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THE EFFECT OF DISABILITY ON CHILDREN WITH CEREBRAL PALSY AND THEIR FAMILIES

A thesis submitted to the Faculty of Clinical Medicine

at the University of Oxford in partial fulfilment of the requirements for

the degree of Doctor of Philosophy

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APPENDICES

Appendix A Survey documents, questionnaires and newsletters
Appendix B Publications arising from the work described in this thesis

The Effect of Disability on Children with Cerebral Palsy and their Families

Thesis submitted to the Faculty of Clinical Medicine
for the degree of Doctor of Philosophy

Christopher Morris
Wolfson College

Trinity Term 2005

Objectives
To describe the 'activities and participation' of children with cerebral palsy, as defined by the International Classification of Functioning, Disability and Health (ICF), using family-assessed instruments.

Methods
A structured review of family assessed instruments appropriate for measuring children's activities and participation was undertaken to identify questionnaires for use in a postal survey. The survey involved a geographically-defined population of children with cerebral palsy between 6 and 12 years old, identified from the *4Child* database in Oxford. Indices of children's 'activities and participation' were families' assessment of the Gross Motor Function (GMFCS) and Manual Ability (MACS) Classification Systems, the Activities Scale for Kids (ASK) and Lifestyle Assessment Questionnaire (LAQ-CP). To determine the reliability of families' assessments their classifications of the GMFCS and MACS were compared to ratings made professionals; the response frequencies and internal consistency of the ASK and LAQ-CP scales were also examined. Details of children's impairments and abilities were then used as explanatory variables in multiple regression analyses to identify the effect of disability on children's activities and participation.

Results
Families of 129/314 (41%) of the children fully participated in the survey and 175/314 (56%) provided a classification using the GMFCS. These children did not differ from children who did not take part by age, gender or characteristics of their cerebral palsy and associated impairments. Although there was not always perfect agreement the families' classifications of children's movement and manual abilities using the GMFCS and MACS were highly reliable compared to those of health professionals (ICC>0.9). Analysis of the ASK and LAQ-CP showed these also to be reliable. Scores for the ASK and LAQ-CP were generally best predicted by children movement, manual and intellectual disability.

Conclusions
Family assessment of children's movement and manual abilities using the GMFCS and MACS was highly reliable compared to health professionals. In concordance with similar studies that used professionally-assessed measures, children's activities and participation were most adversely affected by movement, manual and intellectual disabilities. Family assessment offers a highly reliable method for measuring activities and participation; however currently available instruments do not fully represent all the domains in the ICF.

ACKNOWLEDGEMENTS

The research presented in this thesis was carried out between 2002 and 2005, for which time I was supported by a Graduate Prize Studentship from the Medical Sciences Division at the University of Oxford. However, the realisation of the studentship and completion of the study would not have been possible without the time, help and encouragement provided by a number of people and it is their contribution to this work that I would like to acknowledge.

Whilst undertaking my Masters' research in 1999 I was encouraged by Clare Robertson, a local paediatrician in Oxford, to contact Ann Johnson at the National Perinatal Epidemiology Unit (NPEU). Around the same time I was repeatedly coming across the research output of a group of child disability researchers at McMaster University in Canada, and particularly the work of Peter Rosenbaum. Following correspondence by email I subsequently visited Peter in Canada and was flattered by his willingness to work with me and help me achieve my academic aspirations. The plan for the study was first hatched at that time, and since then I could not have wished for a better mentor. We have developed a great friendship and the journey has not only been very productive and a lot of fun.

Ann Johnson was, in turn, kind enough to recommend me as a researcher to Leslie Davidson, then Director of the NPEU. The plan evolved further when Peter Rosenbaum visited Oxford and a meeting was held involving Ann, Leslie, Peter and also Jo Garcia. Evaluating the reliability of family assessment for the Gross Motor Function Classification System was a principal outcome of that meeting. My initial applications to the MRC for funding were unsuccessful and it was only a chance exchange with David Graham in the Graduate Studies Office which precipitated the subsequently successful application for the University studentship. Also in this regard, I am grateful to Martin Francis, Senior Tutor at Wolfson College, for his sponsorship and award of a college scholarship.

I began at the NPEU in October 2002 as a fledgling researcher with much to learn. My first office partner, Brenda Strohm, is very experienced in co-ordinating neonatal trials and I hope I can retain at least some of the high standards and efficiency that Brenda fostered in me. Incredibly high standards are a feature across the spectrum of the work of the NPEU and it has indeed been a wonderful learning environment fuelled by an apparently never-ending larder of biscuits, cakes and other treats. I would like to acknowledge several key people at the NPEU. Firstly, my thanks to Peter Brocklehurst the present Director of the Unit who, although not directly involved in the study, has been supportive and encouraging. I am especially grateful to Sarah Ayers and Andy King who have spoiled me somewhat by providing such excellent and responsive computing support; and thanks also to Andy Kirk for providing the artwork for the newsletters and posters. The help of the administrative staff has also been invaluable in conducting the study. Over my time at Unit I shared offices with Liz Schroeder, Juan Nardin and Rona McCandlish and I am grateful to each of them for their moral support. Individually and collectively all the members of the NPEU have helped me in one way or another.

In the second term of my first year, thanks to funding from the Orthotic Education and Training Trust, I was able to spend three months at McMaster University and take the Health Research Methodology course in the Theory and Practice of Measurement. The course tutor, Geoff Norman, was an inspirational teacher and my time in Canada was full of fun. I am very grateful to Mary Law and all the *CanChild* team for making me so welcome as a visitor at *CanChild*, which rivals the NPEU as an exceptionally friendly learning environment

despite only having treat-time once per week. Special thanks are also due to my fellow graduate students at *Mac*, Kerry and Sandy, who made my 'time-off' in Canada such fun. Again sponsored by the Orthotic Education and Training Trust, in the second year, I was able to take the course in Advanced Epidemiological Analysis at the London School of Hygiene and Tropical Medicine in London.

Several people have remained on the periphery of the research but whose help has been nonetheless invaluable. I am grateful to Steve Jarvis, Allan Colver and Helen McConachie in Newcastle for their general interest in the study and their assistance in using the Lifestyle Assessment Questionnaire. My thanks also go to Nancy Young for her assistance in using the Activities Scale for Kids, and to Doreen Bartlett and Bob Palisano for enlightening discussions regarding the Gross Motor Function Classification System. In the latter part of the study I was also fortunate to liaise with Anki Eliasson and Lena Krumlinde-Sundholm and use their newly developed Manual Ability Classification System. I have been fortunate to discuss my work and get help from accomplished statisticians including Geoff Norman, Doug Altman and also latterly Maria Quigley. I have also enjoyed discussing associated research carried out by Jan Willem Gorter and his colleagues at *NetChild* in Holland.

The survey that forms the largest part of the work in this thesis would not have been possible without the not inconsiderable time and effort contributed by the children and families who took part, and also the time taken by their health professionals. I hope that they feel that their efforts were worthwhile when they read the report and newsletters from the study.

Aside from the supreme part played by Peter Rosenbaum I have saved the most outstanding contributions till near the end. Unfortunately, the formal supervision initially provided by the university did not work out well; however, as on so many occasions, adversity created opportunity. The problem brought me into contact with Ray Fitzpatrick and Jenny Kurinczuk who have, since the beginning of the second year, been an exemplary supervisory team. Their friendship, help and support has enabled me to not only to complete the work on time and to a higher standard, but also helped me to win a prestigious Training Fellowship in Health Services Research funded by the Medical Research Council. I am very grateful to them both for their support and look forward to future collaboration.

I will lastly mention my clinical background as an orthotist, the profession I trained in with my father when I first left school, and the experience of which has shaped much of my thinking about health services and disability. I am grateful to my father for instilling in me his high standards, enthusiasm and belief in doing a job to the best of one's ability. As a trained teacher my mother always naturally encouraged me to achieve academically. My mother was in fact completing her own Masters' degree in psychology when I arrived in the world and may well have pursued her own doctoral studies if not choosing instead to care for me full time. I therefore dedicate this doctoral thesis, with love and thanks, to my parents who have waited patiently for me to fulfil my potential. I only hope that my academic odyssey will continue to be as fulfilling and enjoyable in the future.

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THE THAMES VALLEY



CHAPTER 1

INTRODUCTION

1.1 RATIONALE AND STRUCTURE OF THE THESIS

The term "cerebral palsy" is a diagnostic label that describes a specific clinical syndrome. This nosological label can only be applied when key clinical observations are found. The most frequently cited definition of cerebral palsy was published by Bax (1964) as "a disorder of posture and movement due to a defect or lesion in the immature brain". The term *cerebral palsy* is now familiar to most health and social service professionals, as well as to many members of the general public as a physically disabling condition. In fact, although cerebral palsy only affects between 2 and 3 per 1000 live births, it is the most common cause of serious physical disability in childhood (Surveillance of Cerebral Palsy in Europe 2000).

Historically cerebral palsy has predominantly been studied in relation to the pathology and aetiology of the impairment. Discussion regarding the definition and classification of cerebral palsy was first recorded in the medical literature during the nineteenth century, predominately in French, German and English language publications. However, what exactly the term cerebral palsy describes has been debated for more than 150 years, and discussions about how the different manifestations of cerebral palsy can be best classified continue to the present day. The major part of *Chapter 1* outlines this history which includes contributions from several of medicine's eminent figures such as Osler and Freud. Then, in *Chapter 2*, an overview is provided of how 'health' has been conceptualised in recent years, focusing on the World Health Organisation's International Classification of Functioning, Disability and Health (ICF). As explained in more detail in *Chapter 2*, the ICF distinguishes the body's core functions and structures from function related to activities and participation in life situations. Children's health status can be measured using a variety of standardised instruments and how these are developed is explained together with an overview of how such instruments have been used to describe the health of children with cerebral palsy.

The knowledge gaps identified in the two introductory chapters are then synthesised in *Chapter 3* into the specific research questions, aims and objectives that were addressed in the studies described in this dissertation. Fundamentally, the thesis considers whether family assessments of children's abilities are reliable compared to those of health professionals and then what effect disability has on the activities and participation, or 'health', of children with cerebral palsy and their families.

One of the key opportunities in undertaking this work was to collaborate with *4Child* Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children to recruit families. It was expected that this would to some extent overcome the bias that occurs from only recruiting children who attend clinics. The main Thames Valley study invited families of a geographically defined population of children with cerebral palsy to take part in a postal survey. The aim was to measure and describe the 'activities and participation' of children with cerebral palsy, as defined by the ICF, using family assessed instruments. Before the survey could commence however, appropriate valid and reliable instruments had to be identified. This preparatory work is described in *Chapter 4* including a structured review of family-assessed instruments appropriate for measuring children's 'activities and participation' as defined by the ICF (Morris et al. 2005) (Appendix B).

The Gross Motor Function Classification System (GMFCS) has become the standard method of describing the severity of the movement disability for children with cerebral palsy (Morris and Bartlett 2004) (Appendix B). Originally the GMFCS required a health professional to make the classification for each child. One aim of the research described in this thesis was to examine the reliability of family assessment of the GMFCS for their child. A preliminary study investigating the reliability of family report for the GMFCS compared to

classifications made by health professionals was undertaken whilst the researcher was a visiting student at McMaster University in Canada in winter 2003. Details of that study are described in *Chapter 4* (Morris et al. 2004) (Appendix B).

Following the preparatory work the main study was conducted with the Thames Valley population. The methods by which the Thames Valley study was carried out are described in *Chapter 5*. The study population was a complete geographically defined population of children with cerebral palsy between 6 and 12 years old. Families were identified and traced, and the permission of children's general practitioner was sought to contact them. Indices of children's 'activities and participation' used were the family's assessment of the Gross Motor Function and Manual Ability Classification Systems (GMFCS and MACS), the Activities Scale for Kids (ASK) and Lifestyle Assessment Questionnaire (LAQ-CP). Classifications of children's GMFCS and MACS were also gathered from health professionals nominated by families. The results of the main study are described in *Chapters 6*, *7* and *8*. *Chapter 6* examines whether the children and families who fully participated in the survey were representative of the study population by comparing the known characteristics of those who did and did not take part. Then, in *Chapter 7*, the performance of the survey instruments was evaluated, including the reliability of family report for the GMFCS and MACS in comparison with classifications made by professionals. Finally, in *Chapter 8*, details of children's impairments, recorded by *4Child* when the child was aged 5 years, and their abilities gathered using family-assessed measures in the survey, were used as explanatory variables in multiple regression analyses to identify the effect of disability on children's 'activities and participation'.

The results are discussed in *Chapter 9* with particular reference to similar surveys with families, and studies that used professionally-assessed measurement of children's abilities. The contribution that the research described in this dissertation has made to our general understanding about the effect of disability on children with cerebral palsy and their families is also discussed. *Chapter 10* presents the conclusions and recommendations arising from this research and indicates to what extent the aims and objectives described in *Chapter 3* were achieved. The remainder of this chapter reviews the historical context of the research described in this thesis.

1.2 HISTORY OF THE CLASSIFICATION OF CEREBRAL PALSY

1.2.1 Before 1900

The quest to correlate brain lesions with their clinical manifestation began with early French publications by pathologists debating the association of hemiplegia of the body with hemiatrophy of the brain identified by post-mortem (Lallemand 1820; Cazauvieilh 1827 cited in Ingram 1984). However, the seminal work describing *cerebral paralysis*, and particularly the related musculo-skeletal issues, was elucidated by an English orthopaedic surgeon named William Little in one of a series of lectures in 1843 entitled "Deformities of the Human Frame". Whilst his lectures focused on joint contractures and deformities resulting from long-standing spasticity and paralysis, Little clearly indicated that the cause of the spasticity and paralysis was often damage to the brain during infancy, and specifically premature birth and perinatal asphyxia (Little 1843). Little also noted that behavioural disorders and epilepsy were only occasional complications and not central to the condition.

At about the same time a German orthopaedic surgeon, von Heine, was reporting similar clinical syndromes as a result of infections such as scarlet fever and vaccinations (von Heine

1860). He cited the work of his compatriot Henoch, who had written his dissertation several years earlier, describing hemiplegia in children (Henoch 1842). It has been suggested that it was actually von Heine, rather than Little, who first distinguished cerebral palsy from the flaccid paralysis caused by poliomyelitis (Bishop 1958; Osler 1889). However, Little was known to have spent some years studying in Germany during the 1830s and it is possible that there was some cross-fertilisation of ideas, although this is not formally recorded. Regardless, cerebral palsy was known for many years after as "Little's Disease".

In his best known work, published in 1862, Little expands on the association between a large number of his patients' clinical presentation and their birth history as recalled by the family (Little 1862). Little differentiated between the congenital deformities observed at the time of birth, such as clubfoot, and the limb deformities that developed subsequent to premature, difficult or traumatic births, due to what he termed spastic rigidity. He demonstrated his familiarity with the work of French, German and Irish pathologists in constructing his theory. Little grouped the clinical presentation of 47 cases as either (i) hemiplegic rigidity affecting one side only, although lesser impairment of the apparently uninvolved limb was frequently observed; (ii) paraplegia affecting both legs more than arms; and (iii) generalised rigidity. Little showed careful consideration for his audience in the published discussion by conceding to the President of the Obstetrical Society of London that for every "one [case] that depended on abnormal or premature labour there were twenty or more from other causes incidental to later life". Sarah McNutt, an American physician, continued to raise the profile of the risks of long term disability arising from birth trauma (McNutt 1885). Notably, the American Neurological Association admitted her as their first female member; but the content of her lectures apparently made her unpopular with some eminent obstetricians whilst she was on a tour in the UK (Ingram 1984).

