expectations of *reciprocity* are equivalent for affected children, carriers and children with normal haemoglobin since all participate in the same way. As shown in Chapter 3[118], a 'negative' result did not satisfy expectations of reciprocity for some participants:

> "…there are parents also asking okay fine my child is negative yeah so how about the child? Why do you just leave the child like that? Okay he participated yes but he is not positive. Does that mean it's all over for him/her? We are [just] waiting for maybe vaccines or maybe for better tabs in future? You see so those already are questions that are lingering in the people's heads, so this issue of study benefits… I think we should also consider immediate benefits…" (IDI08)

Expectations of reciprocity could be seen as an important factor in community demands for return of findings made at the outset of the study, triggering the decision to change the feedback policy to returning results to all homes. This change did not however affect the benefits of participation for families whose children were found to be carriers or have normal haemoglobin. Satisfying demands for reciprocity through returning all findings was a popular decision, but may still leave

[117] I note that a low level of community expectations in relation to reciprocity would not mean that researchers' responsibilities could also necessarily be seen as less, since the exact nature of expectations is clearly influenced by community perceptions about the study and the meaning of participation. Rather, a mismatch between participants' and researchers' views on appropriate forms of reciprocity is a potential cause of loss of trust. Resolving this mismatch may rely on changes to study design or to communication about study design, built on transparent processes of community engagement.

[118] See section 3.3.1.2

room to consider how the balance of benefits and costs of participation for all families could be strengthened.

7.2.2.3 Reciprocity or perceived coercion?

In Chapter 3, I showed that common community perceptions of the research centre as a 'high quality hospital' meant that people's decisions about participation in studies may be influenced by concerns about access to care, as has been described previously in this and other settings (Molyneux and Gikonyo 2005; Masiye, Kass et al. 2008). Field workers perceived some families as influenced by an expectation that participation would strengthen their ability to access these 'high quality' services in future[119], particularly for those families with difficulty in accessing care (section 3.3.1.2). Participating in hope of better access to care suggests a form of reciprocity, based on what might be due to participants in future through their participation in studies now.

The difference between participation linked to expectations of future assistance (which seem almost commonplace in many areas of life) and participation based on fear of withdrawal of future assistance (which is clearly linked to loss of voluntariness) seem likely to sometimes entail quite subtle distinctions[120]. In either case, the underlying influence is the imbalance of power discussed earlier in this section. These effects may occur in any type of research that increases, or is perceived to increase the quality of care available. This is particularly important in countries with over-stretched health resources where failing to access types of care within studies is not only *perceived to* but may in fact have very real implications for health. In the case of information sharing on SCD, the focus on the seriousness and lifelong nature of the condition may increase people's concern to ensure they can access appropriate care in this way. Since the research and the provision of care are both the province of researchers in KGBC, this form of perceived coercion

[119] I gave an example of the latter in Chapter 3, in which the parents in family F07 participated on the basis of a diagnostic interpretation and their concern that refusal would compromise their ability to get 'specialist' SCD care in future if their child developed the disease (see Chapter 3, section 3.3.3.1).
[120] This is same distinction that occurs between giving a token of thanks for assistance, and giving a bribe, without which assistance will not be forthcoming.

has a similar nature to that described for researcher-physicians in clinical research (Kass, Sugarman et al. 1996; Kamuya, Marsh et al. 2011).

Given that the research programme overall, through its collaboration with the District General Hospital, provides significant levels of community benefits[121], researchers' responsibilities to take the potential challenge of perceived coercion seriously should take account of individual studies, like KGBC, and the programme as a whole. This includes communication about access to services during research and as part of community engagement in the programme (Molyneux and Gikonyo 2005; Marsh, Kamuya et al. 2008).

7.2.2.4 Risks of obfuscation in sharing carrier information

In Chapter 5, I described community views on the risk to trust from feeding back *carrier* results in children in a way that is indistinguishable from normal findings[122]. There are several ways in which this policy seems problematic. Firstly, it is a type of deception community members found unacceptable, since the central intention is *not* to benefit the person deceived. Although the reasons for obfuscation concerned important issues, it does not seem likely this would satisfy most people's expectations of truthfulness and due concern in researchers, as well as risking infringement of autonomy.

Secondly, heterogeneity within the community means that the resources needed to ensure benefit in sharing carrier status are likely to be varied[123]. Some families may be able to understand and make use of carrier information more easily than others, including as a result of socioeconomic factors (such as access to private testing) or previous experience. In such cases, *failing to share carrier information*, for example to protect families from unnecessary anxiety, *would seem overly paternalistic.* Concerning heterogeneity, a key group are families with other affected but

[121] see Chapter 1, section 1.2.1.
[122] This policy was based on considering the interests of the community and the nature of the resources needed to ensure benefit in sharing these findings, as described in Chapter 1, section 1.3.
[123] For example, see family F19 in Chapter 3, section 3.3.2, who asked about the implications of the result 'not SCD' for the child's future reproductive risks.

undiagnosed children. Carrier information is important to the child screened in relation to preparedness. It also has the capacity, through information sharing on the disorder, to limit the harms of 'not knowing' for other affected children and the wider family.

Thirdly, where a family believes a child with carrier status is 'normal', this could act to influence the child's future reproductive decisions in harmful ways. If the child is told that she 'does not have SCD', and later comes to learn - for example at school - about SCD carrier status, she may assume she has no future reproductive risk, even if she later knowingly chooses to marry a carrier. The role of researchers in generating harm would seem central if she had an affected child in future on the basis of a misinformed decision. Finally, community members perceived that participants could lose trust in researchers if they later discovered this form of obfuscation. This could occur if a carrier child later gave birth to an affected baby, as above, or if repeat screening in future gave a different result.

Taking these reasons together, the practice of sharing carrier findings as indistinguishable from normal results does not seem to be morally supportable. At the same time, alternative approaches would need to take the limitations underlying the current policy into account, including the availability of resources and the absence of wider government policy and services for SCD.

7.3 Potential for conflation between the roles of researchers and providers

During discussions in Phase Two, I noted that community members often conflated the roles of researchers and doctors in talking about responsibilities for sharing SCD information. In this section, I consider what the effect of this conflation could be upon formulations of responsibility.

The occurrence of some degree of conflation between these roles in KGBC could be seen as almost inevitable. The same confusion is common across other clinical research activities in Kilifi (Molyneux, Peshu et al. 2004; Molyneux, Peshu et al. 2005a) and in many other similar research settings (Marshall and Rotimi 2001; Leach and Fairhead 2006), underpinned by the much greater familiarity of the general population with health care than with health research, alongside the

frequent co-existence of these activities in time and place. The existence of a large literature addressing the nature, underlying causes and ways of addressing the challenges of therapeutic and diagnostic misconceptions of research (Molyneux, Peshu et al. 2004; Leach and Fairhead 2006; Geissler, Kelly et al. 2008) suggests some inevitability in this phenomenon. The mixing of these roles is specifically a risk in studies like KGBC where SCD screening is included (as shown in Chapter 3), and given the provision of services at a clinic run collaboratively by researchers and the District General Hospital. The risk was different within each of the discussions reported in Chapters 4, 5 and 6, as I will discuss later in this section.

In section 7.2.1, I used the partial entrustment model[124] to explore the way responsibilities for sharing SCD information can be seen as specific to researchers. This model also provides an explanation of the difference between these responsibilities for researchers and providers. In the model, different types of relationships are seen to encompass different levels of power imbalance. The extent of the imbalance influences features of trust and responsibility; the level of entrustment the more vulnerable partner must invest is balanced by the level of positive responsibilities the more powerful partner should show towards them.

The relationship between medical providers and patients entails high levels of power imbalance, and therefore generates a high requirement for entrustment by patients (in providers) and strong positive responsibilities for providers (towards patients). This responsibility for providers is seen as down regulated for researchers, where the power imbalance and consequent levels of entrustment and positive responsibilities have all been argued as less (Richardson 2008). However, many commentators have pointed to the difficulty in distinguishing between these different responsibilities for researchers and providers in some situations, both from a theoretical standpoint (Miller and Weijer 2006) and from acknowledging the views of participants (Kass, Sugarman et al. 1996; Molyneux, Wassenaar et al. 2005b).

[124] Also see Chapter 1, section 1.8.3.

Clinical research is the paradigm case in this respect; studies at the SCD clinic in Kilifi district general hospital can be seen as falling into this group. Illustratively, Bosk (2002) writes that 'properly speaking, research and human experimentation are outside the system of care, and therefore, different rules apply. Yet this transition is not easy for either patient/subjects or clinician/researchers to make' (Bosk 2002) (pV68). The resulting likelihood of ethical dilemmas for researcher-practitioners is recognised in the Council for International Organisations of Medical Sciences (CIOMS) guidelines for researchers (2002) which states that: 'Professionals whose roles combine investigation and treatment have a special obligation to protect the rights and welfare of the patient-subjects. An investigator who agrees to act as physician-investigator undertakes *some or all of the legal and ethical responsibilities of the subject's primary-care physician*' (CIOMS 2002) (my italics). In the following paragraphs, I consider the risks of conflation in Chapters 4, 5 and 6 separately.

Sharing information on SCD (Chapter 4)

In the case of community members' perceptions of researchers' responsibilities for sharing information on SCD in affected children, the main reasoning was based on the way potential harms, seen as serious and urgent, could be limited. Where conflation between the responsibilities of researchers and providers occurred in these discussions, the impact on the current analysis is on the understanding of harm limitation as a specific responsibility for researchers. As the principle of harm limitation has moral traction for researchers as well as providers, and community members' perceptions of researchers' responsibilities have, in any case, not been taken as the main reasoning behind their existence in this chapter, the potential for such a conflation to undermine the conclusions in relation to SCD information sharing seems limited.

Sharing carrier information (Chapter 5)

In relation to community members' perceptions of researchers' responsibilities for sharing carrier information, the risk of conflation was much lower across most of the discussions, given that these

concerned healthy children[125]. The case study I used to initiate discussions in Chapter 5 was well understood as a research situation. The main form of benefit for carrier disclosure was seen as giving reproductive choice to the child in future or supporting a form of preparedness that would limit possible harms in the future. The conflation that occurred in these discussions concerned assumptions about researchers' responsibilities to support achievement of this benefit, without consideration of whether the actions needed were more appropriately the responsibility of government health providers, or the wider resource utilisation questions this would raise.

Responding to paternal requests for testing and sharing findings (Chapter 6)

Community perceptions about researchers' responsibilities to undertake paternal testing on request, and disclose findings showing misattributed paternity where these occurred, did generate a fundamental form of conflation between the roles of researchers and providers. In practice, these roles are likely to be close or overlapping for families who participate in studies like KGBC *and subsequently attend the SCD clinic*. Since positive tests are confirmed at the SCD clinic, and family counselling primarily occurs at this site, procedures within these apparently separate areas of activities do in fact overlap. Ideally, the overlap *should* occur for all children found to have the disorder during participation in KGBC. Further, it is likely that any requests for paternal testing will be made to clinic staff, as occurred in family F09[126]. In this case, decisions about the 'responsibilities' of researcher-providers towards children attending the clinic, including those referred by KGBC, should arguably be made jointly by researchers and government health providers *taking account of the CIOMS guidance above*, since the decisions have implications for both the research programme and the district general hospital.

Responses to requests for paternal testing are likely to be seen as decisions made by clinic staff, rather than KGBC researchers. At the same time, since the SCD clinic is widely seen as a research-

[125] In the first group of community representatives, there were some challenges in generating understanding that carriers were – and always would be – healthy. These explanations formed part of the participatory information sharing activities before the discussions. The visual tools used to support discussions (see Appendix C4) were adapted after the first discussion to help to make this distinction clear.
[126] Chapter 3, section 3.3.3.4

led service, community perceptions of the way that the clinic handles such requests (for example, refusals or the withholding of controversial findings) are likely to impact at least as much on attitudes towards the research centre as government health services. This further underlines the need for careful development of long term collaborative arrangements between researchers and providers around such services.

Children with normal haemoglobin

The overlap implicit in the concept of a 'researcher-provider' does not seem to apply to the positive responsibilities of researchers in KGBC overall. Community perceptions about researchers' responsibilities towards the majority of families (more than 80%) who neither have a carrier child nor are referred to the SCD clinic do not suggest an important influence of conflation between researcher and providers since child participants in this case are well.

7.4 Challenges to realising expected benefits of information sharing

So far in this chapter, I have described ways in which responsibilities for achieving benefit through SCD information sharing might be seen as specific to researchers. Even where this is the case, these responsibilities would be impacted by challenges to achieving benefit *that researchers are reasonably unable to counter*. In the following paragraphs, I discuss the nature of three such areas: challenges related to communication (7.4.1); the (unresolved) risk that requests for paternal testing - consequent upon sharing information on parents' genetic roles - could undermine expected benefits of disclosure[127] (7.4.2); and challenges presented by the availability of resources (7.4.3).

7.4.1 Communication challenges

Communication was described as an important challenge to achieving expected benefits of sharing information on SCD and carrier status in Phase Two, given the need to build understanding and acceptance of the condition, including the value of biomedical care and the roles of both parents.

[127] As seen in Chapter 6.

Heterogeneity within the community was a key feature seen to underline the importance and challenges of communication. Particular concerns highlighted in this section are generating acceptance of the chronic nature of the condition, addressing risks of gendered discrimination and ensuring parents can pass carrier information on to children at an older age.

Talking about chronic conditions

As described in Chapter 3, a common understanding of biomedical treatment was that a cure would be the outcome. Even families interviewed in Phase One who regularly visited the SCD clinic spoke of disappointment in their child's progress, wondering, for example, if they could look forwards to a time when they might no longer need to attend. Lack of a cure was a prominent reason for 'switching behaviour' to another form of care (Nyamongo 2002). At the same time, explanations of the *incurable* nature of SCD were seen to carry a risk that the parents would lose hope, potentially damaging relationships between children and their families. Counselling on this genetic condition was seen to need a balance between encouraging realistic expectations of progress and engendering hopelessness. Many community members pointed out that this would require a process, allowing parents to understand and accept the child's situation over time.

Addressing gendered blame

A key challenge throughout the discussions concerned the approach needed to respond to a reportedly common attitude of paternal denial of blame for problems in children, including genetic conditions. Paternal denial of blame led to a view that both parents *should* understand their genetic roles (as well as those of the wider family on both maternal and paternal sides); it also underpinned concerns that giving this information could increase gendered blame and risk of family breakup. The types of communication that might be undertaken in a particular study, like KGBC, clearly cannot address the underlying structural influences that support risks of gendered blame. However, researchers may have some influence through consistent messages and attitudes that counter

harmful structural influences which, from the findings reported here, would find resonance with the attitudes of many within the community.

Giving information to carrier children

There were many concerns about sharing information on carrier status where testing of a future partner was uncertain, mainly related to 'misplaced' and well placed anxieties about the future, and using parents as a channel for this information. Community members were most concerned about the care needed in counselling children and young teenagers; early childhood experiences were often considered important for future adult well being and teenagers seen as particularly vulnerable.

The practical challenges in empowering parents to accurately and sensitively pass carrier information on to their child in future were particularly compelling, and led to reluctance in seeing this as a good *general approach*. Heterogeneity in the community, with the risk that information would be forgotten or misinterpreted by at least some families, meant that working through parents would need significant investment in communication resources. Ideally this would involve direct communication with the child at a later age. In contrast to concerns about autonomy and confidentiality of the child described in guidelines in some northern countries (Borry, Fryns et al. 2006), views in Kilifi about sharing carrier information through parents focus more on practical communication challenges. Given its importance, there is need for more research to assess how well parent-child communication on carrier status could work in this setting, including the influence of age, how communication policies could be best implemented at different ages, and at what cost.

7.4.2 The challenge of responding to requests for paternal testing

In Chapter 6, I addressed the dilemma emerging from Chapters 4 and 5 that explaining parents' *individual* genetic roles in SCD could increase as well as decrease parental conflict and risk family breakup[128]. A highly contested question was raised about the potential for such requests to undermine the expected benefits of sharing SCD information. In that chapter, I showed that there was tension between two positions on paternal testing:

- Paternal testing seen as important to: counter the common and serious influence of paternal denial of responsibility; and to discourage marital infidelity and risks of HIV/AIDS. Some participants felt fathers had a right to this information, but not at a cost to vulnerable children and family stability.
- Paternal testing seen as inappropriate given: likely harmful consequence for mothers and children *where misattributed paternity was shown*, including the breakup of families; and harm to men suspected to be the biological father.

Further difference emerged on whether researchers would be justified in testing fathers but *withholding misattributed paternity results* to avoid the risks of disclosure, with many seeing deception as acceptable where this aimed to protect the affected family's stability. Others believed the risk of deception causing loss of trust in researchers would be too high.

Within these arguments, two specific areas of tension emerged and were unresolved in balancing values. The first area concerns reasoning on prioritising the interests of 'faithful' over 'unfaithful' mothers. The second involves the acceptability of withholding findings from paternal testing. In the following paragraphs, I draw on arguments on the internal consistency of these positions and from a wider ethical literature to determine ways in which these tensions might be resolved.

7.4.2.1 Is it reasonable to prioritise the interests of 'faithful' over 'unfaithful' mothers?

[128] The specific concerns were related to parents' understanding of their future reproductive risks (where their child had SCD); and that paternal denial might strengthen or even generate new suspicions about misattributed paternity, leading to the possibility of paternal requests for testing (where the child had either SCD or carrier status). Structural gendered influences were described as increasing the likelihood of blame and the potential seriousness of the impact on mothers.

Community members were divided on the question of balancing the interests of 'faithful' and 'unfaithful' mothers. Some, including all mothers of affected children in these discussions, prioritised the interests of 'faithful' mothers, but others saw the risks of paternal testing to other mothers as too high to be acceptable.

There are several specific reasons why it is difficult to support a view that children shown to have 'true' paternity should be treated differently to those where this is shown as misattributed. One is a logical flaw that generates wrongful discrimination, particularly given that the main perceived 'problem' with this type of unfaithfulness was the risk of HIV/AIDS. A finding of misattributed paternity may not in fact match up well with sexual behaviour or infection risk in general. In other words, the fact of true paternity being shown in one instance of conception (of the child with SCD) cannot confer a general status of 'faithfulness' on other women. Marital infidelity may have occurred at other times, with or without conception. In addition, the consequences of discriminating in this way would almost certainly adversely affect particularly vulnerable children; since the children could not be held responsible for their parents' actions, the unfairness of the discrimination seems to be compounded. Finally, unlike risks of HIV/AIDS, this form of discrimination is inevitably uneven from a gender perspective (putting aside structural forms of gendered inequity from 'culture'); acts of sexual infidelity in men carry this biological risk of blame for the women with whom they become involved, but not for themselves.

Taking a wider perspective, it also seems possible that there are other more general ways in which men could be convinced of their role in the inheritance of SCD without resorting to investigations whose perceived importance was essentially to act as a paternity test. Alternative approaches could include family counselling, public engagement and school education and peer support programmes. Finally, there seem to be real issues with seeing such 'paternity testing' as a responsibility of researchers, and arguably even of providers. Some community members saw paternity as a private family issue. I would draw on this view that it is not the responsibility of researchers *to protect*

families from knowledge of misattributed paternity to note that - in the same way - it is not easy to see a responsibility for researchers to *demonstrate* such a situation.

In addition, in Chapter 6, I showed that the basis for prioritising the interests of 'faithful' mothers was largely consequentialist in nature. However, there is little evidence to support assessments of the utility of prioritising the interests of 'faithful' over 'unfaithful' mothers. A particular difficulty would be taking account of the risk of extremely severe consequences[129], even if this affected very few cases. In writing about the difficulty for research ethics committees to assess risks from 'worst-case scenarios' in genomics research, Hoedemakers and colleagues (2006) propose arguments based on a safety-first principle, taking public concerns seriously and that neglect of public concerns can lead to less trust and therefore participation in research (Hoedemaekers, Gordijn et al. 2006). In the Kilifi case, public opinion on the risks of sharing misattributed paternity information is divided, but the safety first principle remains highly relevant.

Taking the conclusions of Chapter 6 and the arguments in this section together, I argue that the interests of mothers whose children are shown to have 'true' paternity following paternal testing should not be prioritised over other mothers; a position concurring with that of many in the community.

7.4.2.2 Should researchers test fathers but withhold misattributed paternity findings?

In Chapter 6, I described a further area of unresolved tension between views that:

- The importance of paternal testing was so great that this would justify withholding misattributed paternity results;
- Deception would always present the greatest risk of harm through jeopardising long term trust in researchers.

[129] See for example the opinions of KCR03 group members who feared that mothers implicated in this way could be physically attacked and even murdered.

Both positions were primarily grounded on consequentialist considerations. As I noted in Chapter 6, the rights of fathers to this information was not generally seen as more important than the interests of the family of which he was a part. In this case, the wrong of providing false reassurance to the father about paternity was seen as compensated for by the need to protect the interests of the child.

However, where researchers explain (after testing) that a father is a carrier when this is not in fact the case, community accounts can be drawn upon to suggest that this is likely to generate anxiety to the parents and the wider family about the couple's future reproductive risks. Thus, whilst community members in Kilifi saw *false reassurance* about paternity as acceptable in supporting the interests of the child, mother and family (the main argument against being about loss of trust if found out), the implications of *false anxiety* should also be taken into account. Ironically, in cases of misattributed paternity, there is no future risk of this nature, but families cannot be reassured about this without at the same time implicating marital infidelity. In Kilifi, it is debatable whether lifelong anxiety about the health of all future children or shorter term conflict over an instance of marital infidelity presents the greatest threat to the welfare and stability of families.

The complexity of this prediction is shown by the findings on contextual influences in Chapter 6, particularly issues of jealousy and forgiveness alongside micro and macro level factors. In talking about disclosure of misattributed paternity findings in genetic testing in the UK, Lucassen and Parker (2001) express a concern that couples will make *misinformed* choices where this information is concealed (Lucassen and Parker 2001); in Kilifi, the misinformed choice could be the unnecessary breakup of some families, with costs for affected children and their mothers.

Views on the acceptability of researchers testing fathers and concealing misattributed paternity findings do not seem to fully take account of the potential long term consequences of this action on the stability of the family. Further empirical research may help to understand these risks. The risk

of loss of trust[130] should also be taken into account. On this basis, I argue that researchers should not undertake paternal testing in order to avoid the risk of either disclosing or withholding misattributed paternity findings, a position supported by many community members.