At the time he was resident in America, the eminent Canadian William Osler published articles in 1886 and 1888 before his more notable monograph was published in London in 1889. 'The Cerebral Palsies of Children' comprehensively described his study of a case series of 151 patients (Osler 1889). Osler acknowledged the contributions from his German, French, English and American colleagues and stated that he will "for clearness and convenience adhere to custom and classify cases according to the distribution of the paralysis, whether hemiplegic, diplegic or paraplegic". In fact, he classified his cases into the three categories but used the terms (i) infantile hemiplegia; (ii) bilateral spastic hemiplegia; and (iii) spastic paraplegia. Osler references the synonym *spastic diplegia* for bilateral spastic hemiplegia to Samuel Gee at St Bartholomew's Hospital in London. William Osler later moved from Pennsylvania to become Regius Professor of Medicine at the University of Oxford and was knighted in the UK for his contributions to medicine.

In the year following Osler's seminal book, the neurologists Sachs and Peterson published their series of 140 cases (Sachs and Peterson 1890). They contrasted the comprehensive understanding that had then been achieved regarding the clinical symptoms and pathology of poliomyelitis with the dearth of understanding about cerebral palsy. Sachs and Peterson followed the convention of the time by using the same classification system as Osler: hemiplegic, diplegic or paraplegic. Where possible they investigated aetiology using post-mortem examinations but concluded that any of the three clinical presentations could result from a variety of causes. Despite this lack of correlation they advocated that classification should include "special reference to the pathology of the disease".

Sigmund Freud was of the opposite opinion (Freud 1893). Despite his background in neuropathology he advocated classifying cerebral palsy using only clinical findings. Freud

recognised that, even with post-mortem examination, the pathological findings resulted from a combination of the initial lesion and repair process and therefore were only partially related to the clinical manifestation. His classification system combined previously separate categories under the single term 'diplegia' for all bilateral disorders, as distinct from hemiplegia. The term diplegia was used to describe generalised rigidity of cerebral origin, paraplegic rigidity, double spastic hemiplegia, generalised congenital chorea and double athetosis. Athetosis had already been described, initially by Hammond as involuntary writhing movements in adults affected by hemiplegia (Hammond 1871), and it was later more clearly differentiated from other movement disorders by Gowers (1876). Freud's observations regarding aetiology identified three groups of causal factors: (i) maternal and idiopathic congenital, (ii) perinatal, and (iii) post-natal causes. He noted that it was difficult to know whether later problems resulted from birth trauma, as described by Little, or whether in fact there were predisposing factors that may have caused these infants to have a difficult births. He thought the task of separating congenital from acquired cases impossible in some cases and generally unhelpful. Freud was aware that children with ataxic symptoms might require a separate group, as became the case after the work of Batten (Batten 1903), but at the time of his writing he had not seen enough cases of non-progressive ataxia to be sure.

Freud lost interest in cerebral palsy and instead turned towards his study of psychoanalysis (Accardo 1982). Nevertheless, such was his influence that his lasting statements regarding the futility of attempting to associate clinical syndromes with neuropathology may have predisposed to the dearth of research about cerebral palsy during the first half of the twentieth century. Also, at that time, poliomyelitis and tuberculosis were more common causes of disability and therefore attracted greater attention from medical researchers.

1.2.2 From 1900 to 2000

In the early 1920s, some thirty years after Freud's comments an American orthopaedic surgeon made the next major contribution to our understanding of cerebral palsy (noted by Mac Keith and Polani 1959). Winthrop Phelps pioneered modern approaches to the physical management of children with cerebral palsy advocating physical therapy, orthoses and nerve blocks. In a later article Phelps identified his four treatment goals: locomotion, self-help, speech, and general appearance (Phelps 1941). His approach to surgery was conservative. Phelps acknowledged the need for a neurological classification system for diagnostic purposes but preferred to use his own classification system as a basis for treatment. He proposed that classification should be made on a functional basis including both mental and physical ability, and that a social assessment should precede treatment. Phelps grouped all movement disorders under the term dyskinesia, and used spasticity, athetosis, overflow or synkinesia, incoordination or ataxia and tremor as sub-categories. He noted that these five varieties rarely occurred in pure form. Phelps helped to found the American Academy for Cerebral Palsy in 1947 and was elected its first president. The Academy's mission remains "to foster and stimulate professional education, research, and interest in the understanding of these conditions and in improving the care and rehabilitation of affected persons" (American Academy for Cerebral Palsy and Developmental Medicine 2005).

American neurologist Myer Perlstein recognised the prevailing confusion regarding classification of cerebral palsy and contributed a lucid account of the various systems that existed in the 1940s and 1950s (Perlstein 1952). He recounted methods for classifying children according to the anatomical site of the brain lesion, clinical symptoms, degree of muscle tone, severity of involvement, and aetiology. Thus, he suggested that a modular description using components from each category can be assembled. Minear conducted a

survey with the members of the American Academy for Cerebral Palsy in 1953 and published the resulting classification system based on their majority opinion (Minear 1956). He defined cerebral palsy simply as any "symptom complex" arising from non-progressive brain lesions. Minear's system is similar to Perlstein's in being more of a comprehensive listing of all clinical symptoms with categories for motor impairment, topography, aetiology, supplemental, neuro-anatomical, functional capacity and therapeutic requirement. A separate dimension for functional capacity with four levels is included in the classification but used undefined terms such as mild and moderate limitation of activity.

Meanwhile in the UK, the classification systems used to describe case series by Evans (1948) and Asher and Schonell (1950) comprised different combinations of topography and motor impairment. Wyllie used a confusing combination of neurological and aetiological criteria to define categories (Wyllie 1951). These were (i) congenital symmetrical diplegia, (ii) congenital paraplegia, (iii) quadriplegia or bilateral hemiplegia, (iv) hemiplegia. The selected category was supplemented with a statement of the type of motor disorder: spastic, flaccid, mixed, athetoid or ataxic. Harking back to Freud's argument that it was not possible to classify using aetiology, Ingram preferred a system using neurological and topographical categories, supplemented with an indication of the severity using the terms mild, moderate and severe (Balf and Ingram 1955). The Ingram classification separated hemiplegia, double hemiplegia and diplegia from ataxic and dyskinetic categories. Ingram grouped involuntary movement disorders, such as dystonia, chorea and athetosis, under the term dyskinesia. Ingram pointed out that transient changes in muscle tone seen consistently in children with diplegia would require their continual reclassification if the terms rigidity or spasticity were used as categories.

Again in the UK, in 1957 Mac Keith and Polani convened an informal group called the Little Club that was dedicated to thinking through the terminology for describing cerebral palsy. The Little Club published its definition of cerebral palsy as "a permanent but not unchanging disorder of movement and posture, appearing in the early years of life and due to a non-progressive disorder of the brain, the result of interference during its development" (Mac Keith and Polani 1959). The Little Club classification uses the term 'spastic' with sub-categories of hemiplegia, double hemiplegia and diplegia; the other categories were dystonic, choreo-athetoid, mixed, ataxic and atonic cerebral palsy. Ingram continued his aforementioned criticism citing the changes observed in the series of 1821 patients by Bronson Crothers (Crothers 1951) that would require cases to be moved continually between classification categories (Ingram 1984). Some of the original Little Club members refined the definition of cerebral palsy as "a disorder of posture and movement due to a defect or lesion of the immature brain" and "for practical purposes disorders of short duration, due to progressive disease or due solely to mental deficiency were excluded" (Bax 1964). The group noted the inconsistent interpretations of terms such as spastic between different professional and country cultures. These inconsistencies precluded further progress which led to their conclusion that, at that time, it was "impossible to proceed definitively with classifying cerebral palsy" (Bax, 1964).

In the 1980's another self-assembled expert group discussed how to classify cerebral palsy from an epidemiological perspective (Evans and Alberman 1985; Evans et al. 1986; Evans et al. 1987). They were interested in monitoring rates of cerebral palsy in the population as public health markers of perinatal and neonatal health care. The subsequent 'Evans form' recorded details of central motor deficits in terms of the neurological type: (i) hypotonia, (ii) hypertonia (including stiffness, spasticity and rigidity), (iii) dyskinesia and (iv) ataxia (Evans

et al. 1987). A decision was made to record details of each limb and the head and neck separately. The 'Evans form' also enabled recording of functional mobility and manual dexterity in one of four ordinal levels, the presence of intellectual and sensory impairments, communication difficulties, seizures, congenital and acquired malformations, as well as genetic and other disorders. However, in keeping with previous systems, as far as one can determine, this approach to classification was not subjected to any evaluation of its reliability and validity as a measurement tool.

A summary of several meetings held in Europe and America between 1987 to 1990 was published by Mutch et al. (1992) resulting in a further revised definition to underline the heterogeneity of the condition: "an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development". Notably this annotation also included a revised Swedish classification system which, whilst still not perfect, offered simplicity as its major asset. The three neurological categories were spastic, ataxic and dyskinetic; these were subcategorised in mixed ways as hemiplegia, tetraplegia or diplegia for spastic cases; as either diplegic or congenital for ataxic cases, and as either mainly chorioathetotic or mainly dystonic for dyskinetic cases. Whilst noting that at the time it remained beyond their capability, the authors resuscitated the yearning for an aetiologically-based classification system (Mutch et al. 1992).

The Gross Motor Function Classification System (GMFCS) was developed in response to the need to have a standardized system for classifying the severity of movement disability among children with cerebral palsy (Palisano et al. 1997). Previous descriptive systems had included three levels, such as (i) *mild, moderate* or *severe*; or four levels such as (ii) *non-*

ambulatory or *physiological, household* and *community walkers* (Hoffer 1973); (iii) the Evans system: *not walking, restricting lifestyle, functional but not fluent,* or *walks fluently* (Evans 1985). A five level description of children's ambulatory ability was reported by Hutton et al. in their study of factors affecting life expectancy, though they collapsed the data into only two categories of 'walking' and 'not walking' for analysis (Hutton et al. 1994). However, there were no evaluations of the validity and reliability of any of these systems until the GMFCS.

Palisano and colleagues used the underlying construct of self-initiated functional abilities in sitting and walking and the need for assistive devices, such as walkers or wheelchairs, to develop the GMFCS and systematically tested its validity and reliability (Palisano et al. 1997; Wood and Rosenbaum 2000). The GMFCS describes movement ability of children with cerebral palsy in one of five ordinal levels. The GMFCS currently includes descriptions of children's abilities for each level across four age bands: less than 2 years, 2-4 years, 4-6 years and 6-12 years, with an adolescent age band currently under development. Children in Level I can perform all the activities of their age-matched peers, albeit with some difficulty with speed, balance, and coordination; children in Level V have difficulty controlling their head and trunk posture in most positions and achieving any voluntary control of movement. A full description of children's movement abilities in each GMFCS Level between 6 and 12 years old are shown in Table 1.1. A complete guide to the GMFCS is available in Appendix A in the standard user instructions.

Since its publication the GMFCS has become established as the principal classification system for describing the severity of motor disability for children with cerebral palsy. The classification system has had good uptake internationally and across the spectrum of health

care professions for use in research and clinical practice by providing a system for clearly communicating about children's gross motor function. A review of citations of the GMFCS undertaken in 2003 found over 100 references, with applications in both observational and experimental studies (Morris and Bartlett 2004) (Appendix B). In 2005 the number of citations of the original paper on the GMFCS by Palisano et al. (1997) is nearing 200.

Table 1.1 Descriptions of Gross Motor Function Classification System (GMFCS) Levels between 6th and 12th Birthday (Palisano et al 1997)

Level I
Children walk indoors and outdoors, and climb stairs without limitations. Children perform gross motor skills including running and jumping but speed, balance, and coordination are reduced.

Level II
Children walk indoors and outdoors, and climb stairs holding onto a railing but experience limitations walking on uneven surfaces and inclines, and walking in crowds or confined spaces. Children have at best only minimal ability to perform gross motor skills such as running and jumping.

Level III
Children walk indoors or outdoors on a level surface with an assistive mobility device. Children may climb stairs holding onto a railing. Depending on upper limb function, children propel a wheelchair manually or are transported when travelling for long distances or outdoors on uneven terrain.

Level IV
Children may maintain levels of function achieved before age 6 or rely more on wheeled mobility at home, school, and in the community. Children may achieve self-mobility using a power wheelchair.

Level V
Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At level V, children have no means of independent mobility and are transported. Some children achieve self-mobility using a power wheelchair with extensive adaptations.

1.2.3 From 2000

Following a survey of practice across the continent, the group for the Surveillance of Cerebral Palsy in Europe (SCPE) published their standardised procedures for ascertaining and describing children with cerebral palsy for registers and databases (Surveillance of Cerebral Palsy in Europe 2000). The definition was largely a reiteration of that proposed by Mutch and colleagues (Mutch et al. 1992) and included five key points:

- Cerebral palsy is an umbrella term.
- It is permanent but not unchanging.
- It involves a disorder of movement and/or posture and of motor function.
- It is due to a non-progressive interference, lesion or abnormality.
- The interference, lesion or abnormality is in the immature brain.

The system adopted by SCPE provides a decision flow chart to aid classification into neurological and topographical categories including spastic (unilateral or bilateral), ataxic, dyskinetic (dystonic or choreo-athetotic) or not classifiable. Clearly defined symptoms and requirements are provided for each neurological category. Despite careful planning of the system, there has been little work to demonstrate the validity and reliability of classification. The lack of any defined criteria for recording functional limitations in the SCPE definition was noted by Lenski et al. (2001). Subsequently, and with others, the same research group demonstrated that the inclusion of a description of functional ability markedly improved the reliability of diagnosing children with cerebral palsy (Paneth et al. 2003). Consistent application of the diagnosis is of paramount importance when the prevalence of cerebral palsy from different sources and places is being compared. The utility of less well defined labels such as 'diplegia' has therefore been criticised (Colver and Sethumadhavan 2003) and,

in response to this, the importance of using valid and reliable measures of children's functional ability was emphasised (Morris and Rosenbaum, 2003) (Appendix B).

With rapidly improving imaging technology there is renewed interest in correlating clinical syndromes and neuro-anatomy, challenging Freud's 100 year old statement that this was a futile task. Progress has been made using ultrasound and magnetic resonance (MR) imaging to detect structural impairments of the brain before they manifest as movement disorders (Accardo et al. 2004). MR imaging can also be used to approximate the timing at which the brain was damaged based on normal neurodevelopmental stages (Barkovich 2001; Krageloh-Mann 2004). Using this method brain malformations occurring during the first or second trimesters of pregnancy are thought to be structural, as the developing neuronal cells are then involved in proliferation, migration and organisation. However disturbances to the brain in the third trimester, when brain development is more differentiated, are observed as specific lesions to either the white or grey matter. A further division is made between 'early' and 'late' third trimester events. During the early third trimester the periventricular white matter is at greater risk and towards the end of the third trimester it is more often the grey matter that is affected, particularly the basal ganglia and thalamus. Matching neuro-anatomy with clinical syndromes is more complicated although the topographical location and the extent of the damage to the brain can be indicative of the clinical consequences. Only partially explained to date, Krageloh-Mann (2004) summarises some crude correlations that are emerging between timing and location of the lesion and functional, cognitive and sensory impairments.

The search for a single internationally accepted definition of cerebral palsy continues to the present day. An international multidisciplinary group of experts met in 2004 and some of

the participants then revised the oft cited definition by Bax (1964) to recognise that the key motor deficit is often accompanied by other neurodevelopmental impairments. The new proposed definition is that:

"Cerebral palsy (CP) describes a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy may be accompanied by disturbances of sensation, cognition, communication, perception, and/or behaviour, and/or by a seizure disorder." (Bax et al. 2005).

Whilst welcoming the debate and the desire for consensus the new definition received mixed reviews in the accompanying editorials. Carr considered how the definition and classification would affect clinical practice and the challenge of shifting from traditional modes of thinking (Carr 2005); whereas Blair and Love (2005) considered the precision of the definition to be flawed in the same way as previous attempts, and particularly from an epidemiological perspective. Chiefly they point out that the term "non-progressive" is no further defined than before, neither are the age limits and lower limit of severity for inclusion or what syndromes should specifically be excluded. Blair and Love did not however provide any suggestions to address these issues. Whilst the precision with which the definition is applied by clinicians may have negligible consequences for treatment, the implications for measuring rates of cerebral palsy over time are more profound.

Two major paradigm shifts in health related research over recent years are relevant to the epidemiology of cerebral palsy. First, the World Health Organisation has introduced the International Classification of Functioning, Disability and Health to distinguish impairments

of body functions and structure from consequent activity limitations and restrictions in participation (WHO 2001). The second major shift has been an increasing awareness of the utility of valid and reliable patient-assessed measures for describing health status. The relevance of these two issues to the study of cerebral palsy is described in the next chapter.

1.3 CHAPTER SUMMARY

In summary, it appears that in the 150 years since William Little first associated limb deformities with premature and difficult births, consensus on what the term cerebral palsy really means and exactly what conditions are included remains elusive. One reason that achieving consensus has been so difficult is the variety of different purposes for which classification systems are used. Classification has been used for studying the anatomy and pathology of the neurological impairment, the aetiology or causal pathways resulting in cerebral palsy, and the clinical presentation for the purpose of treatment and surveillance. Those involved in surveillance have been particularly concerned about the definition as minor changes could have profound effects on the validity of rates monitored over time.

The new proposed international definition (Bax et al. 2005) can however be used to identify children with conditions falling under the umbrella diagnosis of cerebral palsy. The SCPE classification of the type, subtype and distribution of the impairment, combined with the GMFCS to describe the severity of movement disability, are currently regarded to be the best methods for consistently describing children with cerebral palsy for surveillance purposes. The new proposed classification includes four elements (i) the type of motor disorder and severity of functional limitations; (ii) associated impairments; (iii) anatomical distribution and radiological findings; (iv) any information on causation and timing of the disturbance should it be available. However, whilst the present classification systems serve the purposes for which they were created they offer only summary accounts of a person's functional status, and provide few details regarding children's overall health and how any impairment affects the lifestyles of the children with cerebral palsy and their families. The classification and measurement of children's health, particularly using family assessment, is the subject of the next chapter.

CHAPTER 2

MEASURING CHILDREN'S HEALTH

2.1 DEFINING AND CLASSIFYING HEALTH

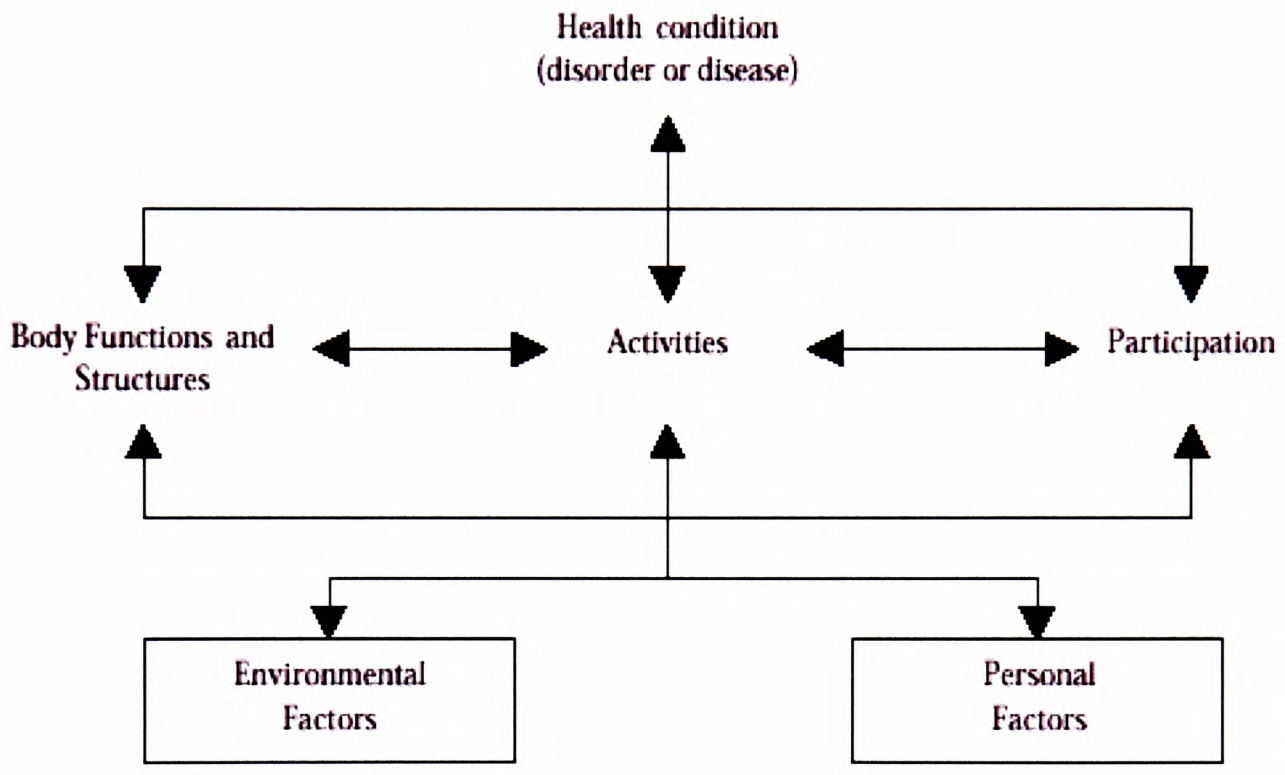
In one of its early reports the World Health Organisation (WHO) recommended that a person's health is defined by their "physical, psychological and social wellbeing and not merely the absence of disease or infirmity" (World Health Organisation 1958). Although not always universally accepted, this triad of domains became the foundation for defining the associated concepts of 'health' and 'quality of life'. The WHO later also emphasised the importance of personal autonomy and others have since proposed economic and spiritual domains (Spilker 1996). The traditional medical model suggested that health is adversely altered as a direct consequence of any illness, causing disability. By contrast the 'social model' of disablement, popularised in the 1970s, conceived handicap as a socially created problem due to the interaction of person and environments rather than as a problem attributable solely to the individual. The importance of this shift, and its widespread acceptance, has been noticeable in the realm of advocacy and rights for disabled people to challenge inequalities, and has also influenced subsequent approaches to defining and classifying health.

2.1.1 International Classification of Functioning, Disability and Health (ICF)

The International Classification of Impairment, Disability and Handicap (ICIDH) was the WHO's initial attempt to categorise the consequences of diseases into different dimensions (WHO 1980). Recognising the difference between the organic, personal and social elements of ill health was an important breakthrough. The ICIDH encouraged clinicians to manage and measure diseases and their consequences with reference to the appropriate dimensions. However, this model was linear and presumed that impairment led to disability which in turn caused handicap without much reference to any contextual factors. The International Classification of Functioning, Disability and Health (ICF) recently replaced the ICIDH and

is an attempt to address the problems encountered with its predecessor (WHO, 2001). Instead of classifying 'disease' and 'consequences' the labels 'health' and 'components of health' were preferred. Figure 2.1 illustrates the components of health and the way in which they interact in the ICF model.

Figure 2.1 Conceptual model of the International Classification of Functioning, Disability and Health (ICF) showing the components of health and interactions (WHO 2001).



'Functioning' is used as an umbrella expression for the neutral terms 'body structure and functions' and 'activities' and 'participation'. Likewise, 'disability' is the umbrella expression for the contrary terms 'impairments of body structure and functions', 'activity limitations' and 'participation restrictions'. Disability is therefore composed of (i) impairments of body functions and structures, (ii) activity limitations, where there are difficulties in the execution of specific tasks, and (iii) participation restrictions, which are problems experienced when people are involved in life situations.

In the current versions of the ICF 'activities and participation' are combined in a single list, although there is acknowledgement that for some purposes the items may be designated to one or other distinct category (WHO 2001 and 2004). The ICF also includes reference to the environmental and personal contextual factors that interact with impairments to either reduce or intensify activity limitations and participation restrictions. Environmental factors are listed in the ICF beginning with those closest to the person and radiating out to the most general factors. Environmental factors include physical, social, attitudinal barriers or facilitators to activities and participation. Personal factors are not classified in the ICF due to their diversity but would typically include gender, age, education, lifestyle, and a person's own preferences. In contrast to the linear and causal inference proposed in the ICIDH, the ICF model illustrates that interactions occur among all the components of health including the contextual factors. The fact that these interactions are represented using bidirectional arrows emphasises that the components of health can potentially influence one another within a changing system.

'Qualifiers' are used to classify the status for each item contained in the domains of the ICF. Body functions and structure represent both physiological and anatomical aspects of health. The qualifiers used indicate the presence of impairment, and where appropriate the severity of that impairment using a five level ordinal scale. Descriptions are provided to improve the validity of the judgements used to rate a person's impairment at one of these levels. The qualifiers 'capacity' and 'performance' are used for activities and participation. Capacity describes what a person could achieve at their best in a standardised environment. By contrast, performance describes what a person actually does in their regular environment and may include use of assistive technology or environmental adaptations. The gap between a person's capacity and their performance can be used to assess whether the environment is

enabling, when performance equals or even exceeds capacity, or conversely when it constitutes a barrier, when capacity exceeds performance. The ICF is arranged in a hierarchical structure with key chapter headings and three sub-domains, which are labelled sequentially second, third and fourth levels. The WHO suggests that the ICF is useful for classification at the level of individuals, institutions or populations and can be utilised for a variety of policy, economic, and research applications.

When using summary measures to compare the health of populations in different countries, Murray et al. (2002) advocate capturing people's capacity for activities and participation. Murray and his colleagues propose that capacity, albeit capacity whilst using any regular personal aids, corresponds more closely to a 'common-sense interpretation of health' (Murray et al. 2002) and also avoids the effect of comparing different environments. However, such an approach is at risk of underestimating the effect of disabilities of people's lives if consequent performance is not also simultaneously assessed. The work in this thesis focuses on children's performance of activities and participation as the aim was to find out what effect disability has on the lives of children with cerebral palsy and their families.

2.1.2 Utility of the ICF for childhood disability research

Whilst endorsing the 'bio-psychosocial model' espoused in the ICF published in 2001, there was criticism of the appropriateness of the range of life situations included to measure children's participation. The life situations included in the ICF were generally described for adults and there was little recognition of developmental issues (Simeonsson et al. 2000 and 2003). Following a study by Cieza and others linking items from standardized health status instruments to specific ICF categories (Cieza et al. 2002), Battaglia and colleagues assessed the applicability and reliability of the ICF for children with complex disabilities (Battaglia et

al. 2004). A checklist and standardized professionally-assessed health status instruments were used to obtain information. The study showed that, for the most part, the findings from the instruments and the ICF checklist were well correlated. The ICF was therefore deemed applicable for children with complex disabilities although some obvious aspects of children's activities and participation, such as 'play', were found to be lacking. In a study by Ogonowski and her colleagues the reliability of ratings between observers who administered three professionally-assessed instruments to parents of children with disabilities during interviews were examined (Ogonowski et al. 2004). They found good agreement between the instruments for around half of the ICF codes, and cited the adult-based examples and lack of guidance on accommodating child development in the ICF as causes for inconsistencies.

More recently the WHO published a separate ICF version for Children and Youth (World Health Organisation 2004), from which the chapter titles of each of the key domains for activities and participation are shown in Table 2.1. Each chapter has a hierarchical structure, for instance, in the chapter on 'self-care', the item 'washing oneself' includes the levels shown in Table 2.2.

The titles of the major chapters for environmental factors are listed in Table 2.3. For children, many of their activities and participation are dependent upon their family or family members, which are listed as an environmental factor under 'support and relationships'. Children will likewise affect the activities and participation of their parents or carers. Therefore, in some domains, such as domestic and economic life, the child's and carers' participation is difficult to separate and might better be defined in terms of the activities and participation of the family unit.

Table 2.1 Chapter titles of the key domains for activities and participation in the ICF.

Major domains of Activities and Participation
1. Learning and applying knowledge
2. General tasks and demands
3. Communication
4. Mobility
5. Self care
6. Domestic life
7. Interpersonal interactions and relationships
8. Major life areas
9. Community, social and civic life

Table 2.2 Sub-levels of item d510 'washing oneself' in the chapter on 'Self Care' as an example of the hierarchical structure of the ICF.

Chapter 5 Self-care
+ d510 Washing oneself
d5100 Washing body parts
d5101 Washing whole body
d5102 Drying oneself
d5108 Washing oneself, other specified
d5109 Washing oneself, unspecified

Table 2.3 Chapter titles of environmental factors listed in the ICF

Environmental factors
Products and technology
Natural environment and human-made changes to environment
Support and relationships
Attitudes
Services, systems and policies

2.1.3 Describing cerebral palsy in terms of the ICF

By definition in cerebral palsy, the primary impairment to the body functions and structure is damage to the brain and, in addition to the motor component, is manifest in some children by altered cognition, vision loss or other cerebral functions. However, damage also manifests as altered functioning in other parts of the body such as muscle spasticity. The impact of skeletal growth during childhood can compound these primary problems if muscles fail to lengthen in proportion to their adjacent long bones causing progressive musculoskeletal deformities (Graham 2002). As a consequence of these primary and secondary impairments of body functions and structures children may experience a variety of activity limitations and participation restrictions that can change over time.

Two studies by Beckung and Hagberg (2000 and 2002) explored associations between neurological impairments, activity limitations and participation restrictions in children with cerebral palsy aged between 5 and 8 years old. In the first study information about learning disability (IQ), epilepsy, infantile hydrocephalus, visual and hearing impairments and movement ability (GMFCS) were available. In the second study a newly developed five level ordinal measure of manual dexterity, the Bimanual Fine Motor Function, was also employed. The first study utilised several domains of handicap in the ICIDH: orientation, physical independence, mobility, occupation and social integration. Their second study calculated participation domain scores from the ICF for mobility, education and social relations. Both studies involved a health professional assessing the children by reviewing their clinical records. The degree of children's participation was coded using modified version of the handicap code from the ICIDH specially created for children. This scale had a five level ordinal grading ranging from no problem to completely restricted. Beckung and Hagberg reported that movement, manual and learning disability were the best predictors of

children's handicap or participation; there was also a high correlation between the GMFCS and mobility score (Beckung and Hagberg 2002).