From the two sections above, on whether the interests of 'faithful' mothers should be prioritised and whether fathers should be tested on request but controversial findings withheld, I conclude overall that researchers should avoid paternal testing where a child is shown to have SCD during participation in research. This conclusion reflects that of community members who supported position #1 in Chapter 6 (section 6.3.1). The main ethical principles drawn upon, from community members' and my reasoning are respect for persons, particularly in relation to avoiding gender discrimination; a duty of care to vulnerable children; and obligations generated by the nature of researcher-participant relations, including the importance of trust.

From this conclusion, it also follows that the importance of explaining parents' individual genetic roles, particularly where their child has been shown to have SCD, remains high in spite of the risk of generating requests for paternal testing. Inevitably, this conclusion may generate tension on the ground, as happened in family F09 in Phase One[131]. Instead of paternal testing, what is likely to be needed is a respectful and consistent refusal accompanied by a clear explanation of the roles of researchers, and supported where possible by approaches to family counselling that acknowledges the importance of "breaking down barriers between feuding couples rather than building them up", as suggested by Turney (2005) from her observations of this situation in Australia (Turney 2005) (p245). In addition, addressing the wider issue of paternal denial that primarily underpinned calls for paternal testing should also be seen an important concern for researchers.

7.4.3 The issue of resources: Influences on information sharing and partnership building[132]

[130] Chapter 6, section 6.3.1.2
[131] Chapter 3, section 3.3.4.3
[132] In most discussions with community members, the costs and source of resources needed to support the types of services required to achieve expected benefits were not directly explored, with the exception of some health managers.

So far, I have argued that researchers have specific and strong responsibilities in relation to disclosure of SCD status information based on considerations of trust, through the partial entrustment model and through building and maintaining trust in relationships with participants. This assessment has taken into account the existence of some forms of conflation between researchers and providers amongst community members. Having also argued that such responsibilities are not negated by the existence of challenges such as difficulties in communication and risks of paternal testing, I now turn to an area that does seem to present important limitations, that is, the influence of resource availability.

In any setting, the services crucial to achieving benefits from sharing information on SCD for affected children include validating screening tests; providing family counselling on the findings; and providing at least standard care for the disorder, ideally in the long term. For sharing information on carrier status, services would include validation of tests and family counselling. These services are likely to be needed for around 1% of children found to have SCD and 18% to have carrier status in areas where the prevalence is similar to Kilifi. As discussed in the previous section, communicating information on these two aspects of the disorder (directly affected and carrier status) will be very different, and therefore have different resource requirements. Many aspects of information sharing for children found to have the disorder will be more demanding. However, the higher prevalence of carrier status would influence the overall level of resources needed to ensure benefit in disclosing this finding. Given the challenges in helping families to understand and accept the chronic nature of the condition, working against embedded structural influences and responding to heterogeneity in the community, the resources needed for effective communication and counselling have already been described as likely to be significant both for affected children and carriers, and to include *publically* as well as *individually* targeted strategies.

There are three main reasons why researchers may not be able to provide resources to support sufficiently effective services in these three areas of validating tests, counselling and ongoing care:

- *Research resource prioritisation:* As described in Chapter 1, the basic nature of research as a scientific enterprise to generate new knowledge presents a moral dilemma in balancing the importance of research as a public good (Ossorio 2006) against the arguments for providing ancillary SCD services made so far in this chapter. Embedded in this reasoning is an acknowledgement that the limitations presented by resource prioritisation for researchers do not mean that their responsibilities are 'negated as a moral consideration' (Shalowitz and Miller 2005). Instead an underlying justice issue emerges from these analyses; how to negotiate the balance between expanding the resources needed to undertake individual research projects where providing ancillary services are seen as a strong responsibility, and still maintain the resources needed to support the public good that social outcomes of research represent more widely.

- *Short term research funding cycles:* The lifelong nature of SCD militates against researchers providing ancillary services including sharing information, given that research funding is typically based on short term cycles. For the same reason, the proposal for sharing carrier findings directly with children at a future date, as suggested by some community members as well as guidelines (Borry, Fryns et al. 2006) seems unlikely to be achievable by researchers.

- *Risks of undermining government health services*: Researchers' efforts to provide long term care may undermine government health services (Georgetown University Workshop 2008) or, if not carefully planned, the relationships between researchers and government health providers.

For these reasons, researchers are not only unlikely to be able to provide sufficient services of this nature unilaterally, but should also arguably always address any such responsibilities in conjunction with those responsible for long term health care in the community, that is, generally, government health providers. The context of research in developing countries serves to amplify the dilemma for both researchers and providers, where shortfalls in resources mean that prioritisation in health care is a crucial activity in itself as well as an important influence on research. In this

way, research and health service planning become mutually interdependent; effective planning for health services needs to draw on information from research, and the ethical conduct of research - particularly around lifelong health conditions - is strongly influenced by the availability of sustainable services, which is the province of health providers.

As in the case of KGBC and Kilifi, it is highly likely that requirements for health services emerging from research activities should be based on joint resources, at least until a clear case can be made (from research) around the inclusion of specific types of interventions within routine medical care. In fact, as stated, the clinic in Kilifi is run collaboratively between researchers and the district hospital, although it is largely supported through research funds at the current time. Strong community views on the importance of SCD information sharing provide support for continued prioritisation of these research-led services at least as long as SCD screening is conducted as part of any type of research within the programme. They also underline the need for research that includes SCD screening to be strategically aligned with government policy on this disorder, both to support the development of future health planning and to allow research to draw on current strategic approaches to care.

For a research programme including studies that undertake SCD screening, central provision of services obviates the need for individual projects to take this responsibility; and ensures sustainability beyond each project. One point of consensus at the Georgetown workshop (2008) was that funders of research should shoulder some of the responsibility for providing the resources for ancillary care in studies. The model of core funding support to the programme in Kilifi for cross cutting services - like the SCD clinic - for a range of studies seem particularly well suited to this purpose (Georgetown University Workshop 2008).

The literature on the role of background inequities in framing researchers' responsibilities for ancillary care[133] is particularly relevant to these arguments. In Chapter 1, I described Lavery's (2010) argument that forms of benefit sharing in internationally funded research should address

[133] Chapter 1, section 1.8

structural health resource inequities (Lavery, Bandewar et al. 2010). Important features of background inequities in this case are the limited public health resources, contributions to harms of 'not knowing' about SCD and structural gendered blame. Cooperative building of SCD and other core services to support both research and health care in Kilifi seems to offer potential for structural 'relief' of such background inequities, amongst other potential benefits.

At the same time, it seems likely that decisions taken collaboratively between health providers and researchers about ancillary care in research may often generate moral dilemmas, especially in relation to resource prioritisation. As well as ensuring that particular decisions on ancillary services are based upon ensuring accountability about the way decisions are taken (Georgetown University Workshop 2008; Fabsitz, McGuire et al. 2010), these decisions may often need to be seen as temporary pragmatic solutions (pending further research, for example, to better inform policy).

7.5 An influence from important emerging values

Two main values emerged as strongly influencing the nature of discussions, including debates over the way that they could be balanced against each other: protecting the interests of vulnerable children/the stability of families; and openness as a form of individual or community empowerment. During these discussions, priority was frequently and consistently (including by community representatives, leaders, staff groups and affected mothers) given to values associated with the *consequences* of actions in relation to these values. In contrast, as has been described, the rights or responsibilities of individuals tended to take a secondary role in explaining or supporting a position argued on account of anticipated consequences, including the responsibilities of researchers. In this section, I explore the nature of the main consequentialist values that emerged, including ways in which these either worked together or at times generated conflicts. Conflicts between values seen as independently important may be of particular relevance to understanding moral dilemmas of this type in a given setting.

As a caveat to this interpretation, the emergence of a strongly consequentialist pattern in reasoning may be linked to the nature of the questions and health issues in the study (Chilibeck, Lock et al. 2011; Geelen, Van Hoyweghen et al. 2011; Horstman and Finkler 2011). Features of this study that may have influenced community views on the importance of particular values in these discussions include the centrality of vulnerable young children; the strength of connection between a child's interests and that of their near family members specific to their age and vulnerability; the nature of SCD as a relatively severe disorder; the genetic nature of the information to be shared; and the means of SCD inheritance as a single gene, implying particular risks and genetic roles in families.

7.5.1 The interests of vulnerable children

The first fundamentally important value that emerged across all the discussions in this thesis was the need for researchers to take special care of *the interests of vulnerable children*. For example, where the relative importance of conflicting values was being assessed, the most common 'bottom line' was seen as the interests of vulnerable children. Although many other values were expressed, and sometimes strongly supported, these were generally not considered more important than the interests of a vulnerable child where conflict emerged. Not surprisingly, these interests were most strongly seen for children thought to be most vulnerable, that is, those found to have SCD. This value framed a parallel duty of care for researchers, taking Beauchamp and Childress (2001) description of such a duty as having 'standards of conduct that are generally acknowledged by those in the profession who are serious about their moral responsibilities' (Beauchamp and Childress 2001) (p5).

7.5.2 The stability of families

In addition to seeing that researchers have a duty of care towards vulnerable participant children, community members also aligned the interests of these children with those of mothers and wider families. The *stability of family units* often took the form of the most important goal underpinning debates on values. Community members across all the types of participating groups expressed this

attitude towards family units[134], seeing these sometimes as representative of individual and sometimes of community forms of a harmonious life.

This position closely reflects reasoning in communitarian approaches to ethics that according to Stephen Holland (2007), in his examination of the role of communitarianism in public health ethics, 'emphasise the social nature of our life, identity, relationships and institutions...' in order to '...restore the notion of community to proper prominence' over more individualistic philosophical stances (Holland 2007) (p40). By a communitarian account, there is incoherence in focusing on the rights and needs of individuals because 'only people with fully formed characters and moral depth can genuinely choose anything'....and 'one acquires the requisite character and depth by being initiated into the culture or tradition of one's community' (Holland 2007) (p41).

There are also similarities with accounts of *the moral significance of families* that reference features of solidarity and collective responsibility (Lindemann Nelson and Lindemann Nelson 1995) (p85). As described earlier, this importance of families in genetic testing is supported by UK guidelines on sharing incidental findings in children that stress that 'the interests of a child cannot be considered separately from those of his parents and other family members.' (Borry, Fryns et al. 2006) (p136). The Lindemann Nelsons (1995) argue for a moral value of families that is partly *instrumental*, towards preservation from harm, nurturing, socialising and providing the basis for the development of individuality of its members. They also emphasise an *intrinsic* value, seen as the 'goods of intimacy' (Lindemann Nelson and Lindemann Nelson 1995)(p86). In Kilifi, community members described instrumental and intrinsic values associated with family stability. Practical and structural gendered arrangements in society that influence access to resources particularly underline

[134] As defined in Chapter 4, the concept of family stability was applied to part of, or the entire, household. At the same time, social constructions of households differ within the community and the nature of a 'family unit' was seen as dependent on functionality (Molyneux 2002). In relation to informed consent processes and access to information from SCD screening, most community members saw the parents and children as the primary family unit, although others included other family members. Broader and more abstract concepts of family (for the example, the blood line) were prominent in tracing sources of the disorder.

the moral role of families in protecting the interests of mothers and vulnerable children in this setting.

It seems reasonable therefore to argue that this widely held value means that the idea of a researchers' duty of care towards a participant child should also be considered, as far as possible, as potentially extending to the family of which the child is a part. Where this stance is seen as important, potential dilemmas may be created where the rights of any individual in the family are in tension with those of the family as a whole (Parker and Lucassen 2004). For example, the needs and rights of the child may not be adequately represented within a collective arrangement, and this balance would always need careful consideration (Parker and Lucassen 2003).

In such a balance, however, community members' views in Kilifi suggest that their priority would generally work towards seeing the child and family *as a whole*, supported by widely held assumptions that children's interests are well represented by their parents. This point is further illustrated by those community members who described their preference not to be told information about misattributed paternity[135], seeing this as risking stability of the families they held as essential to their own well being. Of relevance to an idea of alignment of interests, much more focus in all the debates was placed on 'interests' than on 'rights', wherein the interests of fathers (as before, seen to be aligned with their families) was a more common way of taking account of the paternal position than a rights based perspective.

7.5.3 Openness as empowering individuals and society

The second commonly recurring and strongly supported value across these debates was one of '*openness'*, which I have used in this thesis to capture a widely-held belief that knowledge will contribute towards individual empowerment and societal goals of progress. In a setting like Kilifi, where traditional ways of life and poverty associated with subsistence forms of livelihood are being influenced by social and economic development efforts, including through the work of

[135] Chapter 6, section 6.2.3.2

government, the non-governmental development sector, commerce and a major international research programme (the biggest local employer), many community members perceived a fundamental importance of this need to move 'out of darkness', using the language of a health manager[136]. Sharing information about SCD, as a condition that was little known, and where lack of knowledge was seen to present serious harms to individuals and the wider community, was therefore linked with both individual and much broader development goals.

7.5.4 Tensions between values of a duty of care and openness

In practice, a duty of care and values of openness were often mutually strengthening values, even where tension generated important contingencies[137]. In the case of information sharing for SCD, this convergence led to particularly strongly felt opinions. Thus, sharing information on SCD in research was seen on balance to provide strong net benefits to affected children and their families *and* to promote public health interests of the wider community. Although sharing information on carrier status was more contested, values of a duty of care and openness were still largely aligned, such that promoting the child and family's interests through 'being prepared' fitted well with generating better public understanding of the disorder.

Sharing information on misattributed paternity, however, illustrated a situation where a duty of care to some affected children often conflicted with a value of openness in support of public interests. While a duty of care to children with 'true' paternity or (less importantly) the rights of fathers to knowledge were not generally seen to outweigh the interests of children who might be harmed by such information sharing (those with misaligned paternity), the wider interests of society were often seen as stronger candidates to do this work. The community benefits of sharing information on misattributed paternity were linked to better awareness of the disorder, building transparent relations between researchers and participants, and - most importantly - impacting on the public

[136] Chapter 6, section 6.2.3.1
[137] An example of such a contingency is the importance of effective communication in limiting potential for stigmatisation.

health threats of sexually transmitted infections, particularly HIV/AIDS, by discouraging marital infidelity.

In earlier sections, I have argued - drawing on community reasoning - that it may be difficult to see a perceived importance of openness towards social progress as strongly influencing researchers' responsibilities, where conflicting values are raised. At the same time, it may be useful for ethical reflection to consider the place of these types of values, and the conflicts that may emerge between them, in other studies in settings like Kilifi, recognising that *development itself* may be seen as a very important value to many community members.

7.6 Researchers' responsibilities for sharing SCD status information during KGBC and similar studies

In this section, I bring together the reasoning presented so far in this chapter to describe my conclusions regarding the responsibilities of researchers to share information on SCD and carrier status when these findings are generated during studies like KGBC in Kilifi (and potentially to settings where similar conditions[138] apply).

7.6.1 A strong responsibility to share SCD information with parents of affected children

Research guidelines on the disclosure of genetic findings indicate that not sharing results with clinical and social 'utility' during studies, such as SCD information, generates potential for infringements to autonomy, and - for individual families - fails to meet obligations to offer fair benefits of participation.

Community perceptions of benefit show the case for disclosure as compelling based on strong and agreed views of the nature of the benefits of information sharing as countering likely and severe existing harms to participant children, their mothers and families linked to low awareness of SCD and contextual issues, considered amenable to better understanding. Contextual issues included

[138] Notably, where 'vulnerability' (from partial entrustment model) is likely to be high/information sharing has the potential to limit important harms.

structural forms of health belief, gendered blame and stigmatisation, underpinned by the lack of existing public health resources for families affected by this condition. Background conditions of inequity therefore also make a strong case for researchers to undertake positive action of some sort to respond to this situation. Given the presence of the SCD clinic in Kilifi, run collaboratively between researchers and the district general hospital, there do not seem to be strong arguments based on considering resources against researchers sharing SCD findings[139].

Several aspects of earlier arguments about the role of SCD information sharing in building and maintaining due levels of trust between participants and researchers also importantly support these positive responsibilities. The most important relates to SCD information sharing as a means of appropriately showing due concern and responding to perceptions of reciprocity. Not only are these expectations reasonable, in the light of the preceding analysis, but the high importance - from community and researchers' perspectives - makes due concern a particular priority. Issues of trust, in relation to due concern, reciprocity, rumours and active and passive forms of deceit, are particularly important in settings like Kilifi, where a number of studies recruit participants over time and contemporaneously within a fixed population. In this situation, trust in the researcher-participant relationship often emerges as a collective phenomenon with the potential to affect many different studies. This heightens the risk of mistrust to the validity and sustainability of not just one study but to others as well, and therefore to the capacity of the programme to contribute to the public good that health research represents (Marsh, Kamuya et al. 2010). Given concerns about levels of health research investment and scientific research capacity in developing countries (Global Forum for Health Research 2000), these public goods seem particularly important for long term, large multidisciplinary research centres in Africa. The risks of inappropriately high levels of

[139] In the absence of such services, setting aside resources for their development would have to take into account wider implications for priority setting in research and the need to develop such services collaboratively with the Ministry of Health, and the health priority setting policies this would imply. In other words, development of SCD services to support research would need to draw on a clear public health policy about such care - at district and national levels - in order to ensure that the services were seen as appropriate to national policy, and have adequate investment by governments to ensure long term sustainability.

trust may similarly be influenced by overall perceptions of the centre as providing high quality care.

The risk of diagnostic interpretations of the study has been shown to present the strongest moral counter to arguments for SCD information sharing, with important challenges to the autonomy and respect for participants in relation to the main scientific questions. At the same time, the risk of diagnostic interpretations does not seem a sufficient counter to outweigh the moral reasoning for sharing SCD information presented so far, and the fact that additional communication efforts could be harnessed to address these interpretations.

A specific challenge concerns the way researchers should respond to paternal requests for SCD testing, as a result of paternal denial of responsibility. Community deliberations on this issue were mixed, but I have argued that researchers should not test parents in this situation. Drawing on community deliberations, I have argued that the seriousness of harms likely to be caused by showing misattributed paternity, even if in few cases, outweighs responsibilities to counter paternal denial in this way. If testing is undertaken and misattributed paternity found, it will not be possible for researchers to withhold this information given the risk of loss of trust between families and researchers, and the generation of unnecessary stress to the parents in relation to future reproductive risks. Further there are other ways in which risks of paternal denial can be countered, such as family and peer counselling; and issues of marital fidelity are not generally considered the domain of researchers. Even if a responsibility could be argued for researchers to test fathers (which I have argued against), the resources needed for testing and counselling families would have to be justified as a research cost.

However, the mismatch between expectations of benefit and those reported in KGBC illustrates practical challenges to achieving these. Important ways in which these benefits could be made more likely include addressing the communication challenges reported on earlier in this chapter, mainly concerning the chronic nature of the condition and the genetic roles of parents; and issues of access for some families, which might include decentralisation of services or supporting

transport. Innovative ways of helping families to understand the nature and inheritance of the condition are important; many such approaches are commonly used in genetic counselling. Ways of adapting methods to the context in Kilifi would be important, including the possibility of peer counselling and support.

Given that the responsibilities for researchers to share information on SCD arguably remain strong in spite of risks of diagnostic interpretations (seen as a perverse outcome), the existence of these risks generates a further responsibility for researchers to develop more effective ways of countering diagnostic interpretations of research, which are both likely and morally problematic. Such an approach is likely to rely on more effective communication, including general community engagement as well as increased interactivity with participant families; with for example emphasis placed on providing access to SCD screening - if wanted - independent of a decision to participate in the genomics study.

From the above paragraphs, community engagement will play an important role in developing and maintaining trust in studies that include SCD screening. Community engagement is likely to provide important support to informed consent processes, including consultative processes to explore expectations, and establish and seek consensus on fair benefits and how these will be explained.

7.6.2 Carrier findings: Balancing values

Guidelines for researchers on feedback of genetic findings in studies recommend that SCD carrier status should be shared with participants, on the basis of social 'utility' in making decisions about future reproductive choices. However, this benefit is uncertain in Kilifi, since public access to screening for SCD is not currently available, and there is no reason at present to anticipate a change in this situation. However, different forms of benefit were seen by community members, including future preparedness, with the potential to limit future harms due to low awareness of the condition; and creating wider public awareness of SCD, such that public advocacy could apply pressure for

wider screening services to be set up. Community views were, however, mixed and a few clearly stated a right not to know. In any case, these forms of benefit are less convincing than for information sharing on SCD given that the harms implied carry less certainty and immediacy. Further, it is hard to see creating public demand for SCD services as a responsibility of researchers. In addition, there are important challenges to achieving this less urgent and certain form of benefit, given that new resources would have to be mobilised to validate tests and counsel families on the results, which would occur in a much higher proportion of participants than for SCD. While some community members saw the importance of empowering families to advocate for better SCD screening services, health managers were concerned about the frustration and negative impact on health services that recognition of this set of unmet needs could cause.

In relation to issues of trust, expectations about sharing carrier information are also likely to be less than for SCD on the basis of due concern, given views about benefits. However, expectations in relation to reciprocity and the risk that sharing carrier findings would generate diagnostic interpretations of KGBC are the same as those for sharing information on affected children. At the same time, the practice of merging carrier and normal findings for purposes of feedback seem to present an important intrinsic wrong of deception as well as risking harmful impacts on trust if later 'found out'. Deception is particularly compounded where families would want to know this information or have already been affected by the disorder, such that 'harms of not knowing' are already present; and risks harmful reproductive decisions being inadvertently made in future.

In other words, there may not be a strong case to share carrier findings with participants in studies such as KGBC, but these reasons do not seem to account for harms that might be generated by an overall approach to actively sharing carrier findings that strongly implies 'normality'. Further, requirements for reciprocity should still be met; and diagnostic interpretations of KGBC countered. Thus, if there are - on balance - strong moral reasons not to feedback carrier findings as a general policy, related to the challenges described so far, it is not clear what the nature of any expected

feedback could be, or if no feedback is given (as happened at the start of the study), how expectations of reciprocity can be met.

However, it seems clear that there are some circumstances in which carrier findings should be shared, including situations where participants ask for this information, and those where sharing information is likely to have the same benefits as for SCD (for example, where harms are already occurring in the family through another child being affected). It seems therefore that the carrier feedback process should itself be responsive to varying potential importance at an individual level, including a right 'not to know' about this finding. This means that carrier status should be discussed during the informed consent process, with careful development of the approach to these explanations that avoids over-emphasis and technical complexity, and establishes likely direct experience or knowledge of the disorder. Further, that opportunities to actively or passively feedback individual carrier results should be built into the process for those who are seen to be at high and urgent risk of 'harms of not knowing'[140] or who actively seek this information. The experiences of the newborn screening programme in Ghana on making carrier results available to parents on request may be helpful in this regard (Ohene-Frempong 2005).

7.6.3 A strong responsibility to build partnerships with government health providers

For information sharing on SCD and carrier status, a strong responsibility is framed for researchers to work actively with the Ministry of Health to establish the place of SCD management within national health priorities, including the role of carrier screening programmes (either as part or separate to screening programmes for the disorder itself, such as newborn screening) in order that appropriate forms of information sharing and ancillary services can be developed that respond to the dilemmas seen in research that includes screening for this disorder. This seems particularly important when a significant body of research including SCD screening is taking place, and there is sufficient evidence on prevalence to begin to inform policy development. At a minimum, research that generates findings on SCD and carrier status should be planned in collaboration with

[140] That is, those with an affected family member but who do not recognise the condition.

government health providers to ensure that research and policy are mutually supportive, for example, that withholding carrier findings is seen as a fair strategy by health managers and policy makers. Further, Ministry of Health programmes that raise awareness of SCD would, at the same time, begin to address the responsibilities for researchers towards SCD information sharing in many ways, given the fundamental role of the phenomenon of low awareness throughout the analysis presented in this chapter. It would also have much wider potential to impact the harms of 'not knowing' identified in this study.

7.7 Conclusions

In this chapter, I have drawn upon the findings from deliberative discussions with community members and areas of bioethics literature to address specific thesis objectives on the nature of important ethical questions about disclosure of SCD findings in research, including the responsibilities of researchers during studies like KGBC in Kilifi[141]. The recommendations for researchers are summarised in Figure 7.1, including that information on SCD should be shared with families of affected children, particularly its genetic basis; and that information on carrier status should be made passively but not, in this context, actively available. Further, sharing SCD information should be seen *as a form of ancillary service*, involving ensuring access to, or providing, validation of tests, family counselling and at least basic care. These conclusions are strongly based on community experiences, perceptions, views and values, including the way different values are prioritised in this situation. In the next and final chapter, I consider the implications of this analysis for other studies in different malaria endemic settings where findings on SCD are generated.

[141] As well as being generated around a particular study and setting, the findings are based on a relatively novel approach to empirical ethics. In the final chapter I describe an approach to planning feedback of SCD findings in other types of study and geographic settings, and reflect on methodology, including its strengths and limitations.

The ethical arguments for sharing information for affected children in KGBC are based upon: showing respect for individual and community[142] autonomy; the potential to limit serious and immediate harms for affected children (seen as specific to researchers through the planned inclusion of SCD screening in research, and the vulnerability and dependency of many families in relation to these findings); and the moral importance of building and maintaining trust (in this study and others, now and in future). These have been balanced against: the costs of providing ancillary services for SCD, related to research resource prioritisation; risks of undermining government services/partnerships; and the potential for generating diagnostic interpretations[143]. In this analysis, the high level of importance of sharing SCD information for affected children is taken to outweigh moral reasons *against* information sharing, which are argued as more appropriately seen as *challenges* towards which researchers have additional responsibilities to respond.

Decisions on sharing carrier information present a difficult moral dilemma for researchers and communities, and passive information sharing is argued as a pragmatic and temporary solution. Where a family is already affected by the disorder at the time of participation, however, carrier status should be actively shared. The basis for this argument is respect for autonomy; and that sharing carrier findings a) may not generate anticipated benefits, instead leading to harms in some cases; b) does not have the potential to limit immediate serious harms; and c) requires the investment of long term resources that researchers cannot guarantee and may undermine the 'public good' nature of research.

These arguments include an additional responsibility for researchers to act on the recognition that sharing SCD findings has a high risk of generating diagnostic interpretations of research. Engagement with communities and potential participants should ensure that voluntariness in, and understanding of, studies are sufficiently supported. A further responsibility for researchers,

[142] That is, taking account of the views of community members in these discussions.
[143] Shown in Chapter 3 to sometimes be closer to a form of *distraction from* than *replacement of* the main research question.

towards all participants, irrespective of the nature of SCD findings, is to maintain due levels of trust and reciprocity through the way studies are conducted.

For both sets of conclusions, around sharing SCD and carrier information, further engagement with a wide range of stakeholders, including debate on the conclusions reached here, would be important to develop research policies. The analysis underlines the importance of collaborative partnerships between researchers and government health providers in ensuring long term SCD services are available to children. Most notably, researchers should support the development of clear national policies on SCD within which framework future research and services could be mutually planned. These arguments support the concept that researchers working in developing countries through international collaborations have some responsibility to take account of background inequities, as part of a wider view of the role of research in supporting global justice (Benatar and Singer 2000; Benatar 2002; London 2005; Ijsselmuiden, Kass et al. 2010).

Figure 7.1. Summary of researchers' responsibilities for sharing SCD information during KGBC

1. **For findings on SCD in affected children:** Researchers have a strong responsibility to share this information with families *as a form of ancillary service* (validating tests, counselling and care) for reasons of: showing respect for individual and community autonomy; providing benefits that importantly have the potential to limit harms; and maintaining a due level of trust between researchers and participants. These are countered by issues of research resource prioritisation, and risks of undermining government services/partnerships and generating diagnostic interpretations.

   Concurrent responsibilities are:
   - Working collaboratively with the Ministry of Health/District General Hospital to plan and implement services for SCD;
   - Ensuring counselling services support family stability as far as reasonably possible, taking account of:
     - The importance of sharing understanding on the genetic roles of parents and on both sides of family over many generations; and of long term care, while maintaining hope;
     - Particular sensitivities in counselling, including parents' future reproductive risks, paternal denial of responsibility and suspicions of misattributed paternity; and individual variations in responding to genetic information;

2. **For findings on carrier status:** There is less responsibility for researchers to share this information actively, given: diminished potential for benefit, and limiting harm; and less support from community autonomy, including a right not to know; and counter arguments as for affected children.

   Additional responsibilities are:
   - Providing passive feedback of findings where requested;
   - Providing active feedback where an existing risk of harm is identified in the family that sharing carrier information is likely to limit (e.g. undiagnosed affected sibling).

3. **For SCD, carrier and normal haemoglobin findings:** Researchers have a strong responsibility towards:
   - Building forms of community engagement and informed consent processes that counter diagnostic interpretations of research, taking account of the importance of transparency, maintaining due levels of trust and avoiding over-paternalism, including being clear about boundaries of responsibility in genetic testing.

CHAPTER 8

Wider study implications: Settings, methods and further research

8.1 Introduction

In this final chapter, I turn to considering wider implications of this research, in keeping with the remaining thesis objectives[144]. In doing this, I move away from questions centred on sharing sickle cell disease (SCD) findings during research in a particular study, community and setting described in previous chapters.

As some of these wider implications have been raised in earlier parts of the thesis, my aim in this chapter is to draw together issues noted and provide additional commentary around three main areas: the implications of these findings for planning different types of studies that generate SCD findings, including in different (malaria endemic) settings (8.2); the contributions and limitations of the empirical methods used in this thesis (8.3); and recommendations for further research (8.4). The final section (8.5) provides the conclusion for this chapter. Taken together, section 8.5 and the final section of Chapter 7 (7.7) describe the overall conclusions of the thesis.

8.2 Key considerations for sharing SCD information in new studies and settings

In many research settings where malaria is, or has been, endemic, it is highly likely that SCD will be an important background health problem, although often - as in Kilifi - masked by focus on the greater contribution of communicable diseases. As many communicable diseases are increasingly controlled, an epidemiological transition is occurring in developing countries such that non communicable diseases account for an increasing proportion of health burdens (Weatherall and Clegg 2001). In this context, SCD will become an increasingly prominent background health

[144] See Chapter 1, section 1.2: To propose...key considerations in planning for SCD information sharing in other types of studies and different malaria endemic settings; and to explore the potential contribution of descriptive and deliberative empirical approaches to resolving ethical dilemmas in research, including as a form of community consultation.

concern for researchers working in developing countries. This is particularly important where research draws on international collaborations, where some responsibility can be convincingly argued for researchers to respond to background inequities (Benatar and Singer 2000; Georgetown University Workshop 2008; Ijsselmuiden, Kass et al. 2010; Lavery, Bandewar et al. 2010). The type of studies for which the issue of sharing SCD findings may be relevant is wide. Already, as noted in Chapter 1, screening for SCD status as a risk factor is included in diverse studies in Kilifi, including those on malaria, pneumonia, malnutrition and maternal health. With increasing interest in genomics research in Africa, including the internationally funded H3 Africa initiative[145], there is likely to be increasing need for researchers to consider their responsibilities towards sharing findings on this common, serious and often poorly recognised genetic condition. In this section, I draw on the findings in Kilifi, as discussed in earlier chapters, to propose six key considerations to be taken into account in planning research generating SCD findings, including other types of research and different geographic areas. These considerations (See Figure 8.1) concern the context for research and are *supplementary* to other professional and ethical responsibilities of researchers, including those in national and international guidelines. In summary, these are:

1. **The nature of any benefits and harms** related to information sharing, including from a community perspective, with emphasis on potential for *harm limitation*.
2. **Researcher-participant relations**, including features of *trust*, and some influence from intensity of engagement, particularly *vulnerability* and *dependence* of participants.
3. **Local/national resources and partnership**s, including the nature/existence of *resources* to support disclosure; and collaborative partnerships with government health policy makers and providers seen as essential to ensuring research and health policies are mutually supportive.

[145] http://h3africa.org

4. **Research related issues**, including SCD prevalence; feasibility[146]; social value[147]; and *research resource capacity*.
5. **Perceived rights and responsibilities of researchers** from a community perspective[148].
6. **Balancing values**, including the *process* by which the issues above can be weighed up where conflicts or tensions emerge.

In Appendix F, I expand Figure 8.1 to include important findings from the study in Kilifi, as an illustration of the way this framework could be used (table 8.2).

Using the approach outlined in Figure 8.1 places emphasis on the need to take a wide view of the existing landscape for research, details of the study and the opinions and values of different stakeholders in the process. Many authors have pointed to the importance of engaging local communities in order to strengthen ethical aspects of decision making in research, including through processes of consultation (Sharp and Foster 2000; Emanuel, Wendler et al. 2004; Dickert and Sugarman 2005). The six key considerations identified above propose a move towards greater public involvement in research planning, through: i) Highlighting specific areas of importance for consultation in planning research including SCD screening, and ii) Supporting an approach to *balancing values* - that will almost inevitably be needed - that is based on *in situ reasoning*. At the same time, the methods needed to develop these local understandings for new research settings may not necessarily need to draw on the particular approach described in this thesis, as I will come back to discuss in section 8.4 on implications for new research.

[146] In some instances, research may include use of anonymised samples, or those archived for considerable periods of time, making it difficult or impossible to trace study participants to share findings.
[147] The social value (or overall scientific and social importance) of a study, including to the local population , may influence the balancing of a positive value of SCD information sharing (limiting harm, supporting autonomy) and negative value of its impact on resources, where this undermines the public good of research or government health services.
[148] In Kilifi, seen as a function of balancing harm/benefit outcomes

Figure 8.1 Key considerations for sharing SCD information in studies: Questions about context

1. BENEFITS AND HARMS: Nature, probability, severity and who affected?

- What *understandings* and implications of SCD for the child, family and wider community? Are there any particularly sensitive areas of SCD information sharing? e.g. Beliefs, gender, HIV/AIDS?
- How much would information sharing help affected children and families, including limiting *existing harms*?
- If researchers do not share information/provide support, what would families be able to do? (dependence)
- What challenges exist in achieving benefits of information sharing, and how can they be overcome?
- How is *diversity* within community likely to influence harm/benefit assessment and dependency, including for SCD and carrier status (such as ability to access private testing/care)?

2. RESEARCHER-PARTICIPANT/COMMUNITY RELATIONS

- **Trust:** Do study activities, including community engagement and informed consent support trust between research and participants/community?
  - What risk of diagnostic interpretations? Is understanding of study undermined? Are expectations of reciprocity unrealistically lowered, with potential to undermine fair benefits?
  - Do research activities demonstrate transparency and consistency in participant/community relations?
  - Are expectations of due concern about SCD information being addressed?
- **Intensity of engagement:** What is the relationship between researchers and participants/the wider community during the study, *and for other linked research*?

3. LOCAL/NATIONAL RESOURCES AND PARTNERSHIPS

- **Nature of resources:** What is needed to validate tests, provide counselling and *long term care* for affected children?
- **Existence of SCD services:** What services already in place for SCD diagnosis and management, including public information? How accessible are services?
- **National SCD health policies:** What government national/local policies/programmes for SCD (even if not implemented at all levels)? How would researchers sharing information fit with this?
- **Other health programmes:** What other health programmes are in place that could either support or challenge SCD information sharing (particularly HIV/AIDS)?

4. STUDY RELATED ISSUES

- **SCD prevalence:** Is research being conducted in an area where malaria/SCD is prevalent?
- **Feasibility:** For example, research using anonymised or historical samples, particularly if geographic tracking difficult
- **Social value research:** How important is the research, particularly to the study community? How important is SCD information to the research questions?
- **Resource capacity:** What capacity do researchers have (through planning, including with funders) to take account of resource requirements of SCD information sharing? What impact would this have on funding decisions outside the project?

5. RIGHTS AND RESPONSIBILITIES

- How are the rights of individuals and responsibilities of researchers seen as important, in relation to affected children, those found to be carriers and their families?

6. BALANCING VALUES: WHO DECIDES?

- How can competing values be balanced in a way that maximally supports ethical conduct of research?

8.3 The work done by empirical methods in this thesis: Contributions and limitations

> "[In these discussions] we have given you rice. This rice we have given you, about those issues, has pebbles and other things. So you will go and separate the rice from the pebbles. I hope what will remain will help you a lot in this matter." (KCR03/P6)

These remarks by a community representative at the end of a discussion made a strong impression on me at the time, and have continued to do so throughout the analysis and writing of this thesis. 'Separating rice and pebbles' has been a challenging process, particularly given the relatively novel aspects of some of the methods used. In Chapter 2, I described the approach taken to data collection and analysis, and commented on the difficulties anticipated and the way in which I aimed to address these.

A key challenge concerned the way in which normative conclusions could be derived from empirical research based on *in situ* processes of reasoning. These challenges could be seen as those related to the use of qualitative methods, and those linked to developing a normative account of researchers' responsibilities based on these findings. Using qualitative methods typically generates challenges concerning the relationship of data to 'reality'. Such challenges would include, for example, accounting for the influence of relationships between researchers and participants, group dynamics, unspoken ideas and changes of opinion. However, novel aspects of the methods used in the thesis relate to the second area of challenges, around developing a normative account on the basis of community deliberations. In this section, I reflect on experience of working with a deliberative approach, and the ways in which I have addressed implicit challenges.

The first comment on these methods is that I and my main co-facilitator (Francis Kombe) found the approach engaged the interest and active involvement of nearly all participants. Discussions were lively, interesting and extremely informative. The level of interest can be gauged from the fact that conversation almost always continued after the voice recorder was switched off. In most groups, participants showed great consideration towards others, while discussions remained robust in

nature. It appeared that some prior familiarity with each other, and with me and other facilitators, was important in engaging interest and in supporting openness. Least active engagement came from one of the chiefs' groups (Chiefs 03) whose expectations of the discussion had been to learn about policy on SCD, rather than to contribute to its development. In one community representative group (KCR03), a common reticence on the part of young people towards their elders made it difficult to involve some members; this seems to present a trade-off for openness between features of familiarity and peer identity.

Three linked types of challenges emerged in ethical reflection on these discussions: how to ensure that reasoning was supported by informed opinions; how to account for diversity of opinion within and between groups[149]; and, if diversity could be understood and accounted for, how to develop sufficiently robust normative conclusions that could be taken forwards to influence policy. My approach to working with these challenges is described in the following paragraphs.

8.3.1 Challenges of information sharing

A central component of this study was to ensure that community members had a sufficient understanding of the issues I saw as important to support a process of 'informed reasoning'[150]. In practice, this was not always possible for every member of a group, particularly for more complex areas of the discussion. I observed that group members actively supported others to 'keep up' with the issues in a generally good humoured way, shown by one community member describing - when he finally understood the point - that 'his network had been down'. In some instances, difficulties in understanding the implications of a point became a finding in its own right[151]. Given a background of changing understanding and opinion, it was important that the method for analysis could take account of this 'bigger picture'[152].

[149] Although this is framed here as a challenge, diversity within groups could also be seen as a success, since the aim of these discussions was to explore diversity, and not to develop consensus.
[150] Key areas of information and information sharing activities are described in Chapter 2, section 2.4.3.
[151] See Chapter 6, section 6.3.2 on the likelihood of requests for paternal testing.
[152] See Chapter 2, section 2.4.5 on data analysis in Phase Two.

Overall, there was a balance made between ensuring all members of a group were actively contributing to discussions on the basis of sufficient understanding of the issues, and maintaining a comfortable and active flow of conversation. Interestingly, the groups often maintained this balance themselves. This compromise may to some degree answer criticisms of deliberative ethics that the views of those who are less comfortable with processes of reasoning may be marginalised (Parker 2007b), given features of mutual respect shown within deliberations.

8.3.2 Experiences of exploring diversity

Diversity was explored using Macklin's (1999) approach of mapping agreement and difference as perceptions about 'facts' and values as an organising concept (Macklin 1999). From this mapping process, three important observations are that:

- Some apparent differences of opinion about sharing SCD information were in fact based upon mutual acknowledgement of diversity within the community, dependent upon contextual features (macro and micro level), and associated differences in the perceived likelihood of outcomes. These types of differences were generally locally resolvable where strategies could be found to respond to this diversity (for SCD information sharing, through individual counselling).

- Other types of divergence were based on different ways that community members articulated or prioritised values. In this case, questions under consideration became irresolvable through *in situ* reasoning. However, resolution was achieved by drawing on both *in situ* and *ex situ* processes of reasoning that can be seen as a form of reflective equilibrium[153] in this study (Beauchamp and Childress 2001). This iterative process involved components of:
  i) Exploring the 'fact/value' basis of community members' opinions (as an organising concept);

[153] See Chapter 2, section 2.4.1.1.

ii) Assessing the consistency and strength of diverging views across the findings and in comparison to wider ethical frameworks from the literature and guidelines;
iii) Drawing on values, and prioritisation of values, seen as important by community members;
iv) Assessing the extent to which a proposed 'solution' would be compatible with the views of a significant number of people involved in these discussions.

- The diversity of views expressed in these discussions seems to reflect a complexity that is likely to be found within and between communities when a sufficiently in-depth approach is taken to exploring values. At no time were any views consistently raised across a majority of community members that were incompatible with widely acknowledged moral values, such as respect for persons.

8.3.3 Developing normative conclusions from community deliberations

A further challenge concerns the way the set of conclusions developed from the process described above can be used to influence policy. A direct contribution is limited by the scale and scope of the study described in this thesis. To reach a more specific, supported and widely useful set of recommendations would ideally involve taking the conclusions back to a larger range of local stakeholders - including more bioethics and health policy as well as practitioner and community stakeholder expertise - for further deliberation. In this way, and given the underlying focus on local reasoning, the project follows what might be seen as the preliminary stages of a locally driven hermeneutic approach to ethics, as outlined in Chapter 2 (section 2.4.1.1).

8.4 Recommendations for further research

Over the course of this study and from its conclusions, several areas of future research have been highlighted for research and for policy relating to SCD; these are summarised in the following paragraphs.

Strengthening theories about researchers' responsibilities for ancillary care/services

The findings of this study suggest that the partial entrustment model (Richardson and Belsky 2004; Richardson 2007; Georgetown University Workshop 2008) is a helpful way of considering the theoretical basis of such a responsibility for researchers, but may fall short in some areas. While based on recognition of levels of trust implicit in a professional relationship between researchers and participants, it does not take account of the responsibilities that might be generated in maintaining due levels of trust, including responding to expectations of due concern and requirements for transparency. A failure to provide ancillary services (for example, on the basis of decisions about resource prioritisation) can undermine trust in ways that can compromise the scientific validity and sustainability of studies. This seems as potent a way of undermining the 'public good' nature of research as decisions about resource prioritisation. In addition, assessments based on the level of intensity of engagement between researchers and participants, argued against elsewhere from a justice perspective (Fryer-Edwards and Fullerton 2006; Ossorio 2006), should also take into account the nature of engagement from participants' perspectives. In Kilifi, many different studies conducted at the research centre tended to be seen as one programme of work, with implications for the perceived responsibilities of all researchers. Further work on developing models for researchers' responsibilities for ancillary services is important to support research planning - not least funding (Beskow and Burke 2010) - including exploration of these findings in other settings.

Building understanding of local perceptions and experiences of SCD to support research and health policy development

The findings have provided a rich account of the way that this disorder impacts many lives in Kilifi. As shown, this is critical to understanding the balance of benefits and harms that might be a consequence of sharing SCD findings in research, as well as planning the nature of services. More research on community perceptions and experiences of SCD is important to map out a broader picture of these features, to develop stronger models of the influence of diversity and to understand

the types of services that maximise benefits for affected children and their families. Questions would include: How typical are the structural gendered influences on outcomes of information sharing reported in this study? How likely are families counselled on genetic roles in SCD to subsequently break up, and what are the risk factors and consequences? What forms of communication and counselling best support family stability, and at what economic costs? What is the role of parents in passing carrier information on to their children? In addition, the role of self help groups and measures to support geographic and economic access to clinics seem likely interventions to explore, including decentralising specialist services to peripheral health facilities. Since some parents appreciated the support given by other parents attending clinic at the same time, decentralised care has the potential to act as a focus for local parent support groups.