Rosenbaum and Stewart (2004) illustrated how the ICF enhances the way that health care of children with cerebral palsy can be considered. Rather than focusing solely on providing interventions at the level of the child's impairment, the ICF suggests that supporting the family using the principles of family-centred service is also likely to benefit the child's health (Rosenbaum and Stewart 2004). Tieman and her colleagues demonstrated that children do not always use the mobility methods implied by their capability because of personal or environmental factors at home, school and outside (Tieman et al. 2004a&b). For instance, children who walk using assistive mobility aids at school often preferred to crawl or roll when moving around their home, and used a wheelchair when travelling longer distances outside due to the varying context of the different environments. When a difference exists between a child's capability and performance, judicious alteration of the environment or provision of assistive technology may improve a child's accomplishment of activities and participation, such as mobility (Palisano et al. 2003). Personal preferences may be used to identify realistic treatment goals that are important to the child and family (Ketelaar et al. 2001). The strength of the ICF is the way in which barriers that have the potential to be changed can be identified to improve a child's performance of activities and participation particularly when the impairment to the body structure and functions cannot be corrected.

In terms of the ICF much of the research related to cerebral palsy has historically focused at the level of body functions and structures. Whilst studying characteristics and causes of the impairment is clearly vital, assessing children's activities and participation is also extremely

important for building a more complete understanding of children's health. As yet, we know relatively little about how severely children with cerebral palsy are affected by activity limitations and participation restrictions as defined in the ICF. One of the key reasons for this knowledge gap has been the lack of appropriate measurement instruments. The next section describes how the science of health status measurement has evolved, considers the different types of instruments that exist, and explains how such instruments are developed.

2.2 MEASURING HEALTH STATUS

Traditionally, a person's health is assessed by a doctor on the basis of an interview and clinical examination related to their physiological signs and symptoms. Whist this approach can be satisfactory for individual consultations it is not standardised and makes comparisons about the severity of health status unreliable. Therefore, in recent years, a variety of health status instruments have been developed in an effort to standardise the assessment of the severity of health conditions. These instruments may require assessment by a health professional, be administered by a trained interviewer, or be completed by the person themselves or someone that knows them well. For instance, the ICF is presented as a checklist to be administered and completed by health professionals as a measure of health status. There has been a particular proliferation of patient-assessed instruments professing to measure aspects of a person's health based upon their responses to items in standardised questionnaires. These self-assessed measures are distinct from instruments that require administration by a health professional, interviewer or other assessor as they directly capture the respondent's own perception of their health. Self assessment has a longer tradition in the disciplines of education and psychology where the methodology is well developed for assessing people's personal characteristics (Streiner and Norman 2003).

In order for a measurement of health status to be considered credible and useful, evidence of the instrument's validity and reliability are required (Jenkinson and McGee 1998). Details of assessing validity and reliability, particularly relating to health status measures, are examined thoroughly later in this chapter in section 2.2.2 on instrument design. However, the concepts are defined in brief here. The validity of an instrument relates to whether the instrument is measuring what it claims and not some other phenomenon. There are several approaches to demonstrating different aspects of validity such as content, criterion and

construct validity (Jenkinson and McGee 1998). The appropriate choice of test and which aspect of validity is assessed may depend on the purpose of the particular instrument under examination. Reliability determines how consistently the instrument measures the aspect of health status for which it is valid. Tests of reliability therefore determine how much error is incurred in the measurement of health status (Streiner and Norman 3003).

The triad of physical, psychological and social wellbeing proposed by the WHO became the foundation for self-assessed instruments that were subsequently developed as measures of 'quality of life'. The terms health status, quality of life, health-related quality of life, and functional status are frequently used interchangeably and there is no consistent agreement about what these titles really mean (Fitzpatrick et al. 1998). There is however consensus that quality of life is a multidimensional concept (Spilker 1996). As some dimensions of quality of life are not otherwise directly measurable by an external assessor, such as satisfaction with health, and because the essence of a self-assessed measurement is that it incorporates the respondent's own views, quality of life is also generally accepted to be conceptually subjective (Schipper 1996).

There have been several substantive reviews focusing on measures of children's 'quality of life'. Eiser and Morse reported a comprehensive review of measures of quality of life for children with chronic conditions (Eiser and Morse 2001); and Schmidt et al. reviewed generic measures of 'health related quality of life' for children suitable for population based research (Schmidt et al. 2002). A bibliographic study of all types of patient-assessed health outcomes for both adults and children (Garratt et al. 2002) resulted in the establishment of a database of Patient-assessed Health Instruments (National Centre for Health Outcomes Development 2003). These and other authors (see Colver and Jessen 2000) have emphasised

the conceptual differences between measuring the 'objective' entity of a child's health as distinct from their 'subjective perception' of their quality of life. Despite the apparently clear distinction, as previous reviewers have reported, considerable inconsistency prevails in the literature regarding what is meant by these terms. Quality of life has frequently been used as a 'catch all' expression for any self report health status measurement even when the instrument is capturing objectively verifiable functional ability, activities and participation. By definition both 'quality of life' and 'activities and participation' are multidimensional constructs. However, whilst measuring quality of life ideally requires the subjective responses of an individual about their perception of health status, the performance of activities and participation can be readily observed and therefore be either self-reported or assessed by an appropriate observer. As McDowell and Newell emphasise, the distinction between objectivity and subjectivity is determined by 'what' is being assessed and not by 'who' makes the assessment (McDowell & Newell 1996).

2.2.1 Types of Instruments

Health status instruments can be categorised in different ways based upon their intended purpose. For instance whether they are *discriminative* and intended to tell people apart; or *evaluative* which means the instrument is responsive to measuring important change over time; or *predictive* which means the instrument produces information correlated with some 'gold standard' outcome not available until a later time (Guyatt et al. 1993). Another common way of classifying measures by their purpose is generic, condition-specific, site specific, dimension-specific, summary items and utility instruments (Fitzpatrick et al. 1998). These types of instrument are considered in turn below.

Generic instruments

Generic instruments are intended to be applicable to everybody whether or not they have a medical condition. Generic measures include questions, or items, which are appropriate for people in general and are not associated with any particular symptoms. The Child Health Questionnaire (CHQ) is the most frequently reported generic quality of life measure for children. The CHQ is multidimensional and includes questions related to physical, social, and psychological wellbeing as well as pain and a general relative perception of overall health (Landgraf et al. 1996). The advantages of generic measures are that they can be used across diagnostic groups, in general population health surveys and for research into conditions where no specific measure is available. The main disadvantage with generic measures can be that, because they cover a wide range of domains with relatively few questions, they may provide only crude discrimination between different health states.

Condition-Specific instruments

In contrast to generic measures 'condition-specific' instruments are intended for use among people with a particular diagnosis or disease, such as asthma or cancer. The Lifestyle Assessment Questionnaire is an example of a condition-specific measure of impact of disability for children with cerebral palsy (Mackie et al. 1998b). The advantage of constructing measures for particular conditions is that the questionnaire will be composed of items that should be relevant to the respondents. This means the instrument should in principle be more sensitive to clinically relevant differences in those persons with the condition, but most likely will not be relevant to unaffected populations. Condition-specific instruments can therefore not be used to compare affected individuals with healthy subjects or those with other conditions.

Site-specific instruments

Rather as condition-specific instruments focus on aspects of a particular diagnosis, site-specific instruments aim to elicit characteristics associated with particular parts of the body. The items composing the site-specific instruments will be relevant to symptoms commonly encountered by people who have a problem with that body part but who are not necessarily suffering from the same condition. For this reason site-specific measures should be more sensitive to aspects of life associated with that body part that may not otherwise be detected using a more general instrument. For example, the Scoliosis Quality of Life Index is intended solely for children with spinal deformities (Feise et al. 2005).

Dimension-Specific instruments

As noted earlier, whatever label is used for health status measurement most people agree that the concept is multidimensional. However, there are circumstances when one dimension may be more relevant for investigation such as physical functioning, psychological wellbeing or pain. In these instances an instrument that focuses solely on a particular domain may elicit more relevant information and be more sensitive than a generic instrument. For example, the Paediatric Pain Profile aims to assess pain in children with neurological disability who are unable to communicate themselves by asking parents 20 questions about their child's behaviour (Hunt et al. 2004).

Summary Item instruments

Summary items are single questions that aim to elicit a person's global estimations of their health and wellbeing. Although these questions are often conceptually multidimensional, the respondent is asked to judge for themselves the relative importance of each dimension and provide an overall answer on a given scale. The main advantage is the brevity of the tool in

comparison to the other types of instrument described. The disadvantage is the loss of detailed information about particular dimensions. However summary items such as "overall how do you rate your general health" may be incorporated into longer questionnaires to analyse patterns of response to other items, or instruments that have broader purposes than solely measuring health.

Utility instruments

Utility instruments are generic in nature but involve either a direct or indirect estimation of the value of a person's health state based either on their own preference for health (direct method) or the preferences of a reference population (indirect method). These instruments have gained importance in the field of health economics because decisions about finite resource allocation are often very difficult to resolve on clinical or cost factors alone. The most commonly used utility measures, such as the Health Utilities Index (HUI), employ the indirect method which takes a societal rather than an individual perspective (Feeny et al. 1996). A reference population is first asked to rate a number of health states and the resulting information used to calculate utility indices for each state. The utility instrument may appear to respondents to be a typical generic instrument in which they rate their current health status. However, the utility indices derived from the reference population are subsequently combined with their responses to calculate a numerical utility index of their health status. Utility instruments are particularly useful for comparing health states associated with otherwise non-comparable conditions to assist with complex decisions about what health services should be delivered.

2.2.2 Principles of Health Status Instrument Design

Most health status instruments involve a number of questions, or items, to which the assessor or respondents provide ratings along a given scale. When constructing an instrument, having decided what purpose it will serve, the first stage is to devise the items.

Identifying Items

Items may sometimes be gleaned from existing instruments that measure the same trait or facet of health, providing that they have been shown to be valid and useful. More often, an instrument is being developed precisely because no existing instrument is appropriate. Consequently the items must be devised from first principles. Sources of ideas for items include the extant literature and current state of knowledge on a subject, patients and their families, expert opinion, clinicians, and clinical observations (Streiner and Norman 2003). Information about the components of health either generally or for particular conditions can be elicited from people using qualitative research methods such as focus groups, individual interviews or other survey techniques to identify relevant themes. Questions appropriate to these themes are then derived to create a series of items. In the developmental stages of the process there is no limit to the number of items that are generated and it is important that the items cover all aspects of the construct being assessed. This is to ensure that the content of the instrument is adequately valid to fulfil its purpose. The next stage involves creating a means for people to indicate their responses to each item. This will usually be dictated by the nature of the question being asked. Some questions require a dichotomous answer, such as yes or no, or the selection of one or more nominal or unrelated categories. These types of items include gender, religion or nationality where a respondent can sensibly indicate only one discrete response. However many questions will require the respondent to indicate their

answer along an ordinal or continuous scale, such how frequently they experience some symptom or to what extent they are with a statement.

Scaling Item Responses

Direct estimation scaling techniques include visual analogue, adjectival and Likert scales (Streiner and Norman 2003). Visual analogue scales (VAS) ask respondents to indicate their status along a line of known length with opposing descriptions at each end of the line for the attribute being measured. VAS techniques are often used for constructs which are difficult to describe other than at the extremes, such as pain. In contrast to VAS, adjectival scales use specific descriptive terms to indicate, incrementally, how much the construct has altered at each step along a given continuum; for example, from none to a little, a lot and all of something. Likert scales are similar to adjectival scales in that they use incremental descriptive terms, however, Likert scales by definition use opposite terms at each end of the continuum, such as 'strongly agree' and 'strongly disagree' with incremental variations in between and perhaps a neutral term, such as 'no opinion', at the midpoint (Likert 1952).

Although the methods appear apparently straightforward important design factors must be taken into account to avoid introducing bias in the construction of items. The descriptive terms must be as clear and unambiguous as possible and the validity of the difference between the increments depends upon them being discernable to respondents. Typically, people can make absolute judgements between five and nine increments across a range of physical phenomena such as musical tones, tastes and shapes, and the range was suggested to lay between three and 18 levels depending on the phenomena (Miller 1956). Miller therefore advocated that, to be meaningful and avoid error, the number of response options offered by questions measuring a single attribute on a scale should usually be at least five but

not normally exceed nine. These direct estimation methods elicit ordinal data but responses to several items are often given numeric scores and subsequently summed together and interpreted and analysed as quasi-continuous or interval level measures.

The rationale for converting judgements into numerical scores has its basis in psychophysics where it has been shown that just noticeable differences correlated well with continuous physical measurements of phenomenon such as weight and light (McDowell and Newell 1996). The principles evolved further in psychometrics, where measurement of many social and health parameters is often not possible, under the banner of classical test theory (Lord and Novick 1968; Nunnally 1978). This classical theory assumes that the distances between responses and their subsequent scores are equal. This assumption is controversial and has lead some scale developers to use item response theory (IRT) to overcome the problem, a popular form of which is named Rasch analysis, (Rasch 1960). In contrast to the classical approach, which uses variance in the respondents and their responses to test the properties of instruments, IRT instead examines the properties of the items and is therefore independent of the characteristics of those subjects for whom the scale is used (Fan 1988). The advantage of IRT being that item responses are scaled according to their relative difficulty, therefore the scale developer is more assured of continuous or interval level measurement and enhanced precision over classically developed instruments. However, in practice, there may be little difference in the way the final instrument is constructed (Fan 1998). Streiner and Norman (2003) recommend, with caveats, that "from a pragmatic point of view" in most circumstances responses can be analysed as interval level data without introducing severe bias (Streiner and Norman 2003).

Alternatives to direct estimation techniques are comparative approaches to scaling. Examples include Thurstone's methods of equal appearing intervals and paired-comparison techniques (Streiner and Norman 2003). These methods involve preliminary work to identify an underlying interval level scale that can then be used to calibrate responses from subsequent subjects. Whilst the credibility of interval level measurement offers powerful statistical advantage over direct estimation methods, a great deal of time-consuming preparatory work must be undertaken. This may be indicated when the issues being measured are emotive, and bias is likely using other methods. Guttman scaling is a comparative method that can be used when all the items are assessing a single underlying construct (Streiner and Norman 2003). Items are eventually ordered by increasing difficulty of each item relative to others resulting in an ordinal scale. As mentioned earlier, econometric methods are used to design utility measures based on people's preferences, and usually these are the preferences of a reference population rather than those of the affected individuals.

Selecting Appropriate Items

With many items, the item pool is usually initially too unwieldy to be useful and therefore must be reduced. Any items which are vague or open to misinterpretation, for instance because they use professional jargon, should be removed. The remaining items can then be piloted with a sample of the target population to enable further evaluation of their interpretability and some initial psychometric testing of the scale. Items that are frequently not answered at all or elicit the same response from everybody are not useful as they do not discriminate between people.

When items are to be summed together to create a composite scale score, the scale items must be homogenous, that is, measuring the same underlying concept (Nunnally 1970). The means to undertake this assessment are provided either by item-total correlation, split-half reliability or Cronbach's alpha coefficient. These statistical techniques evaluate the internal consistency of a scale and to what extent consistency is influenced by each item; items that are not associated with the scale can subsequently be removed. With the item-total method each item should demonstrate a Pearson's correlation coefficient with the scale total above 0.2, and any items with lower correlations should judiciously be removed (Kline 1986). Scales that achieve Cronbach's alpha coefficients between 0.8 and 0.9 are generally acceptable for use in populations (Guyatt et al. 1993). However, as internal consistency naturally increases with the number of items in a scale, instruments that show reliability coefficients exceeding 0.9 may simply include a number of redundant items; conversely low reliability coefficients may be associated with too few items in a scale (Fitzpatrick et al. 1998; Streiner and Norman 2003).

With multidimensional instruments the homogeneity of each dimensional scale should be assessed separately. The internal consistency of the multidimensional instrument could be tested by an extension of the item-total method correlating scale totals with an overall total but is usually assessed more elegantly using factor analysis or multidimensional scaling. These techniques mathematically group together items that appear to be measuring a similar construct as explanatory factors. The interpretive task of labelling exactly what each factor is indeed measuring is then left to the scale developers.

Demonstrating Validity and Reliability

- Validity

The validity of a measure is the extent to which the instrument is assessing what it intends and not some other phenomenon. As described, items should have been selected for the instrument precisely because they are most obviously representative of the concept; this is termed face or content validity. To establish construct validity the developers should demonstrate a clear conceptual framework that underpins the instrument, and then provide evidence that resulting measurements correlate well with instruments measuring the same construct, termed *convergent* validity, or correlate poorly with instruments that are measure something unrelated, termed *divergent* validity (Jenkinson and McGee 1998).

Instruments are generally designed to be either discriminative or evaluative. To establish discriminative validity an instrument must detect known differences between respondents. As evaluative instruments aim to measure change within individuals they must in addition demonstrate their responsiveness, which describes the sensitivity of an instrument to detect real and important change. Statistical techniques such as the 'effect size' (Cohen 1988) or 'standardised response mean' (McHorney and Tarlov 1995) have been used to demonstrate this necessary measurement property of evaluative instruments. There are however conceptual limitations with these methods as, for instance, large effects are easier to detect than smaller ones. The amount of change detected by an instrument will therefore depend on the efficacy of the intervention used during psychometric testing and this may not reflect all the situations in which the instrument will ultimately be deployed.

- Reliability

Reliability concerns whether the measurement is reproducible and therefore dependable (Jenkinson and McGee 1998). The reliability of instruments can be determined by repeating administration on different occasions when the subjects have not changed with respect to the construct being measured (test-retest). The reliability of responses from different people can be compared for the same subject, for instance between the child and their parents, teachers or a clinician (inter-observer). These forms of reliability are usually presented as the percentage of raw agreement or calculated statistically as chance-corrected agreement using either the kappa statistic for nominal data (Cohen 1960) or weighted kappa statistic for ordinal or continuous data (Cohen 1968). The Intraclass Correlation Coefficient (ICC) is equivalent to a weighted kappa when calculated using quadratic weights (Fleiss and Cohen 1973). Recommended interpretations of the kappa statistics have been proposed as poor when lower than 0.4, fair to good between 0.41 and 0.8 and almost perfect above this value (Landis and Koch 1977). Reliability coefficients are directly related to the variability in the population in which they are used (Streiner and Norman 2003), however coefficients exceeding 0.7 are commonly regarded as reliable for population-based research and coefficients exceeding 0.9 are regarded as reliable for use clinically with individuals (Fitzpatrick et al. 1998).

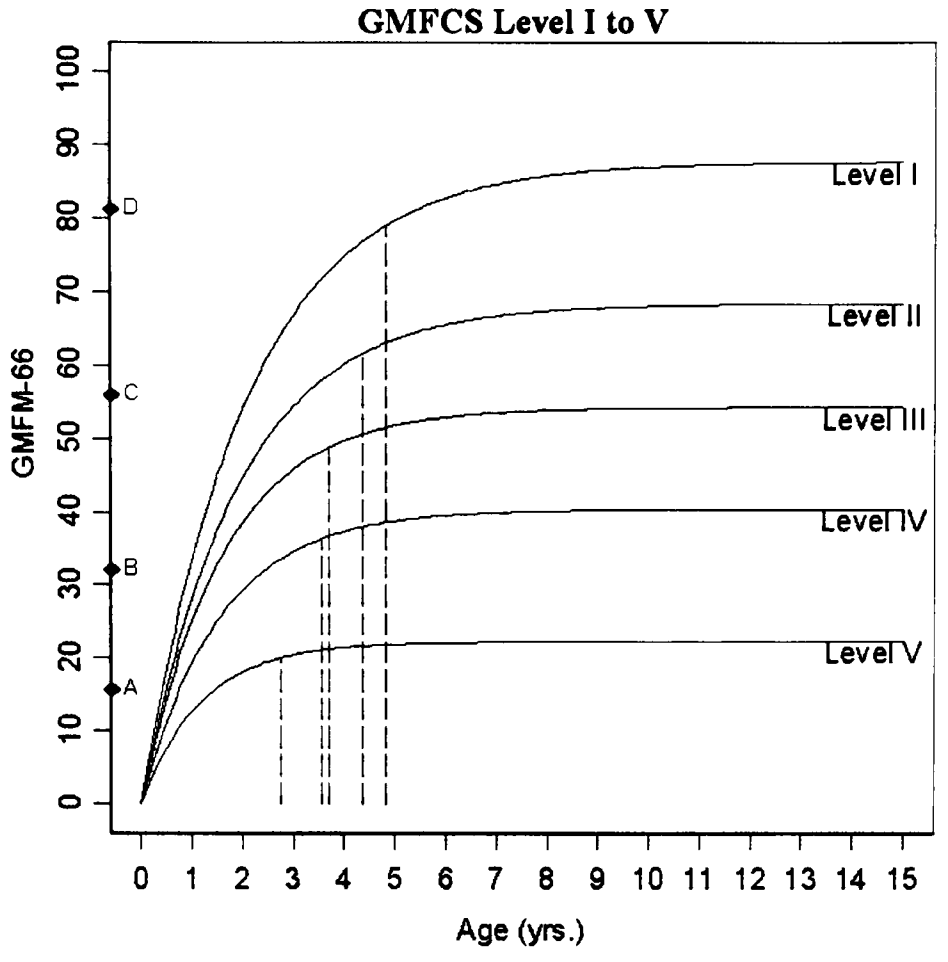
2.2.3 Development of the Gross Motor Function Classification System (GMFCS)

Developed as a professionally assessed system for classifying the severity of motor disability in cerebral palsy, the Gross Motor Function Classification System (GMFCS) has undergone extensive testing of its validity and reliability. The content validity of the GMFCS was originally established through a nominal group process with practicing therapists in Ontario, Canada, and then subsequently using Delphi survey consensus methods with a panel of 20

international experts (Palisano et al. 1997). Further evidence for the construct validity of the system was obtained with a correlation between GMFCS Level and Gross Motor Function Measure (GMFM) score of r = -0.91 (Palisano et al. 2000). The GMFM is generally considered the gold standard criterion test of motor function involving the child in attempting various movement tasks of differing difficulty (Russell et al. 1989 and 2002).

Inter-observer reliability was originally determined using the kappa statistic (κ), with coefficients of κ=0.75 for 40 children aged 2 to 12 years and κ=0.55 for 37 children under two years (Palisano et al. 1997). Through chart review of clinical status from infancy to adolescence for 85 children, the GMFCS was determined to be stable between less than two and 12 years of age using the generalisability coefficient (G), equivalent to test-retest (G=0.79), and the inter-observer reliability in this instance was high (G=0.93) (Wood and Rosenbaum 2000). Longitudinal motor growth curves were subsequently developed for each of the five levels in the GMFCS based on the participants' GMFM scores in the Ontario Motor Growth Study (Rosenbaum et al. 2002). Over 650 children were assessed every six months using the GMFM and classified using the GMFCS. The graphs shown in Figure 2.2 describe clinically important and statistically significant differences in both the rates and limits of motor development between each level of the GMFCS, providing further evidence of the construct validity of the system.

Figure 2.2 Motor development curves created for each GMFCS Level from the Ontario Motor Growth Study (Rosenbaum et al. 2002).



The dashed vertical lines show the mean age at which children in each GMFCS Level achieve 90% of their motor potential as measured by the GMFM; for example, on average, children in Level III reach 90% of their motor potential before their fourth birthday. Whilst the curves plateau after 6 years of age it is not yet clear whether children's motor ability remains constant throughout adolescence and adulthood or deteriorates for some children as is often postulated anecdotally. The flattening out of the curves reflects only that the rate of improvement in children's motor ability, as measured by the GMFM, decreases and stabilises. The curves provide no information about any potential changes in the quality of movement or changes in the amount of energy required to achieve movement. These aspects may continue to develop and improve but were not assessed in the motor curve study (P. Rosenbaum, personal communication).

Reports describing evaluations of the measurement properties of the GMFCS have also been conducted by groups other than the original developers. This represents an important second phase of the development of new health status measures in which translation and replication of acceptable reliability studies consolidate the credibility and utility of new measurement scales (McDowell and Jenkinson 1996). For example, high inter-observer reliability (κ=0.83) has been reported from a group in Sweden after they translated the classification system to Swedish and tested the ability of 12 trained physical therapists to classify children (Lundkvist et al. 1999). Similarly, when the GMFCS was translated into Dutch, physicians were able to classify six children from video recordings reliably with or without a period of training (Gorter et al. 2002b). Slightly lower reliability was observed by a group in Japan classifying 225 children (κ=0.66), however the Japanese authors identified that some words were unfamiliar to clinicians after translation (Kondo et al. 2003).

As a mark of its status, correlations with the GMFCS have subsequently been used to test the validity of other measurement scales such as assessing spasticity (Nordmark and Andersson, 2002) and parents' perceptions of mobility (Thomason et al. 2002). The GMFCS Level at age 2 to 4 years has been used in predictive validity studies of neonatal assessments such as health status 'scores' (Frisone et al. 2002; Haataja et al. 2001), specific MRI and EEG findings (Paciorkowski et al. 2002) and infant movement patterns (Ferrari et al. 2002). The GMFCS has been reported to provide more meaningful classification of children than the traditional method of classifying by distribution of involvement (Gorter et al. 2002; Kembhavi et al. 2002; Kennes et al. 2002).

In summary, the GMFCS has demonstrated sound validity and excellent reliability between assessments made by health professionals for children with cerebral palsy between 2 and 12

years of age. The reliability of rating children less than 2 years of age has not yet been demonstrated satisfactorily and the transition of adolescents passing into adulthood has yet to be systematically described, however both of these issues are currently being studied. One further aspect that had not been investigated is the reliability of families assessing their children's movement ability.

2.2.4 Measuring the Health of Children with Cerebral Palsy

A review of the appropriateness of existing health status instruments for children with cerebral palsy recommended using the Gross Motor Function Measure (GMFM) or Paediatric Evaluation of Disability Inventory (PEDI) to assess children's functional abilities (Ketelaar et al. 1998). These instruments usually require individual administration by a professional. The utility of the GMFM was shown in the Ontario Motor Growth Study to plot children's motor ability with their age (Rosenbaum et al 2002), and the PEDI domains for self-care and social function were found to be particularly associated with GMFCS Levels (Ostensjo et al. 2003).

The LIFE-H is a more recently developed instrument for measuring children's involvement in life situations, analogous to activities and participation (Fougeyrollas et al. 1998). In studies using the LIFE-H, children with cerebral palsy experienced most disruption to situations involving recreation, community, personal care, education, mobility, housing and nutrition (Lepage et al. 1998a&b). Increasing disruption to life habits was most associated with severity of cerebral palsy. However the means by which 'severity' was assessed was of unknown reliability as it was a composite of type and distribution of impairment and a rating of mild, moderate or severe. Type of cerebral palsy, severity, communication and comprehension disorders were associated with greater disruption to life habits in their

subsequent model (Lepage et al. 1998a & b). The LIFE-H is currently being administered by trained interviewers as part of a large European study examining factors affecting the participation and quality of life of children with cerebral palsy across several countries (SPARCLE 2005).

In a structured review of generic instruments for population based research the Child Health Questionnaire (CHQ) was recommended to be the most rigorously developed quality of life measure for children (Schmidt et al. 2002). The CHQ has been used in surveys with families of children with cerebral palsy in Australia (Wake et al. 2003) and North America (Liptak et al. 2001; Vargus-Adams 2005). Consistently in these studies, children with cerebral palsy scored significantly below the mean in all CHQ domains except behaviour and mental health. There were however some differences between studies in general and mental health, pain, self-esteem and the psychosocial summary score. These differences may be attributable to real varying characteristics of children in each study or possibly the different versions of the CHQ questionnaire which were used. Liptak's team targeted only children in GMFCS Levels III, IV and V and administered the 25 item parent report form of the CHQ during an interview; whereas Wake's group administered the 50 item parent form to any families of children attending routine clinics and permitted families to complete the questionnaire at home. The severity of children's motor disability, recorded by GMFCS Level, was significantly associated with physical health scores. However, overall, whilst these studies suggest that the CHQ is able to discriminate between the health status of impaired and unimpaired children, the CHQ may not discriminate between children with different manifestations of cerebral palsy.

The Lifestyle Assessment Questionnaire (LAQ-CP) was developed as a discriminative family-assessed measure of the 'impact of disability' on children with cerebral palsy and their families (Mackie et al. 1998a&b). Summary scores are standardised between 0 and 100 where high scores indicate greater disruption to child and family life. Researchers at the North of England Collaborative Cerebral Palsy Survey (NECCPS) routinely administer the LAQ-CP questionnaire to families of children reported to the survey. The NECCPS team suggest comparing prevalence rates for cerebral palsy using children with LAQ-CP scores above 30% as these were likely to be the 'more reliably ascertained cases' (Hutton et al. 2000). Variation in LAQ-CP scores between districts, after adjusting for the characteristics of cerebral palsy, suggest that the impact of disability is modified by the geographical district where children live confirming the ICF model of interaction between impairment and environment (Hammal et al. 2004).

Parents of children with cerebral palsy were invited to complete the Health Utilities Index (HUI-3) as part of the Ontario Motor Growth Study (Kennes et al. 2002). This study demonstrated that the GMFCS Levels were statistically significantly associated with the mobility, dexterity, speech, and vision domains of the HUI-3, and to a lesser extent hearing and cognition; emotion and pain domains were not associated with GMFCS Level. However, whilst there were statistical associations between these components of health, the authors point out that, apart from the mobility domain, the GMFCS should not be assumed to be a proxy for the presence of other impairments as many of the associations are only moderate. All the instruments described thus far, perhaps apart from the LAQ-CP, have generally either been professionally-assessed measures of physical functioning or family-assessed measures of quality of life. There have, however, been relatively few attempts to use family assessment to measure children's activities and participation.