Building understanding of the value of the methodological approach in this thesis

Given the relatively novel approach to data collection and analysis in this thesis, more experience and research on the methods are important to build understanding of their value. Several aspects of this work seem key. Feeding back the findings described here to community members involved in the study would help to strengthen the trustworthiness (Patton 1999) of the findings and begin an iterative process of testing out the conclusions towards developing stronger recommendations for policy[154]. Seeking more experience with the approach in other settings and around other topics would help to understand its wider usefulness. A closer examination of structural aspects of deliberations, for example through discourse analysis (Schiffirin, Tannen et al. 2001), would provide greater insights into the methodology and its limitations.

Pragmatically, it will also be important to consider ways in which the methods used here could be adapted to support their more routine use in consultation with research stakeholders as part of a process of community engagement. It seems likely, for example, that similar discussion-based approaches that draw on informed reasoning of community members would be helpful in formulating policy in the areas described in Chapter 3 as 'gaps' for genomics research, such as the

[154] See section 8.3.3 above.

acceptability of archiving of samples under a broad form of consent. Any trade off between increasing the feasibility of such a process (for example, by reducing the time and resources needed) and its overall value in ethical reflection would have to be assessed.

Building understanding of the role of emerging values in this research

Two values emerged as central to discussions in this thesis; family stability and openness towards empowerment. Researchers' responsibilities were always seen as secondary to these considerations. Taken together, a consequentialist nature to reasoning was generally prominent, although the emergence and balancing of values is likely to have been influenced by specific features of SCD in these discussions, including the role of vulnerable children, the genetic nature of the condition and its emotional, physical and economic costs. I have drawn strongly on family stability as a value in my reflections in Chapter 7, given its predominance for most questions. At the same time, openness and empowerment represented key values for many community members, seemingly underpinned by a strong perceived need for social progress. The emergence and balancing of these values for SCD information sharing in other geographic settings, and around other dilemmas in research and health care, seem important to explore as features of a moral landscape that have potential implications for normative reflection.

8.5 Conclusions

Although the conclusions in Chapter 7 on moral issues, including researchers' responsibilities, in sharing SCD information were described for a specific genomics study in Kilifi, there are several ways in which this thesis makes a wider contribution to health research and policy in developing countries. Firstly, the findings and analysis in earlier chapters can be used to support the development of policy for other types of research that generate SCD findings, including in different settings. In this chapter, based on research in the thesis, I have proposed six key considerations for different studies and settings.

Given their relatively novel nature, and against a background of controversy on the way in which empirical research can be used to support normative analysis, it has been important to return here to some of the methodological questions raised in Chapter 2 (section 2.5). As a second broad implication of findings in earlier chapters, I have described ways in which the methodological approach taken - particularly the more innovative methods used in Phase Two - were feasible and informative for normative reflection. Arguably, by embedding rich accounts of reality and locally appropriate ways of prioritising values, the approach has strengthened the moral basis of the conclusions presented in Chapter 7, related to a specific study in Kilifi, as well as those described in this chapter. I have proposed that taking the conclusions in Chapter 7 forwards into policy would involve further cycles of deliberation with wider groups of research and health stakeholders, including feedback of the findings in this thesis.

At the same time, presenting a third broad implication, the thesis generates a number of future research questions, including about the methods used. Other areas of future research seen as important are ways of strengthening theories on researchers' responsibilities for ancillary services that take account of broad perspectives and specific forms of background inequities; further research on SCD to build health research and policy for this disorder in settings where it is prevalent; and exploring the nature and role of important research-related values held by community members in informing ethical research practice.

9.0 Bibliography

Abelson, J., P. G. Forest, et al. (2003). "Deliberations about deliberative methods: issues in the design and evaluation of public participation processes." Soc Sci Med **57**(2): 239-251.

Abma, T. A., B. Molewijk, et al. (2009). "Good care in ongoing dialogue. Improving the quality of care through moral deliberation and responsive evaluation." Health Care Anal **17**(3): 217-235.

Adeodu, O. O., T. Alimi, et al. (2000). "A comparative study of perception of sickle cell anaemia by married Nigeria rural and urban women." West Afr J Med **19**(1): 1-5.

Akenzua, G. (1990). "Screening for psychosocial dysfunction in children with sickle cell anaemia." Nigerian Journal of Paediatrics **17**(1 & 2): 15-21.

Allotey, P. and D. Reidpath (2001). "Establishing the causes of childhood mortality in Ghana: the 'spirit child'." Soc Sci Med **52**(7): 1007-1012.

Alswaidi, F. M. and S. J. O'Brien (2009). "Premarital screening programme for haemoglobinopathies, HIV and hepatitis virses: review and factors affecting their success." Journal of Medical Screening **16**(1): 22-28.

Appelbaum, P. S., L. H. Roth, et al. (1987). "False hopes and best data: Consent to research and the therapeutic misconception." Hastings Centre Report **17**(2): 20-24.

Assimadi, J., A. Gbadoe, et al. (2000). "The effects of sickle cell disease on families in Togo." Arch Pediatr **7**: 615-620.

Atkin, K., W. I. Ahmad, et al. (1998). "Screening and counselling for sickle cell disorders and thalassaemia: the experience of parents and health professionals." Soc Sci Med **47**(11): 1639-1651.

Bain, B. J. (2009). "Neonatal/newborn haemoglobinopathy screening in Europe and Africa." J Clin Pathol **62**(1): 53-56.

Bamisaiye, A., C. G. Bakare, et al. (1974). "Some social-psychologic dimensions of sickle cell anemia among Nigerians." Clin Pediatr (Phila) **13**(1): 56-59.

Bazuaye, G. and E. Olayemi (2009). "Knowledge and attitude of senior secondary school students in Benin City Nigeria to Sickle Cell Disease." World Journal of Medical Sciences **4**(1): 46-40.

Beauchamp, T. L. and J. F. Childress (2001). Chapter 9: Method and moral justification. Principles of Biomedical Ethics. T. L. Beauchamp and J. F. Childress. Oxford, Oxford University Press.

Beauchamp, T. L. and J. F. Childress (2001). Principles of Biomedical Ethics. Oxford, Oxford University Press.

Belsky, L. and H. S. Richardson (2004). "Medical researchers' ancillary clinical care responsibilities." BMJ **328**(7454): 1494-1496.

Benatar, S. R. (2002). "Reflections and recommendations on research ethics in developing countries." Soc Sci Med **54**(7): 1131-1141.

Benatar, S. R. and P. A. Singer (2000). "A new look at international research ethics." BMJ **321**(7264): 824-826.

Beskow, L. M. and W. Burke (2010). "Offering individual genetic research results: context matters." Sci Transl Med **2**(38): 38cm20.

Borry, P., J. P. Fryns, et al. (2006). "Carrier testing in minors: a systematic review of guidelines and position papers." Eur J Hum Genet **14**(2): 133-138.

Borry, P., H. Nys, et al. (2007). "Carrier testing in minors: conflicting views." Nat Rev Genet **8**(11): 828.

Borry, P., L. Stultiens, et al. (2006). "Presymptomatic and predictive genetic testing in minors: a systematic review of guidelines and position papers." Clin Genet **70**(5): 374-381.

Bosk, C. L. (2002). "Obtaining voluntary consent for research in desperately ill patients." Med Care **40**(9 Suppl): V64-68.

Burgess, M. M. (2004). "Public consultation in ethics: an experiment in representative ethics." J Bioeth Inq **1**(1): 4-13.
Carter, S. M. (2009). "Beware dichotomies and grand abstractions: attending to particularity and practice in empirical bioethics." Am J Bioeth **9**(6-7): 76-77.
CBS, World Bank, et al. (2005). Geographic Dimensions of Well-being in Kenya: Who are where are the poor? Nairobi.
Chilibeck, G., M. Lock, et al. (2011). "Postgenomics, uncertain futures, and the familiarization of susceptibility genes." Soc Sci Med **72**(11): 1768-1775.
Chokshi, D. A. (2005). "Ethical challenges of genomic epidemiology in developing countries." Genomics, Society and Policy **1**(1): 1.
Chokshi, D. A., M. Parker, et al. (2006). "Data sharing and intellectual property in a genomic epidemiology network: policies for large-scale research collaboration." Bull World Health Organ **84**(5): 382-387.
Chokshi, D. A., M. A. Thera, et al. (2007). "Valid consent for genomic epidemiology in developing countries." PLoS Med **4**(4): e95.
Chui, D. H. and G. J. Dover (2001). "Sickle cell disease: no longer a single gene disorder." Curr Opin Pediatr **13**(1): 22-27.
CIOMS (2002). International Ethical Guidelines for Biomedical Research Involving Human Subjects.
Clayton, E. W. and L. F. Ross (2006). "Implications of disclosing individual results of clinical research." JAMA **295**(1): 37; author reply 37-38.
Collins, F. S. (1999). "Shattuck lecture--medical and societal consequences of the Human Genome Project." N Engl J Med **341**(1): 28-37.
Conrad, P. and J. Gabe (1999). Sociological Perspectives on the New Genetics. Oxford, Blackwell Publishers Ltd.
Cowgill, K. D., M. Ndiritu, et al. (2006). "Effectiveness of Haemophilus influenzae type b Conjugate vaccine introduction into routine childhood immunization in Kenya." Jama **296**(6): 671-678.
Davis, D. S. (2007). "The changing face of "misidentified paternity"." J Med Philos **32**(4): 359-373.
de Melo-Martin, I. and A. Ho (2008). "Beyond informed consent: the therapeutic misconception and trust." J Med Ethics **34**(3): 202-205.
de Vries, R. and B. Gordijn (2009). "Empirical ethics and its alleged meta-ethical fallacies." Bioethics **23**(4): 193-201.
de Vries, R., L. Turner, et al. (2006). "Social science and bioethics: the way forward." Sociol Health Illn **28**(6): 665-677.
Dennis-Antwi, J. A., L. Culley, et al. (2011). "'I can die today, I can die tomorrow': lay perceptions of sickle cell disease in Kumasi, Ghana at a point of transition." Ethn Health **16**(4-5): 465-481.
Dickert, N., K. DeRiemer, et al. (2007). "Ancillary-care responsibilities in observational research: two cases, two issues." Lancet **369**(9564): 874-877.
Dickert, N. and J. Sugarman (2005). "Ethical goals of community consultation in research." Am J Public Health **95**(7): 1123-1127.
Dixon-Woods, M., A. Suokas, et al. (2009). "An ethnographic study of classifying and accounting for risk at the sharp end of medical wards." Soc Sci Med **69**(3): 362-369.
Dixon-Woods, M. and C. Tarrant (2009). "Why do people cooperate with medical research? Findings from three studies." Soc Sci Med **68**(12): 2215-2222.
Dunn, M. and J. Ives (2009). "Methodology, epistemology, and empirical bioethics research: a constructive/ist commentary." Am J Bioeth **9**(6-7): 93-95.
Duster, T. (2003). Backdoor to Eugenics. New York, Routledge**:** 47.
Eastern Biotech and Life Sciences. (2011). "Premarital Screening." 2011, from http://www.easternbiotech.com/Pre-marital-Screening.php.

Emanuel, E. J., D. Wendler, et al. (2004). "What makes clinical research in developing countries ethical? The benchmarks of ethical research." J Infect Dis **189**(5): 930-937.
Fabsitz, R. R., A. McGuire, et al. (2010). "Ethical and practical guidelines for reporting genetic research results to study participants: updated guidelines from a National Heart, Lung, and Blood Institute working group." Circ Cardiovasc Genet **3**(6): 574-580.
Featherstone, K., P. Atkinson, et al. (2006). Mutual Surveillance. Risky Relations. Oxford, Berg**:** 73-91.
Featherstone, K., P. Atkinson, et al. (2006). Practical Ethics and Disclosure. Risky Relations. Oxford, Berg**:** 91-116.
Fernandez, C. V., E. Kodish, et al. (2003). "Informing study participants of research results: an ethical imperative." IRB **25**(3): 12-19.
Finkler, K. (2005). "Family, kinship, memory and temporality in the age of the new genetics." Soc Sci Med **61**(5): 1059-1071.
Flick, U. (2006). An Introduction to Qualitative Research. London, Sage Publications Ltd.
Fryer-Edwards, K. and S. M. Fullerton (2006). "Relationships with test-tubes: where's the reciprocity?" Am J Bioeth **6**(6): 36-38; author reply W10-32.
Fulford, K., D. Dickenson, et al. (2002). Introduction. Healthcare Ethics and Human Values. K. Fulford, D. Dickenson and T. Murray. Oxford, Blackwell.
Fullwiley, D. (2006). "Biosocial suffering:Order and illness in urban West Africa." BioSocieties **1**: 421-438.
Geelen, E., I. Van Hoyweghen, et al. (2011). "Making genetics not so important: family work in dealing with familial hypertrophic cardiomyopathy." Soc Sci Med **72**(11): 1752-1759.
Geissler, P. W., A. Kelly, et al. (2008). "'He is now like a brother, I can even give him some blood'--relational ethics and material exchanges in a malaria vaccine 'trial community' in The Gambia." Soc Sci Med **67**(5): 696-707.
Geissler, P. W. and R. Pool (2006). "Editorial: Popular concerns about medical research projects in sub-Saharan Africa--a critical voice in debates about medical research ethics." Trop Med Int Health **11**(7): 975-982.
Georgetown University Workshop (2008). The ancillary-care obligations of medical researchers working in developing countries. PLoS Med. Participants in the 2006 Georgetown University Workshop on the Ancillary-Care Obligations of Medical Researchers Working in Developing Countries. **5:** e90.
Ghana Sickle Cell Foundation. (2010). "Home page." from http://www.sicklecellghana.org/index.php?option=com_content&view=article&id=24&Itemid=28.
Gikonyo, C., P. Bejon, et al. (2008). "Taking social relationships seriously: lessons learned from the informed consent practices of a vaccine trial on the Kenyan Coast." Soc Sci Med **67**(5): 708-720.
Gilson, L. (2003). "Trust and the development of health care as a social institution." Soc Sci Med **56**(7): 1453-1468.
Gilson, L. (2005). "Editorial: building trust and value in health systems in low- and middle-income countries." Soc Sci Med **61**(7): 1381-1384.
Global Forum for Health Research (2000). The 10/90 Report on Health Research. S. Jupp. Geneva, Global Forum for Health Research**:** xiii.
Goffman, E. (1963). Stigma and Social Identity. Stigma: Notes on the management of a spoiled identity. New Jersey, Penguin Books**:** 12.
Greely, H. T. (2007). "The uneasy ethical and legal underpinnings of large-scale genomic biobanks." Annu Rev Genomics Hum Genet **8**: 343-364.
Green, M. and A. J. Solnit (1964). "Reactions to the Threatened Loss of a Child: A Vulnerable Child Syndrome. Pediatric Management of the Dying Child, Part Iii." Pediatrics **34**: 58-66.

Hartley, J. (2009). "Genetic Identity." Arabian Business.com, from http://www.arabianbusiness.com/genetic-identity-41149.html.
Hedgecoe, A. (2007). Medical Sociology and the Redundancy of Empirical Ethics. Principles of Health Care Ethics.
Hedgecoe, A. M. (2004). "Critical bioethics: beyond the social science critique of applied ethics." Bioethics **18**(2): 120-143.
Hoedemaekers, R., B. Gordijn, et al. (2006). "The complexities of ethical evaluation of genomics research." HEC Forum **18**(1): 18-36.
Hoffmaster, B. and C. Hooker (2009). "How experience confronts ethics." Bioethics **23**(4): 214-225.
Hoffmaster, B. and C. Hooker (2009). "What empirical research can do for bioethics." Am J Bioeth **9**(6-7): 72-74.
Holland, S. (2007). Part One Moral and Political Philosophy: Political Philosophy. Public Health Ethics. S. Holland. Cambridge, Polity Press.
Hope, T. (2004). Medical Ethics: A very short introduction. Oxford, Oxford University Press.
Hope, T. (2008). The possibility of empirical psychiatric ethics. Empirical Ethics in Psychiatry. T. H. G. Widdershoven, J. McMillan and L van der Scheer.
Horstman, K. and K. Finkler (2011). "Genetics, health care, family and kinship in a global perspective: situated processes of co-construction." Soc Sci Med **72**(11): 1739-1742.
Ijsselmuiden, C. B., N. E. Kass, et al. (2010). "Evolving values in ethics and global health research." Glob Public Health **5**(2): 154-163.
Ives, J. (2008). "'Encounters with experience': empirical bioethics and the future." Health Care Anal **16**(1): 1-6.
Ives, J. and H. Draper (2009). "Appropriate methodologies for empirical bioethics: it's all relative." Bioethics **23**(4): 249-258.
Ives, J., H. Draper, et al. (2008). "Becoming a father/refusing fatherhood: an empirical bioethics approach to paternal responsibilities and rights." Clinical Ethics **3**: 75-84.
Ives, J. and M. Dunn (2010). "Who's arguing? A call for reflexivity in bioethics." Bioethics **24**(5): 256-265.
Juengst, E. T. (2000). "What "Community Review" can and cannot do." Journal of Law, Medicine and Ethics **28**(910): 52-54.
Kafando, E., M. Sawadogo, et al. (2005). "Neonatal screening for sickle cell disorders in Ouagadougou, Burkina Faso: a pilot study." J Med Screen **12**(3): 112-114.
Kamuya, D. (2007). Community engagement in biomedical research in a low-income setting: a preliminary evaluation of a new network of community representatives. London School of Hygiene and Tropical Medicine. London, University of London. MPH dissertation http://www.kemri-wellcome.org/dissertations/MSc_2007_Kamuya_DM.pdf
Kamuya, D., V. Marsh, et al. (2011). "What we learned about voluntariness and consent: incorporating "background situations" and understanding into analyses." Am J Bioeth **11**(8): 31-33.
Kark, J. (2000). "Sickle Cell Trait." Retrieved 28th September 2011, from http://sickle.bwh.harvard.edu/sickle_trait.html.
Kass, N. E., J. Sugarman, et al. (1996). "Trust, The fragile foundation of contemporary biomedical research." Hastings Cent Rep **26**(5): 25-29.
Kim, S. Y., R. A. Uhlmann, et al. (2010). "Deliberative assessment of surrogate consent in dementia research." Alzheimers Dement **6**(4): 342-350.
King, L., R. Fraser, et al. (2007). "Newborn sickle cell disease screening: the Jamaican experience (1995-2006)." J Med Screen **14**(3): 117-122.
Kitzinger, J. (2006). Focus Groups. Qualitative Research in Health Care. C. Pope and N. Mays. Oxford, Blackwell Publishing Ltd**:** 21-32.

Knoppers, B. M., Y. Joly, et al. (2006). "The emergence of an ethical duty to disclose genetic research results: international perspectives." Eur J Hum Genet **14**(11): 1170-1178.
Kon, A. A. (2009). "The role of empirical research in bioethics." Am J Bioeth **9**(6-7): 59-65.
Laird, L., C. Dezateux, et al. (1996). "Neonatal screening for sickle cell disorders: what about the carrier infants?" BMJ **313**(7054): 407-411.
Lavery, J. V., S. V. Bandewar, et al. (2010). "'Relief of oppression': an organizing principle for researchers' obligations to participants in observational studies in the developing world." BMC Public Health **10**: 384.
Leach, M. and J. Fairhead (2006). "Being "with MRC": Infant care and the social meanings of cohort membership in Gambia's plural therapeutic landscapes."
Leget, C., P. Borry, et al. (2009). "'Nobody tosses a dwarf!' The relation between the empirical and the normative reexamined." Bioethics **23**(4): 226-235.
Lindemann Nelson, H. and J. Lindemann Nelson (1995). The Patient in the Family: An Ethics of Medicine and Families. London, Routledge.
Lippman, A. (1992). "Led (astray) by genetic maps: the cartography of the human genome and health care." Soc Sci Med **35**(12): 1469-1476.
London, A. J. (2005). "Justice and the human development approach to international research." Hastings Cent Rep **35**(1): 24-37.
Lucassen, A. and M. Parker (2001). "Revealing false paternity: some ethical considerations." Lancet **357**(9261): 1033-1035.
Macklin, R. (1999). Against Relativism: Cultural Diversity and the Search for Ethical Universals in Medicine. Oxford, Oxford University Press.
Makani, J., T. N. Williams, et al. (2007). "Sickle cell disease in Africa: burden and research priorities." Ann Trop Med Parasitol **101**(1): 3-14.
Markel, H. (1992). "The stigma of disease: implications of genetic screening." Am J Med **93**(2): 209-215.
Marsh, V., D. Kamuya, et al. (2008). "Beginning community engagement at a busy biomedical research programme: experiences from the KEMRI CGMRC-Wellcome Trust Research Programme, Kilifi, Kenya." Soc Sci Med **67**(5): 721-733.
Marsh, V. M., D. K. Kamuya, et al. (2011a). "Working with Concepts: The Role of Community in International Collaborative Biomedical Research." Public Health Ethics **4**(1): 26-39.
Marsh, V. M., D. M. Kamuya, et al. (2010). "Experiences with community engagement and informed consent in a genetic cohort study of severe childhood diseases in Kenya." BMC Med Ethics **11**: 13.
Marsh, V. M., D. M. Kamuya, et al. (2011b). "'All her children are born that way': gendered experiences of stigma in families affected by sickle cell disorder in rural Kenya." Ethn Health **16**(4-5): 343-359.
Marsh, V. M., W. M. Mutemi, et al. (1999). "Changing home treatment of childhood fevers by training shop keepers in rural Kenya." Trop Med Int Health **4**(5): 383-389.
Marshall, P. A. and C. Rotimi (2001). "Ethical challenges in community-based research." Am J Med Sci **322**(5): 241-245.
Masiye, F., N. Kass, et al. (2008). "Why mothers choose to enrol their children in malaria clinical studies and the involvement of relatives in decision making: evidence from Malawi." Malawi Med J **20**(2): 50-56.
Memish, Z. A. and M. Y. Saeedi (2011). "Six-year outcome of the national premarital screening and genetic counseling program for sickle cell disease and beta-thalassemia in Saudi Arabia." Ann Saudi Med **31**(3): 229-235.
Miller, F. A. (2009). "The complex promise of newborn screening." Indian J Med Ethics **6**(3): 142-148.