2.3 CHAPTER SUMMARY

The ICF has resulted from the paradigm shift in the health care agenda for people with chronic medical conditions, from solely recording and treating impairments towards examining to what extent those impairments affect people's everyday lives. The structure of the ICF is hierarchical with functioning in 'body functions and structures' and 'activities and participation' recorded separately. The main chapters are then divided into sub-levels to facilitate coding of functioning in specific body structures or the particular activities listed as appropriate. The mediating role of both personal and environmental factors is also emphasised. The publication of the ICF Version for Children and Youth has enhanced the utility of the system still further as a framework for childhood disability research. To date, researchers have largely used professionally-assessed instruments to gather information about the activities and participation of children with disabilities, and these have matched favourably with ICF codes. In the same period family assessment has mainly been used to measure children's quality of life, however, there is no reason why families could not also objectively assess children's activities and participation.

The key knowledge gaps identified in this chapter were the need to identify existing family-assessed instruments to measure children's activities and participation and also to determine the feasibility of using family assessment to classify children's movement ability using the GMFCS. This preparatory work was necessary to enable a subsequent survey to be conducted with families of children with cerebral palsy.

CHAPTER 3

STUDY AIMS AND OBJECTIVES

3.1 RESEARCH OPPORTUNITIES

A number of knowledge gaps have been identified in the two introductory chapters. Details regarding the prevalence of cerebral palsy and possible causal mechanisms have for many years been evaluated by researchers using epidemiological registers and more recently with advances in medical imaging techniques. However the WHO ICF model indicates that much of our knowledge of cerebral palsy relates to the body functions and structures domain and substantially less is known about these children's activities and participation. The research described in this thesis aimed to increase our understanding of measuring the activities and participation of children with cerebral palsy by gathering information using family-assessed measures. Two key issues were (i) recruiting a representative sample of children, and (ii) having valid and reliable instruments that could be administered expediently.

3.1.1 Working with epidemiological databases

Epidemiological databases such as *4Child* Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children present an opportunity to conduct research within a complete geographically defined population rather than with the samples recruited from clinics or referral centres. Clinic-based samples may not be representative of the population if children not receiving treatment are excluded and inconsistent referral patterns may distort the characteristics children available for recruitment. Another advantage of using the *4Child* database was the detailed information held about children's cerebral palsy and any associated impairments such as epilepsy, hearing and vision loss. Three of the four counties under surveillance by *4Child* now correspond to a single UK strategic health authority, the Thames Valley, in the current administrative structure of the NHS. Therefore a study providing information on children's health would also be useful for needs assessment in that geographical area. In collaboration with *4Child* staff, a cross-sectional survey was therefore

planned with families of children with cerebral palsy born to mothers resident in the Thames Valley at the time of their delivery.

3.1.2 Family assessment of the GMFCS

The researcher had previously collaborated with *4Child*, which was then titled the Oxford Register of Early Childhood Impairments (ORECI), as part of his Master's study examining prescription variations for children with cerebral palsy (Morris et al. 2002). One of the main limitations of that study was that there were no good valid and reliable indicators available of children's functional ability to ensure the children being compared were in fact similar. The GMFCS has since become established as the benchmark method for classifying the movement ability of children with cerebral palsy and recording the GMFCS is now part of *4Child's* routine procedures. However there was no clear mechanism for classifying the hundreds of children who had already been recorded on the database, which established in 1984. One possible way would be to survey the families but no evidence of the reliability of ratings made by family assessment was available. Therefore examining the reliability of family report for the GMFCS became a novel and substantive part of this thesis. The team at the *CanChild* Centre for Childhood Disability Research were responsible for the development of the GMFCS and generally have an outstanding reputation for research with children with disabilities. The researcher initially made contact with Professor Rosenbaum, a co-director of *CanChild*, following his Master's study in 2000 and established an ongoing collaboration. There were no plans at that time for any study organised by *CanChild* to examine the reliability of classification for the GMFCS by families and there was therefore support from the *CanChild* group for this work to proceed.

3.1.3 Family assessment of children's activities and participation

There is a small body of literature examining children's activities and participation. To date, research evaluating the health of children with cerebral palsy has either used professionally-assessed instruments to objectively measure children's physical functional performance, or assessed families' subjective perceptions of children's quality of life. However as objectivity in assessment is defined by the concept being measured (McDowell and Newell 1996), and the concepts of health defined in the ICF are observable objective phenomena, as discussed earlier there is no reason why children's 'activities and participation' could not be reported by families. Family assessment would be more expedient and less expensive than using a professional to examine each child or to interview each family but risks being unreliable or invalid without careful selection and appraisal of the instruments. Preparatory work would therefore require critical appraisal of the existing literature to identify whether any existing family-assessed instruments were appropriate for measuring children's activities and participation.

As described at the end of *Chapter 2*, the Lifestyle Assessment Questionnaire (LAQ-CP) was developed specifically to assess the impact of disability on the lives of children with cerebral palsy and their families (Mackie et al. 1998a) and is used routinely with families of children reported to the North of England Collaborative Cerebral Palsy Survey (NECCPS). Professor Jarvis, who was primarily responsible for the development of the LAQ-CP, and Dr Colver, who is the co-ordinator for NECCPS, were both based at the University of Newcastle and receptive to the researcher's approach to collaborate with them.

3.1.4 Collaborating with families

The families of children with cerebral palsy were ultimately the key people whose collaboration would make the survey a success or failure. It was therefore important to engage a representative and sufficiently large number of families to take part in the survey so that the findings would be generalisable. There were indications from a range of sources that families were willing to support research that, whilst not necessarily altering their own circumstances, increases our knowledge of how to improve the lives of children with disabilities and their families. The work of *CanChild* is testament to this fact, with the families of over 650 children involved in the Ontario Motor Growth Study (Rosenbaum et al. 2002). Likewise, the programme of work by the Children and Families Team led by Professor Sloper at the Social Policy Research Unit at the University of York has shown that families of children with disabilities are prepared to participate in a wide range of research studies (Social Policy Research Unit, 2004). More specific to cerebral palsy, a qualitative study in the north of England conducted with families recruited from the NECCPS showed that families wanted to be more involved in related research, and promoted collaboration with families suggesting that the activity would empower them (Miller et al. 2003).

3.1.5 Collaborating with the research community

In order to ensure the research was conducted to the highest standards, and would be credible and useful, a number of associations were fostered amongst the research community. The principal collaboration was with the National Perinatal Epidemiology Unit, where the researcher was based, and the *CanChild* Centre for Childhood Disability Research. Both of these research units have considerable expertise in conducting large surveys and provided stimulating learning environments. Networking with others working in the same sphere of study was generally successful and fruitful and several opportunistic contacts were made

through email correspondence and at scientific meetings. For instance, once the study in the Thames Valley was underway, the researcher became aware of the development of the Manual Ability Classification System (MACS) to describe children's abilities when handling objects; this is described in more detail in *Chapter 5*. The opportunity for the researcher to collaborate with the developers of the MACS was precipitated by opportunistic contact with Dr Krumlinde Sundholm, and Dr Eliasson. There were also several meetings with the team at the University of Newcastle to share ideas and research methods. These and other exchanges greatly enhanced the quality of the research described in this thesis.

3.2 RESEARCH QUESTIONS, AIMS AND OBJECTIVES

The overall aim of this thesis was to describe the activities and participation of children with cerebral palsy, as defined in the ICF for Children and Youth using family-assessed measures. This research would enable the following questions to be addressed:

1. Can families provide reliable assessments of children's abilities using existing valid and reliable professionally-assessed classification systems?

2. What is the effect of disability on the activities and participation of children with cerebral palsy, as defined in the ICF?

3. Do we have adequate family-assessed instruments to measure activities and participation for children with cerebral palsy, as defined in the ICF Version for Children and Youth?

To answer the research questions the following specific aims and objectives were identified.

Aim 1: To conduct a survey with a geographically defined population of families of children with cerebral palsy

- To collaborate with researchers at *4Child* Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children and the National Epidemiology Unit (NPEU) at the University of Oxford, the host organisation for the database.
- To seek permission from the *4Child* Advisory Group and obtain the appropriate ethics approval to conduct a survey with families of children recorded on the database.
- To identify the study population from the *4Child* database and trace the families' current whereabouts and address.
- To recruit and engage a representative sample of families in the goals of the study and gather data about children's activities and participation.

Aim 2: To determine the reliability of family report for the GMFCS and MACS

- To collaborate with researchers at *CanChild* Centre for Childhood Disability Research where the GMFCS was developed, and subsequently with the developers of the MACS.
- To gather classifications of children's movement and manual abilities from both families and health professionals using the GMFCS and MACS.
- To evaluate the agreement and reliability of family compared to professional assessment using the GMFCS and MACS.

Aim 3: To measure and describe children's activities and participation

- To identify valid and reliable family-assessed instruments appropriate for measuring children's activities and participation.
- To seek permission from the developers of selected instruments to use these in the survey.

- To administer the selected instruments in a postal survey with families and gather data for descriptive and statistical analyses.

Aim 4: To examine the effect of disability on the children and their families

- To identify children's type and distribution of cerebral palsy and any other associated impairments from the *4Child* database.
- To explore associations between typical constellations of children's disabilities and their subsequent activities and participation as defined in the ICF.
- To determine how well impairments predict children's activity limitations and participation restrictions.

The next chapter describes preparatory work undertaken before the main survey to identify the family-assessed instruments and explore the reliability of family report for the GMFCS.

CHAPTER 4

PREPARATORY WORK

4.1 CHAPTER OVERVIEW

This chapter describes the preliminary research undertaken in preparation for the main study. The main study was a postal survey with families of children with cerebral palsy in the UK, who were between 6 and 12 years old. Therefore one of the objectives was to find the most suitable family-assessed instruments for measuring children's activities and participation. The first part of this chapter describes a structured review which was conducted to identify family assessed instruments appropriate for measuring children's activities and participation as defined by the ICF Version for Children and Youth. The instruments were identified from a systematic search and then appraised and selected using predefined criteria.

The second aspect of the preparatory work concerned the fact that the reliability of family's classification of children's movement ability using the Gross Motor Function Classification System (GMFCS) was not known. Therefore, the second part of this chapter describes a preliminary study to assess the reliability of families reporting their children's movement abilities using the GMFCS. This work was undertaken in collaboration with Professor Rosenbaum whilst the researcher was a visiting student at the *CanChild* Centre for Childhood Disability Research at McMaster University in Canada.

The aim of these two preparatory studies was to ensure that the methods proposed for gathering information about children's GMFCS in the main Thames Valley survey were likely to be appropriate, valid, reliable, precise, interpretable and acceptable.

4.2 FAMILY-ASSESSED MEASURES OF CHILDREN'S ACTIVITIES AND PARTICIPATION: A STRUCTURED REVIEW

4.2.1 Aim

The aim of this first piece of preparatory work was to identify family assessed instruments appropriate for measuring children's activities and participation as defined by the ICF Version for Children and Youth.

4..2.2 Methods

A systematic search of the literature was undertaken using expressions describing 'activities and participation' with reference to the thesaurus terms commonly used in bibliographic databases. Words that have variable endings were truncated with an asterisk; words that may have been connected with or without hyphens were typed using a question mark, such as 'self?report'. Searches were undertaken combining these expressions with the term 'children' and other variants and synonymous terms. The results were filtered using terms related to measurement: scale and questionnaire; and results were limited to English language publications. Terms describing cerebral palsy were then entered to identify measures that had previously been used and reported in this population. The search strategy is shown in Figure 4.1 on the next page. The reference lists of reviews and original articles identified were also searched. The following databases were searched from their year of inception: Medline, EMBASE, CINAHL, AMED and PsycINFO (all WinSPIRS 4.0 Silver Platter), Sociofile (Cambridge Scientific Abstracts) and the PAHOP database of Patient-assessed Health Instruments (National Centre for Health Outcomes Development, 2003). The search was completed in August 2004.

Table 4.1 Search terms and strategy used to identify family-assessed instruments suitable for measuring children's activities and participation.

1. CHILD* OR ADOLESCEN* OR KID*
2. PHYSICAL FUNCTION* OR FUNCTIONAL STATUS OR HEALTH STATUS
3. ABILIT* OR ACTIVIT* OR HANDICAP* OR PARTICIPATION OR DISABILITY*
4. MEASURE* OR SCALE* OR INSTRUMENT* OR INDEX* OR QUESTIONNAIRE*
5. SURVEY OR SELF?REPORT* OR CHILD?REPORT* OR PROXY?REPORT* OR PARENT?REPORT* OR FAMILY?REPORT*
6. SELF?ASSESS* OR CHILD?ASSESS* OR PROXY?ASSESS* OR PARENT?ASSESS* OR FAMILY?ASSESS*
7. #2 OR #3
8. #4 OR #5 OR #6
9. #7 AND #8
10. #9 AND #1(LIMITED to English language)
11. CEREBRAL PALSY OR CP OR HEMIPLEGI* OR DIPLEGI* OR QUADRIPLEGI* OR NEURODISABILIT*
12. #10 AND #11

Inclusion criteria

In order to be included in the review instruments had to (i) be an objective discriminative measure of activities, physical functional performance or participation; (ii) be suitable for use with children aged between 6-12 years; (iii) have evidence of sound measurement properties; (iv) be suitable for family assessment; and (v) be published in the English language.

Exclusion criteria

Instruments were excluded if they predominately assessed the child or family's subjective perception of the child's 'quality of life', or the instrument required administration by a health professional, trained assessor, or could not otherwise be administered by mail.

Assessment criteria

The criteria used to assess and select patient completed instruments included their appropriateness for the task and an assessment of their validity, reliability, responsiveness, precision, interpretability, acceptability, and feasibility (Fitzpatrick et al. 1998).

- Appropriateness

Appropriateness was assessed with reference to the domains identified in the ICF Version for Children and Youth (WHO 2002), which is in essence an assessment of content validity.

- Validity

A clear conceptual framework underpinning the instrument should be provided with evidence that the measure 'behaves' as expected based on the conceptual framework by which it was created (construct validity); and that resulting measurements correlate well with instruments measuring the same construct (convergent validity), poorly with instruments measuring something unrelated (divergent validity), and differentiate between subjects with known differences (discriminative validity) (Jenkinson and McGee 1998).

- Reliability

The reliability of instruments can be determined by repeating administration on different occasions when the subjects have not changed (test-retest). The reliability of responses from

different people can also be compared for the same subject, for instance between the child and their parents, teachers or a clinician (inter-observer), or between the same subject on different occasions (intra-observer). The internal consistency of unidimensional measurement scales should be reported and, when appropriate, details to support the allocation of items to the subscales of multidimensional instruments.

- Responsiveness

Instruments are generally designed to fulfil one or more functions: to discriminate, predict current or future status, or evaluate change over time. Evaluative instruments aim to measure real change within individuals, when it occurs, from one time to another; responsiveness describes the sensitivity of those instruments to detect that change. Responsiveness is not a property required of discriminative instruments and is not useful in cross sectional surveys.

- Precision

Precision is concerned with the number and accuracy of distinctions made by an instrument. In this review instruments were compared by the number of response categories offered to answer each question, the precision of the numerical values attached to each response, how well the possible responses to each item are distributed over a true range, the potential for floor or ceiling effects, and the precision of the overall scales.

- Interpretability

Interpretability is concerned with how meaningful the scores derived from an instrument are, and therefore assesses what information individual or group scores actually convey.

- Acceptability

Questionnaires must be acceptable to potential respondents and users. Factors such as the number of items and the time necessary to complete the questionnaire influence acceptability, while content might be especially important to potential users.

- Feasibility

Feasibility concerns whether, from the researchers' perspective, the instrument is easy to administer and process.

Data extraction

Information about the instruments selected for the review was extracted by the researcher into tables that compared instrument characteristics using the predefined assessment criteria.

4.2.3 RESULTS

In total, 34 instruments were identified from the databases and reference lists of seven review articles. Of these, five instruments were excluded because they focused only on adolescents, four were excluded because they required administration by a professional and eighteen were excluded because their inquiry focused entirely on subjective perception of their children's health, satisfaction with health state or health care. Seven child or family-assessed health instruments were identified that could potentially be administered by mail to measure aspects of children's activity performance and participation. The seven instruments included for further appraisal in the review are listed on the next page:

- Assessment of Life Habits for Children (LIFE-H) (Fougeyrollas et al. 1998)
- Lifestyle Assessment Questionnaire (LAQ-CP) (Mackie et al. 1998a&b)
- Activities Scale for Kids (ASK) (Young et al. 2000)
- Child Health Questionnaire (CHQ) (Landgraf et al. 1998)
- Quality of Life Inventory (PedsQL) (Varni et al. 2001)
- Child Health and Illness Profile-Child Edition (CHIP-CE) (Riley et al. 2004)
- Health Utilities Index (HUI 3) (Feeny et al. 1996).

Appropriateness

A summary of the characteristics of the seven instruments included in the review is shown in Table 4.2. Five instruments are generic, one is designed for children with physical disabilities (PedsQL) and one is specific to families of children with cerebral palsy (LAQ-CP). One instrument is a measure of how often children perform specific activities (ASK), two assess aspects of participation in life situations (LIFE-H, LAQ-CP), one is a health status utility measure (HUI-3) and three instruments are described as quality of life measures. Only two of the instruments (CHIP-CE and PedsQL) have not been reported as having been used with children with cerebral palsy. The content and domains covered by each instrument vary and, despite three having the primary purpose of measuring children's quality of life, all include some content relevant to children's activities and participation. The domains of the ICF Version for Children and Youth 'activities and participation' described, at least partially, by each of the reviewed instruments, are shown in Table 4.2.

Table 4.2 Description of the instruments included in the structured review.

Instrument	Population	Age range	Purpose of measure	Content / Dimensions	Used for CP
LIFE-H	Generic children's version derived from original adult instrument.	5-13 years & adult	Activities & participation + Quality of life	Nutrition, Fitness, Personal Care, Communication, Housing, Mobility, Responsibilities, Interpersonal Relationships, Community Life, Education, and Recreation.	*Yes*
LAQ-CP	Condition-specific for children with cerebral palsy and their families.	3-10 years	Activities & participation	Mobility, Physical Independence, Clinical And Economic Burden, Schooling, and Social Integration.	*Yes*
ASK	Generic – initially developed with children with musculoskeletal disorders.	5-15 years	Activities & participation	Physical functioning - personal care, dressing, eating & drinking, mobility, play, stairs, standing skills.	*Yes*
CHQ	Generic for children.	5-15 years	Quality of life	Physical functioning, role/social - physical, emotional & behavioural limitations, general health perceptions, bodily pain, parental impact time & emotional, self esteem, mental health, general behaviour, family activities & cohesion, change in health.	*Yes*
CHIP-CE	Generic for children.	6-11 years & CHIP-AE up to 18 years	Quality of life	Satisfaction (with self and health), Comfort (emotional and physical symptoms and limitations), Resilience (positive activities that promote health), Risk Avoidance (risky behaviours that influence future health), and Achievement (of social expectations in school and with peers).	*No*
PedsQL	Children with chronic health conditions.	2-18 years	Quality of life	Physical, Emotional, Social and School.	*No*
HUI-3	Generic for children & adults.	Not defined	Preference-based health utility	Mobility, Dexterity, Speech, Vision, Hearing, Cognition, Emotion and Pain.	*Yes*

Table 4.3 Appropriateness of instruments for measuring major domains of Activities & Participation as defined by WHO ICF Version for Children and Youth.

	Learning & applying knowledge	General tasks & demands	Communication	Mobility	Self-care	Domestic life	Interpersonal interactions & relationships	Major life areas - Education & work	Community, social & civic life
LIFE-H	-	*Yes*	*yes*	*yes*	*yes*	*yes*	*yes*	*yes*	*yes*
LAQ-CP	-	*Yes*	-	*yes*	*yes*	*yes*	*yes*	*yes*	-
ASK	-	*Yes*	*yes*	*yes*	*yes*	*yes*	-	*yes*	-
CHQ	-	-	-	*yes*	-	-	*yes*	*yes*	-
CHIP-CE	-	-	-	*yes*	-	-	*yes*	*yes*	-
PedsQL	-	-	-	*yes*	-	-	-	*yes*	*yes*
HUI-3	*Yes*	-	-	*yes*	-	-	-	-	-

Validity

Some issues of face and content validity have already been identified where each instrument was assessed for its appropriateness to measure activity performance and participation as described by the ICF. A summary of some of the other aspects of validity that have been demonstrated to support each instrument is shown in Table 4.4. Moderate to high levels of convergent validity with other relevant measures has demonstrated for all the instruments. Evidence of divergent validity has only been presented for the ASK and PedsQL. There is, however, evidence that all the instruments can discriminate between children with different severities of disability.

Table 4.4 Evidence for the validity of instruments included in the review.

Instrument	Convergent validity	Divergent validity	Discriminative validity
LIFE-H	Moderate correlations with dimensions from other measures of handicap in adult populations – Spinal Cord Injury and Acquired Brain Injury.	-	Disruption in life habits increased with severity of impairment and disability.
LAQ-CP	Good correlation with scores from Central Motor Deficit Form ($r = 0.76$, $p<0.0001$)	-	-
ASK	Highly correlated with CHAQ* (0.82) & mobility indicator from HUI-3 (0.74) & moderate correlation with CHQ physical function.	Poor correlation with emotion (0.15) & speech (0.09) indicators from HUI-3	Distinguished between 3 level global ratings of severity by clinicians.
CHQ	Small but significant correlations for behaviour, anxiety, depression.	-	Distinguished between healthy children and children with chronic illnesses.
CHIP-CE	Reasonable correlation with CHQ satisfaction with health and physical functioning dimensions.	-	Distinguished between healthy children and those identified by parents as having a major chronic illness.
PedsQL	Psychological and social functioning associated with emotional distress, perceived competence and social functioning; physical functioning associated with days missed from school.	Physical functioning not associated with emotional distress, perceived competence and social functioning.	Distinguished between healthy children and children with chronic illnesses.
HUI-3	Moderate correlation with CHQ pain, physical function – mobility & mental health – emotion.	-	Distinguished between healthy children and children with chronic illness & also severity of disability in children with cerebral palsy.

* CHAQ = Child Health Assessment Questionnaire is a condition-specific measure for Juvenile Rheumatoid Arthritis

Reliability

There are acceptable reports of test-retest reliability for all the measures except the PedsQL (Table 4.5). There is reasonable evidence to support inter-observer reliability for the LIFE-H. The reliability between children's and parents' responses has been demonstrated for the ASK. There is evidence to support the internal consistency of the various domains in the three quality of life measures (CHQ, CHIP-CE, PedsQL) and the LIFE-H, and also the unidimensional scale of the ASK. No evidence was found relating to the internal consistency of the domains in the LAQ-CP. Internal consistency is not applicable to the HUI-3 as this measure only includes single summary items for each domain. However, the preferences gathered and used to devise the weightings for the HUI-2 were replicated almost exactly in a subsequent survey in a separate population (Feeny et al. 1996).

Table 4.5 Evidence of the reliability of instruments included in the review.

Instrument	**Test-retest / Inter-observer** (ICC)	**Internal consistency** (Cronbach's α for each domain)
LIFE-H	Same assessor: from 0.64 to 0.96. Different assessors: from 0.62 to 0.91.	Range from 0.73 to <0.90, except interpersonal relationships 0.40.
LAQ-CP	Correlation between summary scores r=0.97, p<0.0001; ANOVA showed large and stable differences between individuals.	
ASK	Child test-retest = 0.97 Child and parent agreement = 0.96	Unidimensional scale = 0.99
CHQ	Test-retest 0.54 to 0.73.	Child from 0.63 to 0.90 Parent from 0.59 to 0.94
CHIP-CE	Child version from 0.63 to 0.76 Parent version from 0.71 to 0.85	Child version from 0.64 to 0.82 Parent version from 0.79 to 0.88
PedsQL	-	Child from 0.71 to 0.88 Proxy from 0.75 to 0.90
HUI-3	From 2 telephone interviews 1 month apart = 0.77	Not applicable

Precision

The number of response categories in instruments varied from binary options to six ordinal levels. The ASK and CHIP-CE are constructed with consistent 5 level response options to each item throughout the questionnaire. Similarly the LIFE-H offers 5 levels of accomplishment combined with 4 levels of assistance for every item. The PedsQL mainly uses 5 levels but the child self report version has only 3 response levels. The HUI-3 offers either 5 or 6 and the CHQ between 4 and 6 options. The LAQ-CP has the most variable response options including binary, 3 and 5 levels depending of the item and even some open questions requiring free text that has to be coded. However, all responses to the LAQ-CP are subsequently coded to scores ranging from 0 to 4 for each item. The same scoring range of between 0 and 4 is used by other instruments except the CHQ, which scores some items from 0 to 5 or 6, the LIFE-H which uses a range from 0 to 9, and the HUI-3 which uses single utility scores derived by a mathematical function based on the preferences of a reference population.

Whether the responses offered truly reflect the range of children's abilities is difficult to assess from published data. As the HUI-3 scores are based on a reference population these are more likely to cover the whole range of possible responses without floor or ceiling effects. It is not possible to gauge from existing publications whether the other instruments are prone to floor or ceiling effects. In the development of the ASK children with the most severe cerebral palsy who could not self-report were excluded; there is therefore a potential for a floor effect if the ASK is used with those children. The CHQ has been used previously with children with cerebral palsy; scores in all domains were consistently below the population mean except for behaviour and mental health.

Table 4.6 Precision of instruments included in the review.

Instrument	Number of response categories	Precision of numerical values	Distribution of items over true range	Floor ceiling effects	Precision of scales
LIFE-H	5 ordinal levels of accomplishment + 4 ordinal levels of assistance	Item scores 0-9 transformed to percentages for each domain	Probably	None apparent	Not known
LAQ-CP	Variable – Binary & 3 and 5 level ordinal levels and free text.	Item scores 0-4 transformed to percentages for domains (0-10) and weights used to calculate summary score (0-100).	Probably	None apparent	Not known
ASK	5 levels of how often an activity was performed in the last week.	Item scores 0-4 transformed to percentage.	Probably	Potential floor effect	Rasch model
CHQ	4, 5 & 6 ordinal level options & binary for symptoms	Transformed to percentages & then to z scores used for summary scores compared to population mean.	Probably	Potential floor effects	Not known
CHIP-CE	5 ordinal levels	Item scores 0-4 transformed to standardised scores	Probably	Not known	Not known
PedsQL	Mostly 5 ordinal levels but 3 levels used for young children.	Item scores 0-4 transformed to percentages.	Probably	None apparent	Not known
HUI-3	5 or 6 ordinal levels	Item scores transformed to standardised preference-based utility scores.	Yes	No	Good

The precision of the scale scores produced from ordinal responses is questionable for all instruments except the ASK and the HUI-3 for the following reasons. The developers of the ASK used item response theory to confirm the unidimensional nature of the scale and that there is an order of difficulty underlying items in the scale (Young et al. 2000). HUI-3 scores for different children or populations are meaningful when they are based on the same standardised reference population (Feeny et al. 1996). A summary of the precision of the instruments is provided in Table 4.6.

Interpretability

All the instruments produce useful and meaningful information once the user is familiar with the orientation of the scoring system and the construct being assessed. The LIFE-H and LAQ-CP both produce domain based scores from 0 to 10 where higher scores represent greater disruption to life habits and participation for the child and family. The LAQ-CP also produces an overall weighted summary score from 0 to 100 based on a linear additive model that includes weights from the preferences of a reference group of health professionals and families. For ease of interpretation typical participation profiles are described for children with scores of 30, 50 or 70. The developers of the LAQ-CP have suggested that children with scores exceeding 30 are more reliably ascertained for population registries, as children with lower scores may not be detected by health or social services (Hutton et al. 2000).

Percentage scores (0 to 100) are calculated for the ASK, CHQ, CHIP-CE and PedsQL instruments where, in these instances, higher scores represent better health or physical functioning and activity performance. The HUI-3 produces 8 attribute preference-based ability scores and an overall summary score ranging between 0 and 1 where a higher score represents better health 'utility', or preferred health state.

Acceptability

The LIFE-H is by far the longest of the seven questionnaires, with 62 items in the short version and 240 in the long version. However, as the LIFE-H has not previously been administered by mail for self completion (L. Noreau, personal communication) the acceptability for the purpose of the proposed study could not be determined. Whether families would understand the instructions and how much time would be necessary for a family to complete the questionnaire are not yet known. The LAQ is not designed for the child to complete. All the instruments except the LAQ-CP can be completed by children or a parent or family carer as a proxy.

The ASK, CHIP-CE and LAQ-CP are all similar in length at between 37 and 45 items, and are reported by their developers to take between 10 and 30 minutes to complete. The CHQ has both parent and child versions and also versions of varying lengths. No information was found to indicate how long the CHQ and PedsQL take to complete. The HUI-3 only has 8 items and would almost certainly be the most expedient. Response rates of 70% have been reported by the developers of the LAQ-CP, ASK, CHQ and HUI-3; these rates have generally been derived from samples recruited in hospital settings who were asked to return the questionnaires by mail.

Feasibility

The instruments were required to be suitable for administration by mail. The LAQ-CP, ASK, CHQ and HUI-3 have all previously been successfully administered by mail. Available evidence suggested that, at the time of the review, the LIFE-H, CHIP-CE and PedsQL had only previously been administered by trained assessors.

All the instruments provide a user guide or manual to assist the researcher with scoring. Data entry software is available for the CHQ and LAQ-CP making the calculation of scores easier. There is small fee charged for the purchase of some manuals, however the HUI is the only instrument in this review for which a substantial fee (several thousand Canadian dollars per use) must be paid to the developers to extract the maximum potential of the measure.

4.2.4 Discussion

Due to the recent development of the concepts of interest in this study, there are relatively few child or family-completed instruments appropriate for measuring children's activities and participation as defined in the child and youth version of the ICF. A previous review of functional health status measures appropriate for children with cerebral palsy recommended the Gross Motor Function Measure (GMFM) and Paediatric Evaluation of Disability Index (PEDI) (Ketelaar et al. 1998). However, these instruments require individual administration by a trained assessor and were therefore ineligible for this current review. Of the other measures excluded, the Pediatric Outcomes Data Collection Instrument (PODCI) created by the Pediatric Orthopaedic Society of North America was closely considered (Daltroy et al. 1998). However, as the primary purpose of the PODCI is to assess orthopaedic intervention, the instrument includes several wholly subjective elements such as treatment expectations and satisfaction with the outcome; the PODCI has also been considered by others to be complex to score (Pencharz et al. 2001).