Miller, F. A., R. Z. Hayeems, et al. (2009). "Clinical obligations and public health programmes: healthcare provider reasoning about managing the incidental results of newborn screening." J Med Ethics **35**(10): 626-634.
Miller, F. A., M. Paynter, et al. (2010). "Understanding sickle cell carrier status identified through newborn screening: a qualitative study." Eur J Hum Genet **18**(3): 303-308.
Miller, F. A., J. S. Robert, et al. (2009). "Questioning the consensus: managing carrier status results generated by newborn screening." Am J Public Health **99**(2): 210-215.
Miller, P. B. and C. Weijer (2006). "Trust based obligations of the state and physician-researchers to patient-subjects." J Med Ethics **32**(9): 542-547.
Molewijk, A. C., T. Abma, et al. (2008). "Teaching ethics in the clinic. The theory and practice of moral case deliberation." J Med Ethics **34**(2): 120-124.
Molewijk, B., A. M. Stiggelbout, et al. (2004). "Empirical data and moral theory. A plea for integrated empirical ethics." Med Health Care Philos **7**(1): 55-69.
Molyneux, C. S. and C. Gikonyo (2005). Internal report on baseline surveys to evaluate community understanding of KEMRI and health research. Kilifi, KEMRI CGMRC.
Molyneux, C. S., J. Goudge, et al. (2009). "Conducting health-related social science research in low-income settings: Ethical dilemmas faced in Kenya and South Africa." Journal of International Development **21**: 309-326.
Molyneux, C. S., G. Murira, et al. (2002). "Intra-household relations and treatment decision-making for childhood illness: a Kenyan case study." J Biosoc Sci **34**(1): 109-131.
Molyneux, C. S., N. Peshu, et al. (2004). "Understanding of informed consent in a low-income setting: three case studies from the Kenyan Coast." Soc Sci Med **59**(12): 2547-2559.
Molyneux, C. S., N. Peshu, et al. (2005a). "Trust and informed consent: insights from community members on the Kenyan coast." Soc Sci Med **61**(7): 1463-1473.
Molyneux, C. S., D. R. Wassenaar, et al. (2005b). "'Even if they ask you to stand by a tree all day, you will have to do it (laughter)...!': community voices on the notion and practice of informed consent for biomedical research in developing countries." Soc Sci Med **61**(2): 443-454.
Molyneux, S., D. Kamuya, et al. (2010). "Community members employed on research projects face crucial, often under-recognized, ethical dilemmas." Am J Bioeth **10**(3): 24-26.
Munyanganizi, R., F. Cotton, et al. (2006). "Red blood cell disorders in Rwandese neonates: screening for sickle cell disease and glucose-6-phosphate dehydrogenase deficiency." J Med Screen **13**(3): 129-131.
Mutesa, L., F. Boemer, et al. (2007). "Neonatal screening for sickle cell disease in Central Africa: a study of 1825 newborns with a new enzyme-linked immunosorbent assay test." J Med Screen **14**(3): 113-116.
Mwenesi, H., T. Harpham, et al. (1995). "Child malaria treatment practices among mothers in Kenya." Soc Sci Med **40**(9): 1271-1277.
Naveed, M., S. Phadke, et al. (1992). "Sociocultural problems in genetic counselling." British Medical Journal **29**: 140.
NCOB (2006). Genetic screening: A supplement to the 1993 report by the Nuffield Council on Bioethics. Cardiff, Clyvedon Press Ltd.
NCST (2004). Guidelines for the Ethical Conduct of Biomedical Research Involving Human Subjects in Kenya. Nairobi, National Council for Science and Technology, Republic of Kenya.
Nigeria Sickle Cell Foundation. (2011). "Specialised services." from http://www.sicklecellfoundation.com/services.php.
NIH, C. D. C. (1987) "Newborn Screening for Sickle Cell Disease and Other Hemoglobinopathies." NIH Consensus Statements.
Nyamongo, I. K. (2002). "Health care switching behaviour of malaria patients in a Kenyan rural community." Soc Sci Med **54**(3): 377-386.

Nzewi, E. (2001). "Malevolent ogbanje: recurrent reincarnation or sickle cell disease?" Soc Sci Med **52**(9): 1403-1416.
Ohaeri, J. (2002). "Psychological burden of sickle cell disease on caregivers in a Nigerian setting." Journal of National Medical Assocation **94**(12): 1058 - 1070.
Ohaeri, J. U. and W. A. Shokunbi (2001). "Attitudes and beliefs of relatives of patients with sickle cell disease." East Afr Med J **78**(4): 174-179.
Ohaeri, J. U., W. A. Shokunbi, et al. (1995). "The psychosocial problems of sickle cell disease sufferers and their methods of coping." Soc Sci Med **40**(7): 955-960.
Ohene-Frempong, K. (2005, 2nd March 2005). "Newborn screening for Sickle Cell Disease in Ghana." Retrieved 28th September 2011, 2011, from http://www.ghanaweb.com/GhanaHomePage/NewsArchive/artikel.php?ID=76368.
Ossorio, P. N. (2006). "Letting the gene out of the bottle: a comment on returning individual research results to participants." Am J Bioeth **6**(6): 24-25; author reply W10-22.
Parker, M. (2007a). "Ethnography/ethics." Soc Sci Med **65**(11): 2248-2259.
Parker, M. (2007b). Deliberative Ethics. Principles of Health Care Ethics
R. E. Ashcroft, A. Dawson, H. Draper and J. R. McMillan. Chichester, Wiley**:** 185-191.
Parker, M. (2009). "Two concepts of empirical ethics." Bioethics **23**(4): 202-213.
Parker, M., S. J. Bull, et al. (2009). "Ethical data release in genome-wide association studies in developing countries." PLoS Med **6**(11): e1000143.
Parker, M. and A. Lucassen (2003). "Concern for families and individuals in clinical genetics." J Med Ethics **29**(2): 70-73.
Parker, M. and A. M. Lucassen (2004). "Genetic information: a joint account?" British Medical Journal **329**(7458): 165-167.
Parker, R. and P. Aggleton (2003). "HIV and AIDS-related stigma and discrimination: a conceptual framework and implications for action." Soc Sci Med **57**(1): 13-24.
Parkin, D. (1991). Alternative selves: Invasions and cures. Sacred Void: Spatial images of work and ritual among the Giriama of Kenya. Cambridge, Cambridge University Press**:** 160-191.
Parkin, D. (1991). From west to east: The works of marriage. Sacred Void: Spatial images of work and ritual among the Giriama of Kenya. Cambridge, Cambridge University Press**:** 84.
Patton, M. (1999). "Enhancing the credibility and quality of qualitative research." Health Services Research **34**(5).
Phoenix, D. D., S. M. Lybrook, et al. (1995). "Sickle cell screening policies as portent: how will the human genome project affect public sector genetic services?" J Natl Med Assoc **87**(11): 807-812.
Pope, C. and N. Mays (2006). Quality in Qualitative Health Research. Qualitative Research in Health Care. C. Pope and N. Mays. Oxford, Blackwell**:** 82-012.
Rahimy, M. C., A. Gangbo, et al. (2009). "Newborn screening for sickle cell disease in the Republic of Benin." J Clin Pathol **62**(1): 46-48.
Rapp, R. (1988). "Chromosomes and communication: the discourse of genetic counseling." Medical Anthropology Quarterly **2**(2): 143-157.
Ravitsky, V. and B. S. Wilfond (2006). "Disclosing individual genetic results to research participants." Am J Bioeth **6**(6): 8-17.
Richards, M. P. M. (1993). "The new genetics: some issues for social scientists." Sociology of Health and Illness **15**(5): 567.
Richardson, H. S. (2007). "Gradations of researchers' obligation to provide ancillary care for HIV/AIDS in developing countries." Am J Public Health **97**(11): 1956-1961.
Richardson, H. S. (2008). "Incidental findings and ancillary-care obligations." J Law Med Ethics **36**(2): 256-270, 211.

Richardson, H. S. and L. Belsky (2004). "The ancillary-care responsibilities of medical researchers. An ethical framework for thinking about the clinical care that researchers owe their subjects." Hastings Cent Rep **34**(1): 25-33.
Ross, L. F. (1996). "Disclosing misattributed paternity." Bioethics **10**(2): 114-130.
Sadarangani, M., J. Makani, et al. (2009). "An observational study of children with sickle cell disease in Kilifi, Kenya." Br J Haematol **146**(6): 675-682.
Schiffirin, D., D. Tannen, et al. (2001). The Handbook of Discourse Analysis. Oxford, Blackwells.
Shalowitz, D. I. and F. G. Miller (2005). "Disclosing individual results of clinical research: implications of respect for participants." JAMA **294**(6): 737-740.
Sharp, R. R. and M. W. Foster (2000). "Involving study populations in the review of genetic research." J Law Med Ethics **28**(1): 41-51, 43.
Sharp, R. R. and M. W. Foster (2006). "Clinical utility and full disclosure of genetic results to research participants." Am J Bioeth **6**(6): 42-44; author reply W10-42.
Shaw, A. (2006). The Contingency of the 'Genetic Link' in Constructions of Kinship and Inheritance - An Anthropological Perspective. Freedom and Responsibility in Reproductive Choice. J. Spencer and A. Du Bois-Pedain. Cambridge, Hart Publishing**:** 77.
Shaw, A. (2006). The Contingency of the Genetic Link in Constructions of Kinship and Inheritance - An Anthropological Perspective. Freedom and Responsibility in Reproductive Choice. J. Spencer and A. Du Bois-Pedain. Cambridge, Hart Publishing**:** 73-90.
Sheehan, M. (2007). Moral Relativism. Principles of Health Care Ethics.
Singer, P. (2011). Rich and Poor. Practical Ethics. Cambridge, Cambridge University Press**:** 199-202.
Snow, R. W., N. Peshu, et al. (1992). "The role of shops in the treatment and prevention of childhood malaria on the coast of Kenya." Trans R Soc Trop Med Hyg **86**(3): 237-239.
Sugarman, J., N. Kass, et al. (2009). "Categorizing empirical research in bioethics: why count the ways?" Am J Bioeth **9**(6-7): 66-67.
Tangwa, G. (1999). "Globalisation or Westernisation? Ethical concerns in the whole bio-business." Bioethics **13**(3/4): 218 - 226.
Tekola, F., S. Bull, et al. (2009). "Impact of social stigma on the process of obtaining informed consent for genetic research on podoconiosis: a qualitative study." BMC Med Ethics **10**: 13.
Telfer, P., P. Coen, et al. (2007). "Clinical outcomes in children with sickle cell disease living in England: a neonatal cohort in East London." Haematologica **92**(7): 905-912.
Tsaras, G., A. Owusu-Ansah, et al. (2009). "Complications associated with sickle cell trait: a brief narrative review." Am J Med **122**(6): 507-512.
Turney, L. (2005). "The incidental discovery of nonpaternity through genetic carrier screening: an exploration of lay attitudes." Qual Health Res **15**(5): 620-634.
Turney, L. (2005). "Paternity Secrets: Why some women don't tell." Journal of Family Studies **11**(2): 227-248.
UNAIDS (2000). HIV and AIDS related stigmatization, discrimination and denial: forms, contexts and determinants. Research studies from Uganda and India. UNAIDS. Geneva, UNAIDS**:** 13.
UNAIDS (2005). HIV related stigma, discrimination and human rights violations: Case studies of successful programmes. P. Aggleton, K. Wood, A. Malcolm and R. Parker. Geneva, UNAIDS.
Virtual Kenya. (2011). "Virtual Kenya." from http://www.virtualkenya.org/maps/map-images/422-chapter-2-spatial-patterns-of-poverty-and-human-well-being.
Wang, V. and F. H. Marsh (1992). "Ethical principles and cultural integrity in health care delivery: Asian ethnocultural perspectives in genetic services." J Genet Couns **1**(1): 81-92.

Weatherall, D. J., O. O. Akinyanju, et al. (2006). Inherited Disorders of Hemoglobin. Disease Control Priorities in Developing Countries. D. T. Jamison, J. G. Breman, A. R. Measham et al. Oxford, Oxford University Press

World Bank**:** 663-680.

Weatherall, D. J. and J. B. Clegg (2001). "Inherited haemoglobin disorders: an increasing global health problem." Bull World Health Organ **79**(8): 704-712.

Weijer, C. and E. J. Emanuel (2000). "Ethics. Protecting communities in biomedical research." Science **289**(5482): 1142-1144.

Wertheimer, A. (2008). Exploitation in Clinical Research. Exploitation and Developing Countries: The Ethics of Clinical Research. J. Hawkins and E. Emanuel. Princeton, Princeton University Press**:** 63-104.

Wertz, D. and J. Fletcher (2004). Genetics and Ethics in Global Perspective. Dordrecht, Kluwer Academic Publishers.

Wertz, D. C., J. C. Fletcher, et al. (1990). "Medical geneticists confront ethical dilemmas: cross-cultural comparisons among 18 nations." Am J Hum Genet **46**(6): 1200-1213.

WHO (2002). Genomics and World Health: Report of the Advisory Committee on Heath Research. Geneva, World Health Organization.

WHO (2003). Genetic Databases:Assessing the Benefits and the Impact on Patient and Human Rights. Geneva, World Health Organization.

WHO (2006). Medical genetic services in developing countries: the ethical, legal and social implications of genetic testing and screening. Geneva.

Widdershoven, G., T. Abma, et al. (2009). "Empirical ethics as dialogical practice." Bioethics **23**(4): 236-248.

Widdershoven, G., B. Molewijk, et al. (2009). "Improving care and ethics: a plea for interactive empirical ethics." Am J Bioeth **9**(6-7): 99-101.

Widdershoven, G. A. (2007). "How to combine hermeneutics and Wide Reflective Equilibrium?" Med Health Care Philos **10**(1): 49-52.

Williams, T. N., T. W. Mwangi, et al. (2005). "Sickle cell trait and the risk of Plasmodium falciparum malaria and other childhood diseases." J Infect Dis **192**(1): 178-186.

Williams, T. N., T. W. Mwangi, et al. (2005). "Negative epistasis between the malaria-protective effects of alpha+-thalassemia and the sickle cell trait." Nat Genet **37**(11): 1253-1257.

Wolf, S. M., J. Paradise, et al. (2008). "The law of incidental findings in human subjects research: establishing researchers' duties." J Law Med Ethics **36**(2): 361-383, 214.

World Medical Association (2004). Helsinki Declaration. W. M. Association.

Young, A., S. J. Kim, et al. (2009). "Discovering misattributed paternity in living kidney donation: prevalence, preference, and practice." Transplantation **87**(10): 1429-1435.

Appendix A1: Phase One Interview Guide Families with SCD/non KGBC Part 1

The story of SCD in this family	
Which children affected?	Names and ages now, ages when noticed problem (try to find out about any that have died - CARE)
By end of interview: age, education, religion and occupation	Avoid direct questions where possible, ask at end if not come out
FROM THE BEGINNING/BEFORE TESTING FOR 1ST OR ONLY CHILD (If 1st affected child died, ask for oldest living child but come back to first experiences later) ***When did you first realise there was a problem and what did you notice?***	
SIGNS at start	All signs e.g. swelling (where?), pain, crying, pale, thin, not eating, how long did it last?
Ideas about **CAUSES** at first and later, including changes	Family line, curse (how?), environment, mix? Any change? What led to changes?
MANAGEMENT – what done, how decided first and later, including changes	*For 1st child in this early stage, what done first, and then subsequently:* • Who consulted, where, when, what advice or explanations given, what treatment given, did it work well? • Who influenced treatment seeking decision and later changes in this? • What factors influenced treatment seeking decision and later changes in this: distance, time, cost, providers' attitudes, own experience/beliefs?
Nature and timing of next episodes	Roughly: how many episodes per month/year; signs?
Parent's **FEELINGS**: worries, fears	What concerns: health status, telling others, future children, affording/managing treatment, looks like HIV, other?
NAME given to illness	Before given info at testing, where did you first hear this name?
WHEN TESTED	
When was child/children tested?	Age(s) at testing and who first? (the oldest?)
For 1st child tested: Where, when, who went with child for test?	Open description
HOW DECIDED to have test done	Who influenced this decision, and how? What factors considered in making decision?
What **INFO GIVEN** before test	About illness or test
What **INFO GIVEN** after test, and when	• How long waited for results, and how given back • What information given about result – status, meaning of this, genetic cause, management for future, permanent problem
Parents immediate **FEELINGS** about test result	What concerns: child's health status, telling others, future children, affording/managing treatment, looks like HIV
For any other children tested: any **DIFFERENCES**?	Where tested, info given before and after testing, and parents' reactions/concerns about results

SINCE TESTING UP TO NOW: ***REFLECT ON ILLNESS IN FAMILY OVER TIME SINCE 1ST CHILD - CHANGES?***	
GENERAL HEALTH for all affected children	Including carefully find out if any deaths
For all children: What **MANAGEMENT** followed now, any **CHANGES** over time, **WHY** these made, any **DIFFERENCES** between children	• *If for child who died – ask about this in general* • Who usually consulted (formal public, formal private, traditional, home etc), where, when, what advice or explanations given, what treatment given, did it work well? Is there MIXTURE? • If traditional: how much spent? • If clinic: how often? • Reasons for change – observed effects, others influence, related to changes in ideas about cause, others? • Reasons for differences in management between children
Ideas about **CAUSE** – changes?	• Own ideas: Family line, curse (how?), environment, mix? • What about older relatives affected: what do you think was cause? What did they think was cause? • If "curse" – what due to? Any taboos? Specific to SCD? • Any change? What led to changes? • Mixture of beliefs about cause and management? Describe how this works
NAME given to illness – changes?	• Where did you first hear this name? • Ever heard other names e.g. from older relatives? • Describe what these other named "illnesses" are like – signs? cause? treatment?
Parent's **FEELINGS** over time – changes?	• What concerns: health status, telling others, future children, affording/managing treatment, looks like HIV • Changes? How and why?
Effect on **RELATIONSHIPS** in family over time	• Find out: how many people in hh and how many children (ages and gender) in this family • Impact on child – siblings; child – parent; child-friends; parent – parent; other family-family; family – others *(work, school, neighbours)* • If any co-wives, wife-wife? • Is there anything someone in family used to do but has now stopped/changed? What and why? *GENDER?? STIGMA??*
OTHERS AFFECTED	
Any other affected families known?	Who, where living, how many, how affected, what signs – sound like same illness? Avoid personal details.
Any relationship with these families	Social, support, related?
Other names for illness ?	As for families – explore what any named illnesses are like
How managed by others	Any differences to own?
Others see as cause?	Other explanations of cause they know about
VALUE OF TESTING	
What positive/negative outcomes of testing? Would you recommend to a friend to have child tested for SCD? Why/why not What benefits and problems in going to clinic?	

Appendix A2: Phase One: Interview Guide Families with SCD/KGBC Part 1

A. Visit by Genetics FW

What happened when FW visited

- When did they visit? Who met them? Mother/father/other (same as now?)
- What did FW explain? What was understood about AIM of visit in relation to:
    - KEMRI/research (*note: also discussed in introduction*)
    - genetics research
    - SCD/ testing

Agreement to participate

- What was parents' response to information given by FW before testing? What benefits seen? What costs/ concerns seen? HEEL PRICK – concerns? Rumours?
- Why decided to participate?
- How many visits made, why if more than one?

Awareness of SCD before testing

- Had they heard of SCD before? Any other child affected? (*links to section B*). NB Any deaths - CARE
- If other child affected, did they think of testing this one...why/why not?
- Any signs of ill health before? What was thought to be problem?

What understood when results brought?

- When were results brought, how did parents get result (direct/indirect)?
- What was understood?
- What response of mother/father/others? CONFLICT IF WELL AT TIME: feelings/thoughts?
- What happened after given result, and why?

B. Story of SCD/signs in this child and any other children

SIGNS at start

- Has this child had any persistent/recurrent signs of illness since birth?
- What signs e.g. swelling (where?), pain, crying, pale, thin, not eating, how long did it last?
- Other children in this family: ages, names, positions...*note down*
- Any other child with same/similar signs
- Who was first affected?
- **FOR ALL AFFECTED CHILDREN, INCLUDING INDEX CHILD: General health?** ***If any child died, ask general questions only***

First decision to test (if not done as part of genetics study)

- Where, when, who went with child for test? Who influenced and what factors considered in making this decision? What info given? (status, meaning of this, genetic cause, management for future, permanent problem)
- How long waited for results, and how given back

What MANAGEMENT- at first and later, any CHANGES over time, WHY made, any DIFFERENCES between children

- Who consulted (formal public, formal private, traditional, home etc), where, when, what advice or explanations given, what treatment given, did it work well? Is there MIXTURE?
    - If traditional: how much spent?
    - If clinic: how often? Date of last visit?
- Reasons for change – observed effects, others influence, related to changes in ideas about cause, others?
- Reasons for differences in management between children

Ideas about CAUSE – at first and later (any changes)?

- Own ideas: Family line, curse (how?), environment, mix?
- What about older relatives affected: what do you think was cause? What did they think was cause?
- If "curse" – what due to? Any taboos? Are these specific to SCD?
- Any change? What led to changes?
- Mixture of beliefs about cause and management? Describe how this works

NAME given to illness – changes?

- Where did you first hear this name? Ever heard other names e.g. from older relatives?
- Describe what these other named "illnesses" are like – signs? cause? treatment?

Parent's FEELINGS over time – changes?

- What concerns: health status, telling others, future children, affording/managing treatment, like HIV
- Changes? How and why?

Effect on RELATIONSHIPS in family over time ***GENDER?? STIGMA??***

- Find out: how many people in hh and how many children (ages and gender) in this family
- Impact on child – siblings; child – parent; child-friends; parent – parent; other family-family; family – others *(work, school, neighbours)*. If any co-wives, wife-wife?
- Is there anything someone in family used to do but has now stopped/changed? What and why?
- How do family meet extra needs? COPING

C. OTHER AREAS

- Any other affected families known?
    - Who, where living, how many, how affected, what signs – sound like same illness? Avoid personal details.
    - Any relationship with these families: Social, support, related?
- Names/ how managed/cause for illness by others – same or different?
- What positive/negative outcomes of testing?
- Would you recommend to a friend to have child tested for SCD? Why, why not?
- What benefits and problems in going to clinic?

Appendix A3: Phase One: Interview Guide Families with SCD Part 2 (KGBC and non KGBC)

A. Story of SCD in family

Additional questions/clarifications from last visit

[insert after Part 1 interview conducted]

B. Talking about inheritance

TOPIC	PROBE
For SCD in this family: explore who affected and what explanations for these individuals being affected	• Map family tree to show who known to have SCD • How could this affect these individuals and not others? • Is it being passed on from one to another? • Probe for beliefs about mechanisms and materials that are believed to be possible (direct physical; indirect non physical e.g. thoughts, curses, spiritual; other?)and HOW this happens
For recognised physical characteristic in own family	• Use cards to show inheritance of height in family • Probe for beliefs about mechanisms and materials that are believed to be possible (direct physical; indirect non physical e.g. thoughts, curses, spiritual; other?)and HOW this happens • Probe for gendered role of parents – same, different – how? Is this random or tied to certain inherited characteristics?
For SCD	• Probe for gendered role of parents – same, different – how? • Contrast/compare with beliefs for role of mother and father for inheritance of height • Explore any gendered differences

Appendix A4: Phase One: Interview Guide Families in KGBC/Non SCD

What happened when FW visited?

- When did they visit? Who met them? Mother/father/other (same as now?) - for each visit made
- What did FW explain? What was understood about AIM of visit in relation to:
    - KEMRI/research (*note: also discussed in introduction*)
    - genetics research
    - SCD/ testing

Agreement to participate: Why decided to participate?

- What was parents' response to information given by FW before testing?
- Any previous knowledge on KGBC? Where from? How did this influence? (e.g. sensitisation meetings, neighbours)
- What benefits seen?
- What costs/ concerns seen? HEEL PRICK – concerns? Rumours?
- How many visits made, why if more than one?
- Where were tests done on sample?

Awareness of SCD before testing

- Had they heard of SCD before?
- Ideas about **CAUSE**
    - Own ideas: Family line, curse (how?), environment, mix?
    - What about older relatives affected: what do you think was cause? What cause?
    - If "curse" – what due to? Any taboos? Are these specific to SCD?
- **NAME** given to illness: Where did you first hear this name? Ever heard other names e.g. from relatives?
- **MANAGEMENT:** Who usually consulted (formal public, formal private, traditional, home etc) and why?
- **RELATIONSHIPS with affected families? *STIGMA??*** Impact on child / siblings; child/parent; child/friends; parent / parent; other family/family; family/ others *(work, school, neighbours)*. If co-wives, wife/wife?

What understood when results brought?

- When were results brought, how did parents get result (direct/indirect)?
- What was understood?
- What was response of mother/father/others?
- What happened after given result, and why?

What views about KEMRI's activities in the area?

- What is KEMRI? Type of organisation; main aims
- What concerns about KEMRI's work?
- What benefits seen for KEMRI's work?
- Have they participated in any other research? Which? What happened?
- Would they participate in research in future? Why/why not? What type of research?

Appendix B1: Final Codes for analysis in Phase One

1. KEMRI

1.1 Nature
- Aims
  - As long standing inst
  - As research inst
  - As service or aid provider
  - Doing good
  - No idea/NGO
  - Understanding consent
  - Utafiti/uchunguzi
- Compete TH
- It's for you
- Many studies
- Motorbikes
- Relationship to KDH
- Staff relationships
- *Wazungu*

1.2 Rumours/concerns
- Bednets
- Blood taken
- Contraception
- Devils and snakes
- Different opinions
- Fears non specific
- Follow up at home
- Giving illness
- Knowing our names
- No concerns
- Taken by KEMRI
- Taking mucus
- Transport and fares

1.3 Would participate

2. KGBC

2.1 Concerns
- Blood
- But child well
- Fares
- Gave child illness
- Help prick reactions
- Regret participation

2.2 Field worker
- Info on KGBC
- Info on SCD results
- Relationship
- Repeat visits
- Trying to remember

2.3 Getting results
- Reaction neg test
- Test before or after signs
- Waiting for results

2.4 Only dad

2.5 Reasons for participation
- Decided by another
- General health check
- It's a mystery
- Not aware
- Research
- SCD check
- Signs already

2.6 Tested in Kilifi

3. Religion
- Accepting God's will
- Explaining inexplicable
- Influence on TSB

4. SCD

4.1 Awareness
- Curable
- Following a relative
- How aware
- Names and syndromes
- Signs
- What affects

4.2 Cause
- As normal illness
- Due to witchcraft
- Folk-no explanation
- *Aware from ukoo (see inheritance)*

4.3 Effect on life
- Burden illness and changes over time
- Schooling
- Other life changes

4.4 Effect on relationships
- Child parent
- Parent
- Sibs friends
- Others
- Stigma

4.5 Feelings SCD
- Accepting/fatalism
- Distress child's condition
- Distress no cure
- Feeling bad generally
- Feeling better
- Other say like AIDS
- Results 1st reaction

(SCD cont'd)

4.6 Info from
- FW
- Hospital/clinic
- Others

4.7 TSB
- Being tested
- Diet and home
- Traditional
- KDH
  - Benefits SCC
  - Doors and benches
  - Getting treatment
- HW attitudes
- Private clinics
- Faith healing
- Comparing/combining
- Making decisions
- No treatment

Appendix B2: Extracts from Phase One Sample Analysis Chart family interviews (Analysis chart I KEMRI KGBC reasons for participation)

IDI	KEMRI aims	KEMRI general	KGBC reasons for participation/only dad
1	M believes that KEMRI provide proper examination and treatment for individuals, but also does research to find better drugs for the future. People would be prepared to accept whatever KEMRI requests (even LP) if (like M) they have had child with severe illness and KEMRI has helped, some don't want to cooperate with KEMRI but "run" there if later need medical care, does not agree with this attitude.	Power: if you refuse to listen/cooperate, may cause issues if you later need help from KEMRI. Relationship with KDH: SCD clinic seen as KEMRI only.	NA
4	KEMRI's work is research, meaning finding out how the illness started (in her child) and following course of illness over time.	Power: financial obligation – given money for fares so had to go; general powerlessness –"were chosen" Rel to KDH: talked of as different entities – adm to KDH but FU by KEMRI; down at KEMRI benches there is another hospital where every part of body is examined.	NA
6	Uses uchunguzi and utafiti interchangeably in talking about KEMRI's work. Was told research is to understand better about the disease, so each visit take blood for testing. Anything they do on the child I was ok.	Mbikes associated with taking blood – when asked if had heel prick, says that she hasn't been visited by those that come with mbikes.	Was recruited into KGBC from SCC after diagnostic testing, understood that research was being done at clinic = understanding more about SCD, not clear if understood as for benefit of others. Was asked if she would agree to participate and to sign form.
7	KEMRI research takes blood to find out whats wrong. F believes KEMRI are not providing good treatment for child with SCD, maybe treating wrong illness. M: utafiti in KGBC was to find out if illness is truly there or not.	Power: Expertise – accept test for illness even though child looks well as KEMRI are experts and have drugs; if refuse may later have issues with KEMRI; testing out expertise of staff – losing trust if cannot answer questions. Knows about early malaria vaccine trials on plantation (Bejon) Notes motorbikes for 2 diff KGBC FWs	Agreed to participate to check if child had ugonjwa wa mifupa, and F thought that would not be able to take child to KEMRI clinic later with this illness if they refused check now. Also believes that told that child would not be able to be adm to KEMRI hospital in the future if they refused to participate now. 1st visit, F did not want to agree without M as didn't have time to listen, 2nd visit met with both parents who did not agree on whether to

			participate – F agreed, M disagreed but F made final choice.
8	Utafiti (KGBC) was to find out if child had SCD, was explained voluntary.	Power: obligation for health? Agrees to test even though already knows positive. Some social element as knows FW? Rel with KDH: SCC run by KEMRI, doesn't know if KDH involved.	Agreed although already knew that child had SCD because thought that maybe KEMRI had not gone through files properly, and KGBC FW said test was to prove. Note also knew KGBC FW. Felt could not refuse (quote).
9	KEMRI are treating older sibs (as reason why agreed to cooperate with KEMRIs work), KEMRI seen as providing a service (through checking for illnesses e.g. KGBC). F wonders if drugs given at KEMRI are those being researched, feels not given enough information at SCC.	Told by FW that KEMRI does many diff types of research (diff diseases) Rel to KDH: seen as distinct – KEMRI and KDH have own ways of doing things. Talks of '*mzungu'* doctor who gave blood for child to be transfused (even own fathers blood did not match)	KEMRI are treating older sibs so agreed to participate KGBC. Child had already had some fever and parents were concerned about him. F also says FW told him would be able to tell from test if SCD from mothers or fathers side. M says she could not refuse this offer of help as knew that treatment saves lives, once child is born anyone has right to "see" him. ` F gave consent, no information given to M. M seems resentful "even if you don't know how to read, cant it b explained to you?" Understanding depends on intelligence not literacy. [Power dynamics in family: PGM stops F from talking by putting her hand out; PGM says wife must always agree to sex out of respect to husband).
10	M was reassured by fellow wives that KEMRI is good and important because will treat until a child becomes fine.	Power: obligation through health (what did you feel when told?) What could I have felt? I was told they were sick and I must take them to clinic.	Thought that test was for HYM and says was told that this illness makes children not able to walk, but if treated would be fine. Understood testing as an assistance to check for illness and start treatment early. M sought permission from F.
11	Not asked.	Power: inequity in expertise. Me to advise? It's for you not me!	Thought that blood was to test for HYM or malaria or any other disease. Older brother also known to have HYM but did not mention this as a reason for participation. In fact did not mention HYM as one of the illnesses being screened for until later in interview.

Appendix C1: Phase Two Information Sharing with Staff Groups: Fact Sheet

Sickle Cell Disease: Some Facts

What is Sickle Cell Disease (Homa ya mifupa)?

- Sickle cell disease is a condition that affects the blood and is always inherited from both parents. This means that people are born with it, just as they are born with other characteristics such as height, skin colour and being a boy or a girl.
- Although the disorder is inherited, the parents of a child with sickle cell may not have any signs of the disorder. The reason for this is explained in the section below on "How is sickle cell inherited?"
- Sickle cell affects the blood by making it more 'sticky' and less strong. The stickiness is due to some of the cells (small particles) in the blood having an abnormal shape. Instead of being round, some cells are shaped like a sickle (English word for *koropio,* or cutting tool), giving the disorder its name.

- **A person with sickle cell can attend school, college and work as normal. They should attend a clinic regularly for health checks and take life-long treatment that will help them to stay healthy.**

What are the signs of sickle cell?

- As blood circulates to all parts the body, the sickle-shaped red blood cells can get stuck anywhere in the body and cause signs, such as pain, in that part. Signs usually start after the age of four months and can include pain, frequent fevers due to infection, swelling of the stomach, anaemia (not enough blood) and jaundice. The problems of sickle cell are very variable.
- Most children go through good and bad patches. Some people are mildly affected throughout their lives and do not have pain often. Others have frequent and severe pain. Doctors cannot predict who will be severely affected.
- Unfortunately, severely affected children have a high chance of dying unless they get good long term medical care. With good medical care, people with sickle cell can often have normal productive adult lives.

How is sickle cell inherited?

Every child grows from materials (called genes) they inherit from both their parents at the time that they were conceived in the womb. Every new baby gets half of these materials from their mother and half from their father. As we see all around us, later they will grow up to show some characteristics that are like their mothers and some that are like their fathers. Of course, other things that happen in their lives will also affect the way that they grow up, such as any illnesses they have or the food they eat. Most people know many of the things that are inherited from parents, such as gender, height, skin colour or body build. The shape of blood cells is also one of the things that people inherit from their parents, including whether blood cells have a round or a sickle shape.

Sickle cell runs in families in two different forms:

1. One form is serious and very easy to see because it leads to the symptoms described above. This is the form that is called sickle cell.
2. The other form cannot be seen without a blood test because the person has no symptoms at all, but they can still pass the condition on to their children. This form with no symptoms is called being 'a carrier' for sickle cell.

The possibility of having a child with sickle cell depends on whether the parents have one of these forms of the condition. For example:

- *If both parents are carriers*, they could have children who have sickle cell. They could also have children who are carriers, like themselves, or children who have normal blood.
- *If one parent is a carrier and the other has normal blood*, they could have children who are carriers or who have normal blood.

Many people find it difficult to understand that sickle cell is in their family because they do not realize that some family members who appear to be unaffected must actually be **carriers**, without knowing. As for any inherited condition, we can say that sickle cell in the blood "**never gets lost**."

- **Both parents of a child with sickle cell must have some form of the condition in their blood, even if they do not realize this. In the same way, both family lines must be affected.**

How likely is it that parents who are carriers will have a child with sickle cell?

Just as it is not possible to know if a baby will be a boy or a girl before they are born, it is not possible to know in advance whether a baby of carrier parents will be affected. However it is possible to know *how likely* it is that a child will be affected by sickle cell if both their parents are carriers:

- There is a 1 in 4 chance that the child will be born with sickle cell
- There is the same chance (1 in 4) that the child will have normal blood cells
- It is twice as likely (1 in 2) that the child will be a carrier, like the parents

Some people wrongly think that a 1 in 4 chance is about what could happen for all their children thought about together, rather than for each child. They may think that if they have one child with sickle cell, the next 3 children are likely to be unaffected. But these are chances for each child and can't be thought of for the family as a whole. This is like the chances of having a boy or a girl during a particular pregnancy, which is always equally likely (1 in 2). But by chance, the children may turn out to be only boys or only girls in several pregnancies running. In fact, it is very uncommon for families to have equal numbers of boys and girls. In the same way, some parents who are carriers may have several children with sickle cell and other carrier parents may never have an affected child.

Appendix C2: Information sharing with KCR/mothers: Guide to information sharing session

Introduction

- We're conducting some research on condition called SCD, to find out peoples' views/advice on what information should be given to families about different aspects of the condition.
- This research involves talking to many different people in community, including affected families, chiefs and medical and research staff, as well as KCRs.
- Does anyone know someone with SCD? [Check if family member, also what they know about the disorder]
- Would like to book group for meeting on …….. to listen to views about this, and in preparation would like to spend some time talking about SCD today so that people have enough information to give an opinion.
- Check date, time and place for meeting

What is SCD?

- SCD is an inherited disorder that causes one type of blood cells to sometimes have curved shape instead of round
- Show picture of sickle and of normal/sickle blood cells (chembechembe – blood cells)
- Problem caused by sickle cells 1) being more likely to get stuck in blood vessels, cutting off blood supply to that part of body and 2) being more likely to break, so affected person tends to be short of blood (anaemic)

How important is SCD?

- The origins of SCD are linked to malaria because people with SCD are more resistant to malaria parasites, which need to enter blood cells at some point in a malaria infection.
- Therefore SCD is mainly a problem in countries where malaria is common, especially Africa, although now occurs all over the world through human migration
- In Kilifi, it is estimated that about 1 in every 100 children is affected by SCD at birth

What problems does SCD cause?

- SCD is very variable – some are more and some less severely affected
- With regular long term management at clinic, most people can lead full and normal lives – attend school, work normally and have families
- However, without special care, many young children with SCD will have serious health problems and may even die
- The symptoms start around 6 months of age, and depend on which part of the body is affected by the 'blocked' blood vessels
- In small children , this is usually the hands, feet or limbs, which become swollen and very painful for hours or days before settling down. These episodes of painful swellings will come and go over time.
- Children are often also pale, get tired easily, do not grow well, and have a swollen stomach due to a large spleen (hard swelling in stomach). There are other causes of a large spleen, such as malaria.
- People with SCD also more easily get infections than other people, such as pneumonia, skin and bone infections.
- SCD cannot be passed from one person to another, except from parents to their children.

How is SCD inherited?

- SCD is always (and only) inherited by children from both their parents
- There are many conditions that can be inherited. The way that different conditions are passed from parents to their children is different, depending on how strongly they are inherited. Strong conditions can be passed from only one parent. Less strong conditions must come from both parents. Because of this, the inheritance described for SCD here does not apply to other inherited conditions, or even to other inherited features that we all see around us (height, facial features, gender, skin colour etc)
- But all conditions that are inherited come from one or both parents through something called genes, which are very small bits of body material that are carried inside the cells of the body. These '*chembechembe ndogo'* are too small to be seen, except with very strong microscopes in the laboratory. They are also called 'genes'.
- In the case of SCD, it is not very strongly inherited so a child can only be affected if both their parents have passed on the SCD genes. The child will then have two SCD genes, and this will cause the symptoms of SCD.
- If they inherit one SCD gene from one parent, and one normal gene from the other parent, this will not be enough to affect their health, although they will be able to pass these genes on to their own children later.
- With cards, show way that sickle gene can affect people:
  - explain symbols for normal and sickle gene, and that these are found inside the blood cells seen earlier
  - demonstrate, by giving different people cards to hold and using question/answer approach, that: 2 SCD cards = sickle cell disorder; 1 SCD card and 1 normal = carrier; healthy but can pass on to children; 2 normal cards = not affected and cannot pass on to their children
  - explain carrier: important to realize that they will feel perfectly well and not have any problems throughout their lives from this one SCD gene, only issue is ability to pass on to next generation
- Demonstrate inheritance with card game, asking 2 participants to volunteer to be carrier 'parents' and tossing a coin to see which gene they will donate. Demonstrate ways in which inheritance can occur and role of chance
- Sum up options using cards illustrating 4 options for SCD inheritance where parents are carriers, linked to game above. Explain chances of options as:
  - 50/50 to have a child who is carrier, like parents (highest chance)
  - 50/50 to either have a child with no SCD genes at all, or a child with SCD (lower chance for each of these)
- Respond to questions

Appendix D1: Phase Two Question Guide/Scenarios for Staff Discussions

Scenario1: A two year old child is brought to hospital by his mother having had 2 episodes of very painful swelling of the legs and hands in the previous 6 months. He has a SC test which is found to be positive and is started on treatment.

Q: What should mother/parents be told about SC as a condition?

1. **What should they be told about genetic/inherited nature of SC? Why/why not**? What considerations?

SC is an inherited condition
Both parents must have passed on
Risk to future pregnancies

Should:

- Value information/knowing? Right to know? Duty to tell?
- Future reproductive planning for parents? *No more children, being prepared in future?*
- Awareness of risks for sibs?
- Wider PH value – especially when shortage of resources for health
- What blaming attached to alternative=traditional belief about curses/devils, use of TH?

Should not:

- A parent (mother?) gets blamed? *Who? How likely? How serious? What would make a difference? What situation father blamed?*
- Parents split up so can seek another partner to reduce risk? *How likely? What would make a difference?*
- Risk of finding out about non-paternity? *What consequence likely?*
- Consequences of conflict for all family? *Who mainly affected? Why? How?*

2. Who/when/how to give this information?

3. What if **mother does not want father to be informed** about genetic nature or future risks?

- What rights does father have? How weigh this up against mother's rights?
- How could <u>outcomes</u> influence this decision:
  - *If informed:* future reproductive planning possible (if not, may have more affected children)
  - *If not informed:* avoid family breakdown (future reproductive risks, non paternity)

4. How should we respond to **requests for testing** from parent(s)? *Insist that both parents must be carriers? Agree to test? Why or why not? What would make a difference?*

- What rights do parents have? Duties of staff?
- What influence of risk of uncovering non paternity? Other consequences?

5. What **responsibilities do medical and KEMRI staff have** in counseling about genetic nature SC? How would this affect their response to issues above?
6. Is this **different to non inherited illnesses** affecting children?
7. How can we **lessen** any such risks? Does explaining 'both must have passed on' lessen risk of blaming? (1 in 4?)

- **What can we conclude about information that should be given on genetic nature of SC, how, to whom and when?**
- **What would make a difference to this?** *[summarize from earlier discussion/explore again]*

Scenario 2: A research project is trying to find out more about SC, including how common it is and how it affects children's lives. All families in the area are visited and asked to participate by having any child in their first year of life tested for SC. If a child is found to be positive, they are referred to clinic for long term care.

Q1: What should be done about carrier results, why?

- *Disclose:* Child's future reproductive decisions; parents may benefit from knowing that SCD in family; public health gain from wider awareness

Disclose AS routine/requested/deferred/not
Encourage later AS testing
Effect (non)availability 'national programme'

- *Don't disclose:* Not an illness but may be stigmatized/treated differently; may not remember results later; child has not agreed to be tested; could be given result/tested later in life when old enough to consider *(describe church screening programmes in Ghana)*
- If to defer testing/disclosure: **WHEN ideally**? Before marriage? Adolescents? (national screening?)
- **How weigh child's and parents' interests re. testing infants?**

Q3: How is SC generally seen within the community? How much/what way do families with affected members experience stigmatization by the wider community?

- What age are children/teenagers/adults likely to experience any stigmatization? How? What due to?
- Does it affect boys and girls similarly or differently, why/how?
- [Any reincarnation stories?]

- **OVERALL what can we conclude about:**
  - **Who should give consent for this type of research?**
  - **Should AS results be taken back in research testing healthy children?**
  - **What are wider community members attitude towards families with SC, what implications for research?**
  - **What main focus for SC control programmes? (NBS, testing parents, B4 marriage, schoolchildren)**

For all discussion points above, explore **opinions/ideas** and **reasons** for them, with specific reference to:

a) Which peoples' interests are being considered; and how are the interests of different people being weighed up (whose interests count most, and why?)
b) What outcomes are being considered as important, which types are these (health, social, other), are they short or longer term and how are they being weighed up?
c) Are there any fundamental values being expressed ie that don't seem to have a 'reason why'?
d) How are the interests of individuals weighed up against outcomes?

Appendix D2: Phase Two Question Guide/Scenarios Chief/KCR/affected mothers

Scenario1: A two year old child is brought to hospital by his mother having had 2 episodes of very painful swelling of the legs and hands in the previous 6 months. He has a SC test which is found to be positive and is started on treatment.

1. Should we give any information about inheritance at all? What should we say and why?
Options: From family(ies)/parent(s), one/both sides, don't mention inherited

- *Should say inherited:* more likely to come to clinic, less likely to use alternatives, they should know, we should explain, part of increasing awareness/ reducing stigma in wider community since will pass information on to others
- *Should not say inherited:* will not understand, risk of blaming
- *If reasons not to talk about inheritance, why not just explain serious life long illness ('natural')?*

2. If blaming is a risk, how does this happen?

- *Who usually gets blamed and why? (mothers, either parent, family)(observations, 'cultural')*
- *What does this depend on? (age, education, religion, personality)*
- *What for? (Causing thru inheritance OR causing thru unfaithfulness)*

3. **What effect of blaming mother for passing on to children? (where father denies his role)**

- *What does this depend on? How much risk of breakup as a result?*

4. **What effect of blaming mother for unfaithfulness? (*accept, deny, breakup, ?violence)***

- *What does this depend on?*
- *How much risk of breakup as a result?*
- *How much of a risk that we will find non paternity?*

5. If father did not accept he is carrier and asked for test, what should we do?

Refuse to test:

- No need as can tell both parents they are carriers without testing
- should not take risk of showing he is not carrier (therefore not father) as serious consequences for mother and child

Agree to test:

- likely he is actually a carrier and would help to convince him
- only a minority would show non paternity
- father has right to be tested/know result
- if he asks, this is our job as health professionals
- if we refuse will affect our relationship with him, for refusing
- if carrier we can avoid telling him (polite lie)
- if not a carrier we can tell him (polite truth) since family social issues not our responsibility
- If we refuse, he can go elsewhere anyway (may be less supportive)

  - ***Summarizing: If we test father and result shows he is not a carrier, what should we say/ why?***

6. **What if you were father?** *If reasoning different than for 'others', why different?*
7. **What difference if we explain from parents *specifically* (see diagrams) or from both families more *generally*?** *(to blaming, to anything else?)*

Scenario 2: A research project is trying to find out more about SC, including how common it is and how it affects children's lives. All families in the area are visited and asked to participate by having any child in their first year of life tested for SC. If a child is found to be positive, they are referred to clinic for long term care.

---

1. **Should we give any information at all? What and why?**

Explain carrier:

- Can help child choose partner later in life to avoid risk SCD in own children
- Can help child be more prepared if has child with SCD later in life
- Child should know (for sake of knowing?)
- We should explain (our responsibility?)
- Will help more information getting to wider community, create better awareness, more acceptance
- Not fair to say 'not affected' as may later get more information and *discount* as assume negative (i.e. *misinformation*, not just not *withholding* information)
- [Since (at least) one of parents is also a carrier and well, will find it easy to accept]
- [Whole family will understand they have this risk]

Don't explain carrier:

- May not be able to test partner later in life, so no point
- Will cause unnecessary worry, e.g. relate all illnesses in future to this
- Risk stigmatizing child, especially girls at age of marriage
- [Will use too many resources]
- [Will not understand anyway]
- [Will not remember later in life to help in planning own future]

2. **Would you want to know if your child was found to be a carrier?** *If different, why?*
3. **What difference would it make to this reasoning if there was a public information campaign on SCD and counseling/testing was available to anyone?** *Why this difference?*
4. **What views about when is best time to find out that someone is a carrier? Why/why not?**

- *After birth/before marriage/at school? (why?)*

Overall considerations for ways people are thinking/deciding between 'reasons': *What importance is given to following, why are these thought most important? (what being compared with, how?)*

- Interests of individuals? which individuals are thought most important?
- Real life effects of decisions? which effects most important/who affected? long/short term effects?
- Basic values of truth telling/knowing truth for its own sake (regardless of consequences?)
- Our responsibilities (as health research or care providers)

Appendix E1: Phase Two Analysis charts KCR 01 (extract)

1. Giving information on the role of parents, including testing

IDI	*For/against inheritance information*	*About testing of parents*	*Weighing up*
P1 (F, 59y, 7y edu, C)	Follow and agrees with points made by P2 about why inheritance from parents should be explained, plus: - Parents currently not aware - may have other affected children/family members now or in past without realizing, able to help those children, able to reassure family about deaths occurring in past -important not to conceal information, compares with positive changes in attitudes to HIV once became more open (p4) - prays that condition will become more widely understood so that people will stop taking child to TH rather than SCC, leading to deaths (p5) **BUT:** May cause a lot of concern so need to explain in way to reduce this/reassure (p5) After P3 mentions risk of parents blaming each other or mother being accused of unfaithfulness, P1 agrees that could happen, particularly in families where parents already do not trust each other (p9). Can lead to separation/divorce.	Countering P4's idea about mothers being generally more likely to be blamed for 'bringing in' inherited illnesses, as opposed to fathers 'taking them out', P1 points out that fathers spend more time at home than out, so would still be as likely to be responsible for inherited condition in family as mothers. She gives a local saying that 'only the mother can know for sure who are the parents of the child' (*mwana ni mameye*) Does not directly say if would offer testing to parents, but comments that giving information on inheritance may lead to them asking for testing, and does not frame this as a problem, more as a possible consequence (p31) If father asks for testing – sees this as their decision. Compares with situation for discordant couples for HIV testing, where it's only advice that can be given (p33) Agrees with colleagues who say should be open.	Strongly concludes that parents should be fully informed about inheritance and their role in it because alternative is that will not understand and child will continue suffering (TH?) Also important to know truth (p30) Good explanations and support can prevent divorce for people 'who have wisdom' and love each other. Emphasis on truth telling –' but the truth should be told, the truth, because if its not told, it will be, it's not good' (p31)

P2 (F, 32y, 16y edu, M)	Explain condition, not disease, and carried through family line across generations, so that (p4): - 'will take it in a good way' (p4) - understand why parents are carriers and yet are healthy. - do not think due to parent's mistakes (?STI) but from their own parents/families. - be better prepared for the future - advise children (other children?) not to marry someone with same problem to avoid future generations being affected. **BUT** Following discussion of separations/ divorce due to blaming (by P1), adds that may be many who choose to separate/divorce to avoid having any more affected children, especially if affects 1st child (p11). Describes this as stigmatization (in English) (p10) Could also lead to denial because of these implications for future (p10)	Says that 'mistrust' of mothers by husbands is not usual, but that where father feels he is well (or has been tested and shown normal) and condition said to be inherited, mother likely to be blamed. Sees allowing parents to be tested for SCD generates problems in potentially showing MP (p23), suggests that this is a significant problem 'those cases are there' *If father asks for testing*: 'let him be tested, you have done your part' (p32) 'The results that come out belong to them' [*rights of father/responsibilities of Dr*] 'It's not now your problem' 'Dr's part is to educate, that's been done, the results are for him and his family, they will decide on their own the way they will live' (p33) Later adds that before testing, father should be counselled and told of consequences but if he insists, then will have no alternative but to test him (p33) When M explains other people have suggested 'hiding the truth' about father's results, she points out that might cause complications in future e.g. if has child with another woman and that child has SCD [but is mixed up here as would be told AS when actually AA, so there would be no chance of having affected child with this wife or another woman] Concludes that although can bring difficulties, right thing is to tell father truth, not good to encourage mother to go outside, maybe bringing more sick children into the home. 'You will have done your duty' (p37) 'maybe they will love each other so much they may not divorce but now those things are their things now, yours, you have done'	Finally strongly concludes important to explain inheritance is from both parents – will not necessarily decide to separate to avoid risks in future, may also decide not have more child, some fathers might decide to take 2nd wife. Explanations are key for next generations of that family (p28) as well as for the child's care, can also help to reduce some forms of blaming. Would not offer/persuade parents to be tested because of issues of MP but if father insists, would test and give true results with counseling before on possible outcomes. Main reasons are seeing this as family not medical issue, rights of father, not encouraging unfaithfulness and risk of greater complications in future.

Appendix E2: Phase Two Summary analysis chart KCR01 (extract)

Q1: What information should be given about inheritance of SCD and what implications of this information for family/child?

1. Overview

- There was spontaneous agreement within group (no evidence of any important forms of influence between participants or from moderators) that important to give *information* about inheritance of SCD, including that this is *from both parents and family lines*. The main reasons were:
    - Benefits to child/family in future: addressing current common and harmful misconceptions that influence health care and family relations, benefitting child directly through assumed better uptake health care, including in long term, and possibility of reducing risks to family in future (reproductive choice)
    - Parents' rights to information: decisions about child and marriage are family responsibilities, not medical. Parents have right to information as ones who look after child; and have an obligation of care to child.
    - Public health benefits: greater awareness of SCD and management; supporting relationships of trust between doctors or researchers and clients/participants
    - Openness/truth telling: as a value, to support trust between doctors/patients, to support trust between parents (discourage unfaithfulness)
- The main reservations for giving information concerned risks of conflict within family as a result of parents blaming each other, especially mothers; generating suspicions of MP; and the responsibilities of doctors/researchers to prevent this. But this were not considered sufficiently likely/important to undermine reasons for giving information on inheritance. One participant felt that in some situations, might be better to talk about 'family lines' than about roles of 'individual parents' to reduce risks of parents blaming each other.
- P4,5 and 6 (at least) took time to understand MP issue, at first being certain that better explanations could address mistrust/suspicions of unfaithfulness.
- Once understood, most (5 of 6) participants wanted <u>to limit information</u> to exclude any discussion of misattributed paternity (MP) as a mechanism in SCD inheritance i.e. this seen as most serious risk. However, if parents asked questions about MP or testing was requested by father, 5 participants would be open with information, testing and giving results [*clear difference in obligations between actively giving information and passively responding to requests for information*]. P6 was less clear, with main concern being stability of family, where he would try to avoid testing/talking about MP but not refuse to test or conceal results. One participant (P5) was more inclined to proactively recommend parental testing as well as giving results; this was largely based on value of openness as well as feeling that this would reduce risks of blaming (i.e. higher risk is of father wrongly blaming mother for bringing into family than for testing showing MP).
- One participant (p3) was very quiet in discussions, although also was the person who raised issue of parental blaming/conflict as possible consequence of understanding inheritance.

2. Reasons for explaining inheritance SCD

2.1 Addressing harmful misconceptions/not knowing

- Current common belief that SCD/unknown diseases are due to bewitchment. Affected children taken to TH and do not get better. If understand normal condition that's inherited, more likely to take to hospital and child will benefit.
- Parents may have other children/family members currently affected without realizing, so they could benefit from better understanding as well as index child. Similarly if have lost children in past, can reassure them that these were 'natural' deaths not due to witchcraft
- Parents fear/worry about unknown diseases, and better understanding can reduce this. These kinds of fears can even lead parents to separate e.g. blaming each other, fears about HIV
- Parents may interpret as mistake by one of them (including STI, unfaithfulness, curses), so explaining as a condition inherited from both sides across many generations important, also in helping to see that they could pass on without any signs.

2.2 Supporting access to effective treatment (biomedical) for child/other family members affected

- Understanding basis of condition will encourage to bring child to clinic for management, also in long term when understand chronic, intermittent nature
- Many of above misconceptions also lead to low uptake of medical services
- Information will be passed on to others in community gradually, so others may also benefit in same way

2.3 For the future

- Will be better prepared for the future, any effects of this disorder within family
- Can advise the child to have a future partner tested to reduce risk of future generations being affected

2.4 Openness, truthfulness

- Most parents currently not aware
- Important not to conceal information, compare with change in attitude to HIV with increasing awareness/openness, hiding truth can lead to later complications
- Should explain as parents are living with this problem – child's pain is known to them

2.5 Obligations/duties

- Parents have obligation to their child so must know best way to fulfill this (P4)

2.6 Public health benefits

- Information will spread across community to help other families affected, and offer potential for families to put pressure on government to provide services
- Builds relationship of trust between doctors/researchers and clients/participants

3. Reasons for not/reservations about explaining inheritance

Generating new issues:

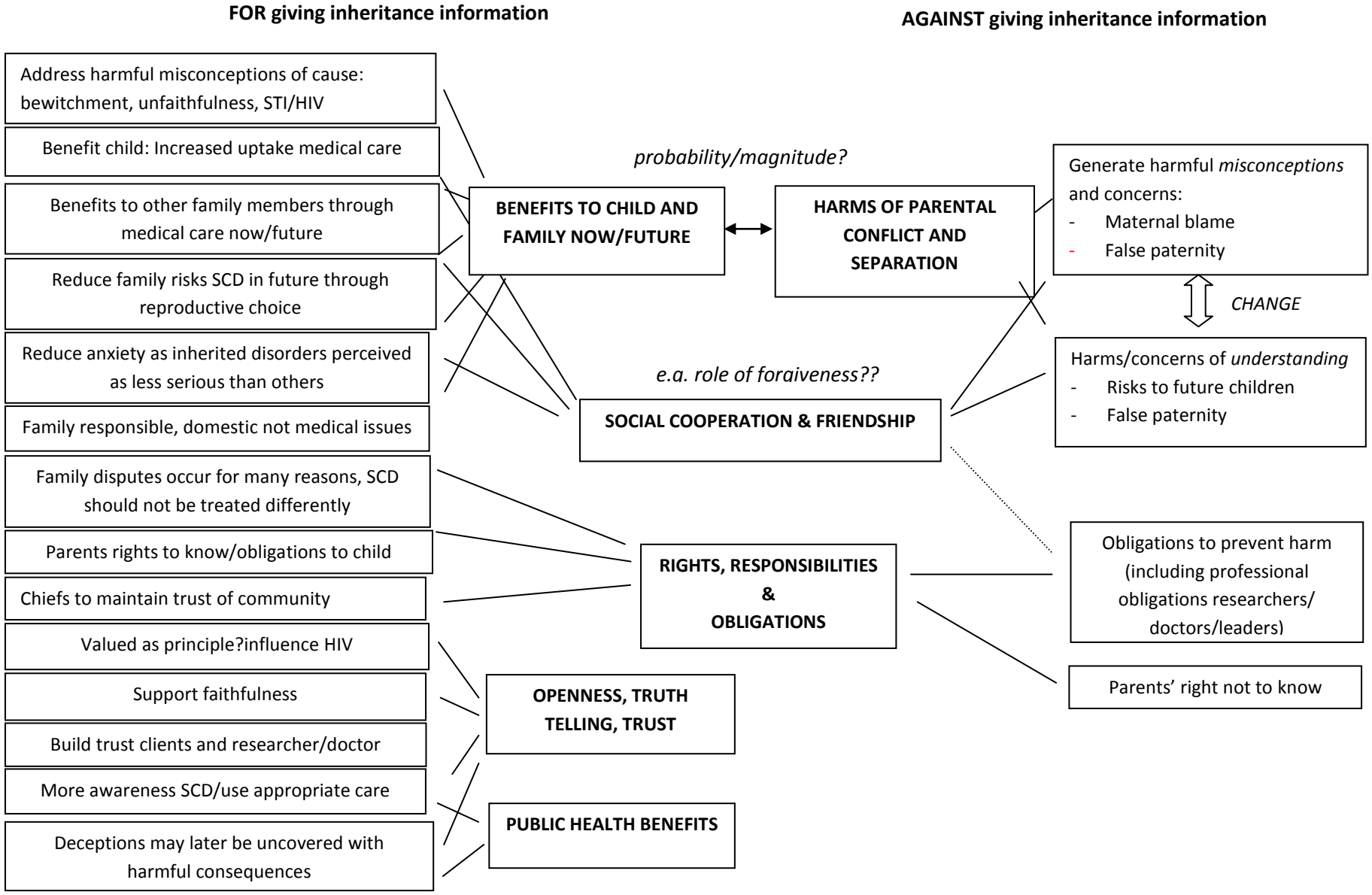
- Since currently unknown, may cause worry about what this means and implications of inherited nature for child and parents e.g. understanding that future children could also be affected – pressure on parents relationships (P2 says especially if 1st child affected); parents worry about own health
- Link with inheritance can raise risks of blaming: towards either parent for having brought into family OR for unfaithfulness (towards mother only). P1,2 think this likely if there is already problem with trust between parents. P1 thinks could lead to separation.
- These concerns may lead to denial of inherited nature
- Father may ask for test to show whether he is a carrier or not, this would have potential for showing MP in some cases and generating parental conflict, and possible harm for child also

4. Summary weighing up pros and cons: conclusions by participant

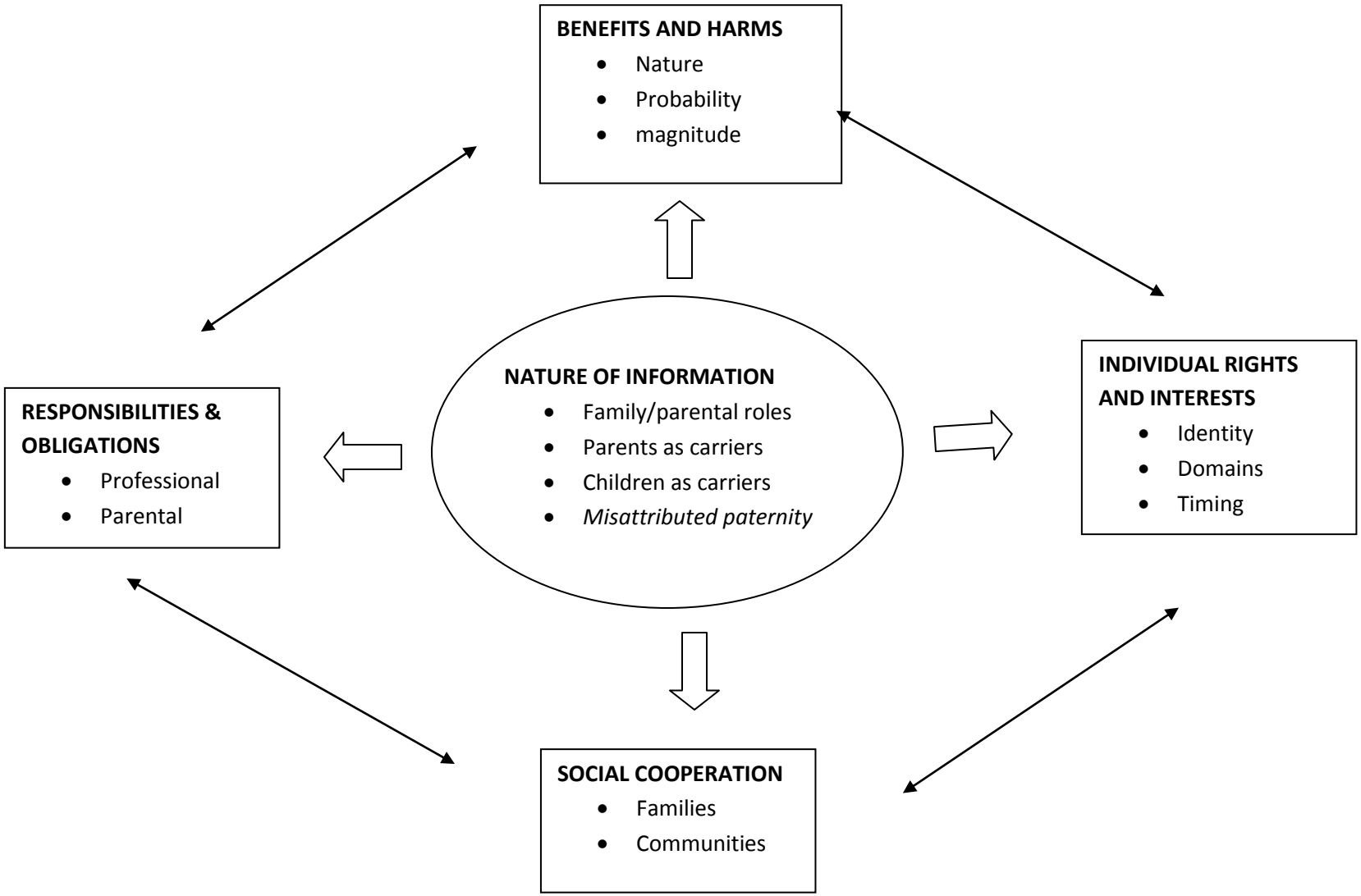
ID	***Reasons for explaining inheritance***	***Weighed up against/conclusion***
P1	• the interests of the child, since best way of ensuring gets effective care (*main concern*), as well as any other children who might be affected in that family • need for openness and telling truth as a fundamental value – also makes comparison with public health benefits for HIV/AIDS in doing this (*seems to be both for its own sake and for effects of openness*) **AND** (about factors against) • separation not inevitable – those who love each other and have wisdom may not separate • For testing father: not completely seen as responsibility of dr. to protect parents in this way (its only advice they can give)	• risk of causing parental worries • agrees knowledge about inheritance may lead to parental conflict/separation (for causing and MP) • acknowledges traditional beliefs and sayings that point to MP as a locally important issue (*e.g. mwana ni mameye*) **Therefore** • Strongly believes should give parents information about SCD inheritance • Would not actively test parents/give information on role of MP but would test and give negative results to fathers if testing requested
P2	• benefits to this child and all succeeding generations in understanding what the condition is, including not marrying someone else with same condition in family • reducing blaming in family as will realize this is a condition that has been passed on from both sides of family, from many generation, so not a mistake made by parents, also why parents can pass on without having any illness themselves. • For testing father: dr's main responsibility is to advise and explain, not to try and protect families from parental conflict – those are the parents own decisions – you will have done your duty	• the risk of more blaming, particularly of mothers for causing SCD or MP where father denies being a carrier, or separating to avoid future risks (especially where 1st child affected). • Risk that father will request testing because he denies responsibility, and that this might show MP and lead to separation • (not commented on by her, but discussed in group) the adverse effects on the child if parents separate)

	• if there is an real issue of MP, important not to encourage mother, as may continue to bring sick children into the home • For MP: not inevitable that parents will divorce, depends on couple (e.g. if they love each other so much) • importance of truth telling as can often lead to worse complications to hide truth **AND** (about factors against): • Conflict between parents (as a result of information or requests for testing from father) only likely to happen where there is already mistrust; not so usual in families • some families may decide either not to have any more children or to take 2nd wife rather than separate (Muslim?) For testing father: • can ameliorate risks by making sure father has good pre test counseling so he understands what consequences could be • not Dr's responsibility to protect, family responsibility (strongly believes)	**Therefore** • Strongly concludes important to explain SCD inheritance to both parents, even if this risks separation • Would not actively test parents /give information on role of MP/suggest testing parents but if requested would test and give father negative results, again even if this could cause conflict in family

Appendix E3: Phase Two Exploring themes (early)



Appendix E4: Phase Two Exploring themes (later)



Appendix E5: Table 6.2 Summary chart of agreement and difference by group type for responses to requests for paternal testing

Community representative and 4 affected mothers

KCR 1: 6 local residents from an urban setting. Higher overall level of education that other community groups, with the exception of one man with 3 yrs of schooling. Three had full secondary education, one of whom was also a graduate (Muslim woman). The KCR chairperson, an older Christian man was included. However all bar one were very outspoken and contributed actively.

Overall quite strong consensus (only one person disagreed) that should not proactively offer parental SCD testing. By end discussion, five agreed that if father asked to be tested, doctors should agree and give the results even if showing MP. One person remained ambivalent.

Key views:

- Family breakup a possible outcome of sharing information on misaligned paternity, but perceptions of outcomes of disclosure do not seem to be as severe in this group, and no mention of physical harm.
- Misaligned paternity is a domestic not a medical issue, and therefore not a responsibility of researchers to try to protect families
- The possibility of forgiveness, based on religious faith
- Common reference to HIV/AIDS control e.g. the importance of openness as experienced in that programme
- The importance of openness in counteracting rapid increase in SCD (seen as 'epidemic)
- Not wanting to encourage unfaithfulness that might bring other diseases like HIV into family.
- Possibility that separation might be a positive outcome, with less emphasis on family stability (minority view)

Participants were divided in their reasoning, so that although conclusions largely agreed (importance of disclosing results), some based this on liberal views about openness in spite of potential for harm, while others tended to support a more paternalistic stance but drew on religious beliefs to conclude that forgiveness was possible. The participant who was opposed to disclosure at the start (chairperson, 50 y Christian male) remained uncertain because of potential for harm but eventually came to agree with others that he, personally, would be able to forgive his wife by drawing on his faith and mechanisms with his church to support confession/forgiveness. On this basis, his general position became less certain.

KCR 2: 6 local residents from a rural area with relatively high prevalence of SCD in comparison to others. The KCR chairperson and secretary were included; the latter being the mother of an affected child. This was an active conversation involving all participants. Four participants were Christian, one Muslim and one following a traditional faith. Three participants had attended secondary school, and three had full or nearly full primary education.

There was disagreement during this discussion on whether or not researchers should offer or agree to test fathers or provide misaligned paternity results.

Key views:

- Greater threats associated with supernatural explanations and paternal denial than sharing misaligned paternity, since latter not common
- Importance of openness based on an analogy with HIV/AIDS, and the acceptance of unfaithfulness this has brought
- Acceptability of concealing/lying to protect the person lied to
- Misaligned paternity as a domestic (in fact mothers) and not a medical responsibility
- Doctors role 'not to harm'
- Possibility of forgiveness drawing on organized or traditional faith values (all agree)

Most participants began with some reservations on testing and sharing information on MP. One woman changed her position in favour of offering testing including disclosure, based on a comparison with HIV/AIDS policy. She was supported by the other women (one of whom had a child with SCD) and one man in arguing for openness, but with more reservations about risks of harm. She was quite strongly opposed by one man, who felt throughout most of the discussion that researcher should not give this information because of risks to family harmony, but towards the end modified his attitude to think that forgiveness could be possible based on Christian beliefs, in

general agreement with others on this role of faith. The discussion was often polarized between these two individuals, with many references to gender (raised by issue of unfaithfulness in women) and to the role of religion in forgiveness.

KCR 3: 6 local resident from a rural area. This discussion was dominated by three older participants. The group included three women and three men, all Christians except for one man. One person (and older man) had secondary education, and the remainder at least 7 years of primary schooling. The KCR chairman and secretary as well as their 'vices' were included.

By the end of this discussion, there was general agreement that doctors should not offer parental SCD testing and should not disclose MP results, but should test fathers if this is requested.
Key views:

- Prominence of jealousy in marriage in this community, little chance of forgiveness where misaligned paternity known and high chance of serious harms to mother and child, including severe physical violence (all agree)
- Better to test father if asked (not clear why)
- Not good to lie but even worse to harm; different types of lies
- Fathers' rights not to know, to maintain family harmony (his interest as well as others)
- Doctors' role not to harm, and lies to protect/help are already accepted medical practice
- Openness and benefits seen in HIV/AIDS progammes (one younger person suggested briefly)

Over the course of the discussion, a few members expressed different views about whether it might be possible to disclose misaligned paternity results, such as the benefits of openness seen in HIV/AIDS programmes. However alternative positions were eventually either abandoned or not voiced. There was also consensus that fathers should be tested if requested, leading to a justification of 'good lies' as a way of handling results.

Mother's group: Three mothers of children with SCD; two have two and one has one affected child. All were participants in phase 1. One mother has secondary education and the others 6 years of primary schooling. All are Christians. Active conversation although with one mother talking more than others.

There was a difference in opinion about testing and disclosing misaligned paternity results, with one participant (primary education) moving quickly from an initial doubt to feeling strongly that it's important to test fathers if requested, and then also important to be open about results. The other two participants were more ambivalent, with concerns about the potential for harms, but one tended to agree that should be open as far as possible.
Key views:

- Misaligned paternity results will lead to separation or child not being treated as well as others
- Most mothers are faithful so testing fathers will benefit mothers and families in the majority of cases (all agree)
- Risk that father may have another test later and find out that was given incorrect information, losing trust in doctor and bringing risk of family breakup anyway
- Hiding results will encourage mothers to be unfaithful (all agree); doctors should not be complicit in deception

Given their assessment that most wives are faithful, all mothers believed that testing fathers was an important way to convince fathers of their role, but two had concerns (one more than the other) that the risks to the mother and child might outweigh these advantages for those where misaligned paternity was a risk . The conversation was dominated by one participant who explored a range of ideas very quickly, working towards a strong argument for openness in the end. However the others, while quieter, were not completely swayed from their concerns about harm.

Community leaders

Chiefs 01: 6 assistant chiefs from relatively urban areas outside central town, all secondary school educated, one Muslim and remainder Christian, two women and four men. Ages between 38 and 56 years. Active participation, interview started in English and later mixed English/ Kiswahili; responses to Kiswahili much more rapid. All participated actively.

After considerable debate, there was agreement on the undesirability of generating information about misaligned paternity. For this reason, two would refuse to test fathers while the others (4) would agree to test

but give a false interpretation of a negative carrier result.

Key views:

- Misaligned paternity is important risk to family stability
- Main issue is welfare of child and stability of family, others concerns are secondary to this.
- Possible to refuse to test father
- Acceptability of giving inaccurate interpretation of inheritance (e.g. either from father or wider family/changes over time) to account for negative results
- Understanding healthy carrier status well will reduce motivation for paternal testing
- SCD as a new disease for which community should understand possibility that control will demonstrate (and therefore discourage) unfaithfulness
- Father's right not to be cheated

Participants explored many options for handling requests for testing during this discussions, balancing up perceived undesirability of showing misaligned paternity (high costs to family) and that of concealing information. Many individuals changed position several times over the course of the discussion.

Chiefs 02: 6 assistant chiefs from relatively rural areas, including higher SCD prevalence areas. All secondary school educated, Christian men between ages 41 and 53 years of age . Active participation, interview started in English and later mixed English/ Kiswahili; as above responses to Kiswahili much more rapid. Active participation although one chief was absent for most of discussion.

Disagreement within the group on testing and disclosure of misaligned paternity results. Four would conceal results, one would disclose and one would prefer to disclose but felt this would often not be possible.

Key views:

- High risks of conflict and separation in family where misaligned paternity shown
- Priority is family harmony
- Deception for a good cause is acceptable; may be part of chiefs' roles in protecting community; doctors sometimes deceive patients in their own interest
- Truthfulness important as may later be found out, generating worse harms that if had been truthful
- Truth telling as a Christian principle
- The way that information is shared is very key, over time can move from deception to truth. Need to base this on individual assessments including education and marital stability

Initially all participants thought that it would be too potentially harmful to families to disclose misaligned paternity results, but during the discussion two participants changed views to supporting greater openness. In part this seemed to be a response to being asked what they themselves would want, but mainly to one chief linking this situation to a story he had heard in which a deception involving an HIV test result was later uncovered through a second test.

Chiefs 03: 6 chiefs from urban (within main town) and rural areas, including higher SCD prevalence areas. Only one not secondary school educated, he has primary education. All were Christian men aged between 48 and 55 years. Active participation, interview started in English and later mixed English/ Kiswahili; as before responses to Kiswahili much more rapid. One chief is openly HIV positive and drew on these experiences in this discussion. Able to draw more on experience of talking to communities than interacting with individuals within the community.

There was general but not very clear agreement within the group on how to handle requests for parental testing and disclosure of misaligned paternity results; the majority (4/6) prioritised openness as much as possible, with a final decision based on individual counselling to assess likely reactions. The remaining two did not see this type of information giving as their responsibility and were not able to make a recommendation.

Key views:

- Priority is family harmony
- Most wives are faithful
- Openness/truth about role of unfaithfulness in SCD important for people to know to promote faithfulness and

improve health of community. Example of Kadhi courts requiring HIV testing before marriage

- Some men, particularly older ones, can accept a child from 'outside' marriage (four agree this is possible e.g. dowry paid); children are an asset to the home
- Importance of counselling before testing to ensure fathers can accept any result, drawing on model of VCT for HIV/AIDS
- Chiefs/assistant chiefs and village elders can support in family conflict (minimise harms)
- Father's right to know includes being able to avoid inadvertent harm to others (one person, from experience of HIV)

At start of this discussion most participants expressed reservations about disclosing misaligned paternity, although supporting wide public awareness campaigns on SCD generally. But over time several changed their views to support more openness. In part this was by association with VCT in HIV/AIDS policy, in that misaligned paternity results could be shared after pre test counselling to ensure/assess whether (social) father would accept any results. Another change in opinion emerged during the refreshment break when participants chatted about the cultural acceptability (particularly in more traditional rural areas) of accepting a child born out of wedlock into the (social) paternal home, linked to dowry payments having been made and a general positive attitude towards children. One participant remained doubtful about disclosure throughout.

District health managers

MOH: Participants were all members of the District Health Management Team, including the District Medical Officer, District Public Health Nurses in charge of Public Health and Medical Services and the District Disease Surveillance Officer. One DPHN was a woman, the others were men. Two officers were from Kilifi district originally. All were Christians and between 37 and 48 years. All interviews were conducted individually, with active engagement.

There was agreement on the general desirability of openness but differences in final conclusions about whether or not to test parents and disclose misaligned paternity results.

Key views

- Awareness of ethical dilemma posed by testing parents; fathers rights to information (accepted principle, from HIV/AIDS control) and threats to family stability/interests of mother/child. "Ethics vs. facts" (or ideals vs. pragmatics)
- Fathers and family interests aligned
- Critical role of counselling: pre/post test to ensure that disclose to fathers who can accept (cf. HIV/AIDS policy) and that can work with others over time to disclose results in best way possible
- Doctors responsibility to take social and ethical issues into account alongside medical issues
- Health managers responsibility to consider long term health interests of community
- Health professionals responsibility to look after interests of sick child/family more than rights of father (whose health cannot be affected)
- Limitations imposed by lack of SCD control programme in district, and strongly positive attitude towards strengthening
- Positive attitude towards 'information campaigns' and role of better understanding in addressing harms
- Acceptability of giving false interpretations of negative carrier results depending on predicted outcome
- Policy will get easier to implement over time (like HIV/AIDS)

These issues were explored as part of articulating the need to balance the values associated with openness and rights to knowledge with the interests of sick children and families. Participants all drew on current HIV/AIDS policy on counselling and testing, tending to support openness as a policy as far as possible. However, the recognised challenges to some families underpinned final differences in opinion about the best option at present.

Research staff

20 participants, interviewed individually (5 participants), in pairs (2 pairs) or groups of 3 to 4 (3 groups). Staff groups were mainly field workers (12) or community facilitators (5) but also included 2 data entry clerks and 1 early career researcher. Most were aged between 27and 43 years, with one 51 year old. 3 were Muslims and the remainder Christian. All were originally from the district and had at least secondary education, 2 were graduates

and 6 held diplomas. One participant had 2 affected children and had lost 2 children to SCD. One was a SCD carrier with a carrier son, and had lost her older brother to SCD. One participant's sister had 2 affected children. These were generally very open and active conversations, held in English and facilitated by the interviewer's existing working relationships with participants. The first 7 interviews were held in 2 parts one week apart to allow more time and create an opportunity for participants to test out ideas with others.

Across staff groups, opinions were diverse and sometimes very strongly held, but the most common view was in support of testing where requested, and openness about the results, although often with the proviso that the couple had been counselled and the mother had consented (to the father's test). However 4 individuals disagreed with testing and disclosure (2 in IDIs and 2 included in groups of 4) and 2 were unsure (1 IDI and 1 from group of 4). Of the 14 who supported openness, 2 were in IDIs (including the early career researcher and staff member who is a carrier) and the others discussed the issues in groups. Staff groups were all clearly talking about a research situation.

Key views:

- Importance of openness in health policy for individuals and communities, both as an intrinsic and instrumental value
- Father's rights to know
- Likelihood of knowledge about misaligned paternity leading to family breakup, including influence of traditional ways of handling conflict, underlying relationships
- Special interests of child/mother/family (aligned)
- Responsibilities of doctors/researchers concern health not social issues (argued as reason for and against disclosure)
- Use of counselling to assess reactions and support acceptance; including mother's consent to testing in father
- Commonness of unfaithfulness and misaligned paternity
- Knowledge of father's negative status means family can plan for future children without worry
- Breakup of family may be an opportunity (one person)

Reasoning on the importance of openness drew on interpretations of intrinsic and instrumental values. Where these values were prioritised, participants often drew on comparisons with HIV/AIDS and research consent processes. Values were placed on openness and rights more commonly than paternalism. However these conclusions assumed inclusion of counselling processes and usually maternal consent. For many participants, views emerged through debate over time. The most marked shifts in attitudes were seen in the groups involved in 2 separate discussions, where a movement from being more to less paternalistic was the most common pattern. However at least 2 participants made this move in the other direction, starting with strong views about openness but concluding that researchers cannot risk the welfare of sick children and vulnerable mothers. Researchers should therefore not test or should conceal controversial results to protect the family.

Appendix F: Figure 8.2 Assessing researchers responsibilities for sharing SCD information during studies: Questions about context and their implications *(* denotes information generated by this study)*

QUESTIONS	*IMPLICATIONS FOR RESEARCH PLANNING*
1. BENEFITS AND HARMS	
From the perspectives of researchers, participants, the wider community and other health stakeholders: • What *understandings* and implications of SCD for the child, family and wider community? Are there any particularly sensitive areas of SCD information sharing? e.g. Beliefs, gender, HIV/AIDS? • How much would information sharing help affected children and families, including limiting *existing harms*? • If researchers do not share information/provide support, what would families be able to do? (dependence) • What challenges exist in achieving benefits of information sharing, and how can they be overcome? • How is *diversity* within community likely to influence harm/benefit assessment and dependency, including for SCD and carrier status (such as ability to access private testing/care)?	• For disclosure SCD status to act as a benefit during research, information sharing should be seen as a form of *ancillary service*, including ensuring access to validation of tests, family counselling and (for SCD) health care. • Stronger responsibilities for researchers to share information are generated where there are higher levels of potential benefit from this action, and of participants' dependency on researchers to achieve these benefits. • Planning for information sharing & care should be based on local assessments of how SCD is understood & managed; and take account of diversity within the community. • *Ability <u>*to limit harm*</u> through sharing information (& providing care) presents particularly strong responsibilities, although not necessarily specific to researchers. This feature should be *specifically looked for* where: – There is low general awareness of SCD as a genetic disorder – Harmful alternative explanations for illness (e.g. some supernatural beliefs) are common – Paternal denial of responsibility is common pattern, including through the influence of patrilineal cultural traditions, leading to gendered blame/stigmatisation • *Not all areas of information sharing are likely to carry the same sensitivities; these should be matched to different types of information sharing activities e.g. most sensitive information should be shared within family counselling situation while less sensitive areas could be included in public outreach. • *A particularly sensitive area is the possibility of accusations of misattributed paternity directed at mothers, leading to requests for paternal testing; this is not likely to be seen as a responsibility for researchers. • *The extent to which benefits and harms are prioritised by stakeholders can support resolution of conflicts with competing values (see section 5 on rights and responsibilities). In Kilifi, for example, the interests of vulnerable children and stable families were highly prioritised in decision making about sharing SCD information. • *Where active information sharing not seen as appropriate *on balance* (see section 4 below on 'balancing'), particularly for carrier status, researchers should consider *passive* forms of disclosure, and inclusion of this option as part of informed consent process.
RESEARCHER-PARTICIPANT/COMMUNITY RELATIONS	
Trust: Do study activities, including community	• *Since sharing information from SCD screening has high risk of generating diagnostic interpretations of studies,

engagement and informed consent support trust between research and participants/community? • What risk of diagnostic interpretations? Is understanding of study undermined? Are expectations of reciprocity unrealistically lowered, with potential to undermine fair benefits? • Do research activities demonstrate transparency and consistency in participant/community relations? • Are expectations of due concern about SCD information being addressed?	undermining informed consent, researchers should counter through: – Community engagement & informed consent processes – Research design? (e.g. de-link consent for SCD screening and main study) • *To maintain appropriate levels of trust, researchers should: – Understand and address expectations of due concern and reciprocity (including ensuring fair levels of perceived reciprocity in absence of diagnostic interpretation of research) – Ensure transparency and consistency in relations/activities • *Loss of trust in one study can have implications for other research, at the same time or in the future.
Intensity of engagement: What is the relationship between researchers and participants/the wider community during the study, *and for other linked research*?	• Higher levels of intensity of research engagement with participants generate increased responsibilities for information sharing & care, according to principle of reciprocity. • *But low levels of engagement do not necessarily *undermine* researchers' responsibilities, given importance of justice (particularly important for researchers, given their primary role to support a public health good). • *Participant/community perceptions of intensity of engagement may differ from those of researchers; these should be taken into account in policy on information sharing and/or community engagement.
LOCAL/NATIONAL RESOURCES AND PARTNERSHIPS	
Nature of resources: What is needed to validate tests, provide counselling and *long term care* for affected children?	• Important to take account of *community perceptions of SCD* to assess resources needed e.g. through local consultation and/or formative research. • Likely that these resources/costs can be anticipated in advance of study, and therefore possible to include in research planning & funding. • *Long term care important but unlikely researchers can provide. Partnership with Ministry of Health (MoH) is essential in planning short/long term SCD health services to support research, including communication strategies.
Existence of SCD services: What services already in place for SCD diagnosis and management, including public information? How accessible are services?	• Existence of adequate SCD services increases researchers' responsibility to share information, since competing responsibilities for resource prioritisation are reduced. • *Absence/inadequacy of services increases researchers' responsibilities to work with MoH on implementing/strengthening services; may reduce responsibilities to share information directly with participants *in short term,* depending on research resource prioritisation and MoH policy.
National SCD health policies: What government national/local	• *Strong partnership with MoH at national and local levels are an important basis for studies involving screening/testing for SCD; this feature increases with greater prominence of SCD research agenda.

policies/programmes for SCD (even if not implemented at all levels)? How would researchers sharing information fit with this? What other health programmes are in place that could either support or challenge SCD information sharing (particularly HIV/AIDS)?	• *Need for partnership with MoH underlines importance of taking national/local SCD policy into account, since researchers cannot adequately meet long term responsibilities for provision of services alone. • *Research involving SCD screening/testing should consider ways in which studies can help to develop policy, if needed.
2. STUDY RELATED ISSUES	
SCD prevalence: Is the research being conducted in an area where malaria/SCD is prevalent?	• Higher prevalence of SCD/malaria increases researchers' responsibilities to share information given the associated increased level of harm; and may increase resources needed to ensure benefits, depending on existing services.
Feasibility Is it possible to return SCD findings, or are there reasons that this is unlikely to be possible e.g. anonymised or historical samples	• *In some situations it may be impossible for researchers to share SCD findings with individual participants, but responsibilities remain to share research findings with MoH, and to contribute to strengthening SCD services in keeping with local/national policy or policy making processes (where no policies are in place).
Social value research How important is the research, particularly to the study community? How important is SCD information to the research questions?	• The acceptability of a policy *not* to share SCD findings during studies with affected families may be increased where the research questions are of high social value, particularly if this relates to SCD, even where other features suggest that these findings should be shared (see balancing).
Resource capacity What capacity do researchers have (through planning, including with funders) to take account of resource requirements of SCD information sharing? What impact would this have on funding decisions outside the project?	• Costs of sharing SCD findings (validating tests, counselling & care, in collaboration with MoH) should be anticipated and budgeted for as far as possible. • An important factor that may limit researchers' responsibilities to share SCD findings is the cost to researchers of undertaking this action, particularly where costs importantly affect feasibility of research of high social value. • The acceptability of a policy not to share SCD findings with affected families during studies may be increased for research teams who are nationals of a developing country where research is being conducted (for local benefit), and who do not have access to international funding (see balancing).
3. **RIGHTS AND RESPONSIBILITIES**	
How are the rights of individuals and responsibilities of researchers seen as important, in relation to affected children, those found to be carriers and their families?	• Stakeholder perspectives on the importance of individual rights to information, or rights not to be given information, and researchers' responsibilities to share information, can present counter arguments to claims based on a risk/benefit analysis (as in section 1 above). • These are important areas to explore locally since variation in the way competing claims are balanced may occur

	in different settings and should be taken into account in developing policy as far as possible (see balancing in section 6 below). In Kilifi, for example, rights and responsibilities were consistently seen as less important than the interests of vulnerable children and the stability of families.
4. BALANCING VALUES: WHO DECIDES?	
How can competing values be balanced in a way that maximally supports ethical conduct of research?	• Discussion-based processes with local stakeholders can support exploration of views, including divergence and agreement. Divergence may often be resolved internally by identifying a basis in different perceptions about 'facts', including that some divergence in 'facts' may represent diversity within communities. • Fundamental differences in values may only be resolved by reference to external ethical benchmarks; their validity seems strengthened where local stakeholders also held 'externally' supported views. • Where decisions concern national/local health policies, iterative discussion-based processes would be important to test out proposed solutions to divergence with additional local stakeholders, for example, national/local policy makers, local bioethicists, health providers and community members. These should be transparent & accountable processes, and decisions reviewed over time.