Of the measures included in this review the LIFE-H provides the most complete coverage of the content of the ICF. This is almost certainly due to the fact that they were both founded from the same Disability Creation Process (Fougeyrollas et al. 1998). The LIFE-H has been used in a population of children with cerebral palsy but has not been previously administered

by mail (Lepage et al. 1998a&b). The reliability of family assessed responses could not therefore be assured and any effect of the length or complexity of the questionnaire on the response rate was therefore unpredictable. The HUI-3 is the shortest questionnaire, has been extensively developed and previously used to describe health status in a population of children with cerebral palsy (Kennes et al. 2002). The HUI-3 might be useful if further statistical analysis was planned that incorporated population preferences for health or economic evaluation. However, despite the statistical robustness of the HUI-3 it has limited coverage of the ICF domains, does not describe what activities children perform and is also extremely expensive.

The potential for generic quality of life instruments to contribute to measuring activities and participation was represented by the three most cited instruments which were retained in the review for this purpose. The Child Health Questionnaire (CHQ), Pediatric Quality of Life Inventory (PedsQL), and the Child Health and Illness Profile-Child Edition (CHIP-CE) are all extensively developed, self proclaimed quality of life instruments. The CHQ and CHIP-CE are generic measures whilst the PedsQL was developed for children with chronic health conditions. All three instruments are multidimensional and some of the constructs included are in fact difficult to distinguish from those encompassed by the ICF. In different ways, each of the instruments sets out to capture details of children's physical functioning, educational performance and involvement with family and peers. The CHQ, CHIP-CE and PedsQL have confirmed elements of their measurement as valid and reliable by comparing children's self reports with proxy responses. The fact remains, though, that despite their excellent psychometric properties the overall purpose of these instruments is to measure subjective quality of life. The instruments therefore include pain, self esteem, symptoms,

and satisfaction domains, which, although important in terms of quality of life, are not commensurate with objectively measuring activities and participation.

The ASK and LAQ-CP both have a broad coverage of most domains of the ICF. The ASK contains several items asking how often children play sports. These are associated with 'recreation and leisure' which is part of the domain 'community social and civic life'. The LAQ-CP was based on the predecessor to the ICF and was initially proposed to measure 'handicap', the antecedent to 'participation restrictions'. The LAQ-CP contains items about child-parent relationships which are considered part of 'interpersonal interactions and relationships'. The LAQ-CP is a parent-assessed instrument and contains some items relevant to the participation of the respondent themselves or family as a whole. This is logical as the participation of the child is in many ways likely to be interconnected with and inseparable from that of their family. The ASK is a child-completed instrument although proxy report by parents is highly reliable. Neither the ASK nor LAQ-CP covers issues listed in the ICF under 'communication' or 'learning and applying knowledge'.

Of note, and not included in this review, is the Children's Assessment of Participation and Enjoyment (CAPE) which is a new instrument for children with disabilities developed by the *CanChild* Centre for Childhood Disability Research (King et al. 2004). Details of its psychometric properties were not available at the time of this review. The content of the CAPE covers recreational, active physical, social, skill-based, and self-improvement / educational activities which are five of the nine dimensions of the ICF. The criterion of responsiveness was not assessed in detail as the design of the study in the Thames Valley was cross-sectional. However, reasonable evidence of responsiveness to important change was only available for the ASK and the three quality of life measures.

4.2.5 Conclusions

There are no existing child- or family-completed instruments specifically for measuring 'activities and participation' as defined in the WHO ICF for Children and Youth. No single instrument was favoured from this structured review of seven potential instruments. The LIFE-H is the most appropriate instrument as assessed by its content but the reliability of child or family self-assessment using the questionnaire is not yet known. The complexity of the rating procedure and large number of items in the unabridged version render the LIFE-H too complex for self-assessment; the developers currently recommend the instrument only be administered by a trained interviewer.

At the time of this review, using the predefined criteria of appropriateness, validity, reliability, precision, interpretability and feasibility, the instruments recommended for mailed surveys with families of children with cerebral palsy are the Activities Scale for Kids (ASK) and the Lifestyle Assessment Questionnaire (LAQ-CP). Both instruments provide a broad description of how frequently children performed some typical activities. The LAQ-CP also measures aspects of participation restrictions imposed on the family unit and hence provides additional contextual information. The instruments included in this review provide most information about children's physical mobility, self-care, general tasks and demands, and domestic life. The selected instruments, the ASK and LAQ-CP, provide notably less information about communication, educational participation and applying knowledge. Therefore there remains much scope for developing valid and reliable instruments corresponding to the ICF dimensions of activities and participation.

4.3 RELIABILITY OF FAMILY ASSESSMENT FOR THE GMFCS: A STUDY IN CANADA

4.3.1 Aim

The aim of this study was to devise and test a new questionnaire for gathering family reports for the GMFCS and to assess the reliability of classifications of the GMFCS made by families in comparison to health professionals' judgments.

4.3.2 Methods

This study was a cross-sectional survey approved by the Hamilton Health Sciences / McMaster University Faculty of Health Sciences Research Ethics Board in Canada undertaken whilst the researcher was a visiting student in the winter 2003. The survey was designed to collect data from families using a short questionnaire which in this case was sent using the regular postal service. Return of the completed questionnaire was taken as evidence of consent.

Study sample

The study population included families of children with cerebral palsy known to the *CanChild* Centre for Childhood Disability Research from the Ontario Motor Growth study (Rosenbaum et al. 2002) (n = 632 at the time of this study). Each of these children had several ratings of their GMFCS level made by a physiotherapist as part of that previous study; only the most recent professional rating was used for this study. The therapists had been provided with the standard user guide instructions for the GMFCS and were asked to make the assessment after observing the child during regular therapeutic activities.

Families participating in concurrent research studies co-ordinated by *CanChild* and families who had requested no further contact or who had already been lost to follow up were excluded. The families of children who were between 6 and 12 years old in February 2003 were specifically selected, to correspond to the current upper age band for the GMFCS (6-12 years). Five families who had specifically asked to receive any future correspondence in French were excluded. Thus the study population for the pilot study were the families of 221 children.

Questionnaire Design

Professionals in the Ontario Motor Growth Study had been provided with the standard GMFCS literature in the form of a three-page fold-out booklet. The booklet describes the abilities of children in each of the GMFCS levels according to the four age bands: less than 2 years, from 2-4 years, 4-6 years and 6-12 years. This booklet was considered unwieldy for families, partly because the text contains an element of jargon but mainly because for this study only the upper age band (6-12 years old) was relevant. Therefore a new questionnaire was produced.

Several ways of designing the questionnaire were considered. The options were (i) a checklist of several key activities by which the parents or carers could rate the child's ability either dichotomously (i.e., can or cannot do) or on a five or seven point ordinal scale (such as how often they achieved some activity); or (ii) a single summary item requesting a global estimation of which of the five levels provided the most accurate description of the child's movement abilities. Although checklists appear to offer a more systematic approach, there is no demonstrable improvement in reliability compared to the global estimation approach (Van der Vleuten et al. 1991). It has been suggested that this is because experts, those who

are very familiar with a subject or person, do not necessarily think in the same stepwise manner upon which checklists are based to make their judgment (Hodges et al. 1999). A parent's competence to provide a reliable global estimate of the child's abilities might therefore be undermined by a series of task oriented questions. Thus an approach was selected using a single question that asked the respondent to indicate which of five provided descriptions best represented their child's movement ability, methodologically corresponding to a Guttman scale (Streiner and Norman 2003).

In a stylistic change from the leaflet distributed to health professionals, the description of children in level V, those least able, was purposefully placed at the top of the page so that families of children with the greatest limitations would not have to read through a list of activities their child could not do before reaching their own child's ability. Also, in order to monitor any effect of families having been informed of the rating made by health professionals, an extra question was included asking if a health professional had ever discussed the GMFCS with the family. The single page questionnaire was copied onto brightly coloured paper to help it stand out from the accompanying letter, and so that it could be more easily noticed if misplaced in the family home. Questionnaires were then individualised with an identifying number unique to each child in the study.

Survey Procedure

The survey was prepared following the published recommendations for maximising response rates from mailed questionnaires (Edwards et al. 2002; McColl et al. 2001; Streiner & Norman 2003). Each invitation letter was personally addressed to the parent or carer and stated the name of the child about whom information was being requested. Each letter displayed the research unit logo in colour and was personally signed by a researcher. Return

envelopes were individually stamped and addressed to the research unit at McMaster University. The letter, questionnaire and return envelope were packaged inside an envelope bearing the last known address for the family and were posted using the regular Canadian postal service. For pragmatic reasons, based on the limited duration of the researcher's time in Canada, families were given a short deadline of three weeks to return questionnaires and there were no reminder letters or attempts to telephone families to increase the response rate.

Data Cleaning and Entry

Upon receipt of the returned questionnaires, as a quality control procedure, data were twice entered into an SPSS database on separate occasions by different people. This was done first by the researcher and then by a fellow postgraduate student. One discrepancy was found between entries that was resolved with reference to the original questionnaire.

Statistical Analysis

Chance-corrected agreement was determined using the kappa statistic (κ). The GMFCS uses an ordinal scale with five levels, one of two indices of reliability could have been used for these ordinal data. However, the weighted kappa and the Intraclass Correlation Coefficient (ICC) have been shown to be equivalent when quadratic weights are used to calculate the kappa statistic (Fleiss and Cohen 1973). The ICC was determined using repeated measures analyses of variance (ANOVA) in SPSS to calculate the mean squares (MS) due to the children's ratings ($MS_{subjects}$) and the mean square error (MS_{error}). The calculation was then performed as shown in the following equation where the symbol σ represents variance.

$$ICC = \frac{\sigma_{subjects}}{\sigma_{subjects} + \sigma_{error}} = \frac{MS_{subjects} - MS_{error}}{MS_{subjects} + MS_{error}}$$

For each ICC reliability coefficient, 95% confidence intervals were determined by hand using Fisher's *Z* transformation (Fisher 1925; Streiner and Norman 2003). An ICC was calculated separately for families who had, and those who had not, discussed their child's GMFCS level with a health professional. Another ICC was calculated for the combined sample. An ICC was also separately calculated for the children whose last professional rating was made when the child was in the lower age band of the GMFCS (4 - 6 years). This provides a measure of the stability of the GMFCS across different age bands, as the family rating was made when the children were between 6 and 12 years old using descriptors relevant to that age group.

4.3.3 Results

From the 221 invitations sent out to families 20 letters were returned by the post office as undeliverable and 97 responses were received. Of the families that could possibly have received invitations there were 97 / 201 questionnaires returned (response rate 48%). Four questionnaires were spoiled, having been marked in a way that a single level could not be interpreted for those children. The mean age of the 93 children for whom a family report of the GMFCS was admissible was 9.4 years (S.D. 1.1 years, range 6 to 11 years), similar to the survey population's mean age which was 9.6 years (S.D. 0.97 years, range 6 to 11 years). Table 4.7 shows a comparison of the known physical characteristics of the children for whom there was an admissible response and the total study population. There were no significant differences apparent in the types or distribution of impairment between respondents and the total eligible study population. Children with all types of cerebral palsy were represented in proportions consistent with general populations, the majority having spastic cerebral palsy. Most of the children were recorded as having leg or four-limb dominant impairments.

Table 4.7 Characteristics of children for whom a response was admissible compared with the total eligible study population

	Responders		Total population	
	n	**%**	**n**	**%**
	93		**201**	
Gender				
Male	53	*57*	118	*59*
Distribution of impairment				
Leg Dominant	35	*38*	69	*34*
Three-Limb Dominant	8	*9*	16	*8*
Four-Limb Dominant	35	*38*	80	*40*
Right Hemiplegic	6	*6*	14	*7*
Left Hemiplegic	6	*6*	15	*7*
Not Given	3	*3*	7	*3*
Type of impairment				
Spastic	67	*72*	148	*74*
Dyskinetic	7	*8*	14	*7*
Ataxic	4	*4*	5	*2*
Mixed / unclassifiable	15	*16*	34	*17*

The type of cerebral palsy (spastic, ataxic, dyskinetic, mixed) and distribution of involvement (number of limbs affected) for the children included in the analysis are shown in Tables 4.8 and 4.9, with respect to the GMFCS Level assessed by a physiotherapist.

Table 4.8 Type of impairment compared with GMFCS Level assessed by a physiotherapist for children included in the analysis

	GMFCS Level					
	I	II	III	IV	V	Total
Spastic	19	7	10	19	12	67
Dyskinetic	2	1	1	-	3	7
Ataxic	1	-	3	-	-	4
Mixed / unclassifiable	2	1	6	2	4	15
Total	24	9	20	21	19	93

Table 4.9: Distribution of impairment compared with GMFCS Level assessed by a physiotherapist for children included in the analysis

	GMFCS Level					
	I	II	III	IV	V	Total
Leg Dominant	11	7	12	5	-	35
Three-Limb Dominant	3	-	0	5	-	8
Four-Limb Dominant	1	2	6	9	17	35
Right Hemiplegia	5	-	1	-	-	6
Left Hemiplegia	4	-	1	1	-	6
Not given	-	-	-	1	2	3
Total	24	9	20	21	19	93

The number of children in each level of the GMFCS, as families and physiotherapists rated them for the 93 admissible responses, is shown in Table 4.10.

Table 4.10: Number of children assessed in each GMFCS Level as classified by families and professionals

		Family-assessed GMFCS Level					
		I	II	III	IV	V	Total
Physiotherapists	I	17	7				24
	II	1	7	1			9
	III		1	14	5		20
	IV			4	14	3	21
	V				2	17	19
	Total	18	15	19	21	20	93

ICC = 0.92 (95% CI 0.90 to 0.93)

The ICC for professionals and families who had discussed their child's GMFCS level with a health professional (n=35) was 0.97 (95% CI from 0.96 to 0.98); for those who had not (n=52) the ICC was 0.92 (95% CI from 0.91 to 0.93); and for the whole sample (n=93) the overall ICC was 0.94 (95% CI from 0.90 to 0.96). The overall chance-corrected agreement was $\kappa = 0.68$. Regarding the stability between ratings made by health professionals for children when they were in the 4-6 year age band and ratings made by families for the same children when they were in the 6-12 year age band (n= 35) the ICC was 0.96 (95% CI from 0.95 to 0.97). This latter result included 21 families who had not discussed their child's GMFCS level with a health professional, 11 families who said they had, and 3 who did not indicate either way.

4.3.4 Discussion

These preliminary findings suggest that family ratings of the GMFCS made using the new questionnaire were reliable across the spectrum of impairments and disability. Although most of the available guidance for obtaining a substantial response from mailed surveys was followed the response rate was 48%. This was a reasonably good response to a single mailing which could almost certainly have been increased by persisting with either mailed or telephone reminders. The short period of time in which the pilot study was conducted, limited by the researcher's time at McMaster University, meant that reminder letters could not used. The addresses to which the invitations were mailed had not been updated since the previous study two years earlier. So, although 20 letters were returned as undelivered, how many invitations actually reached the intended families is not known.

There is a risk of respondent bias, in that families who returned questionnaires, particularly after only one mailing, may not be representative of the whole population. Although a comparison of the characteristics of the children in terms of age, gender and type and distribution of impairment was conducted, no information was readily available to compare other characteristics of families, for instance indices of their socio-economic status. There was no obvious tendency to respond with respect to the type and distribution or severity of children's cerebral palsy.

There were four out of 97 responses from which a single GMFCS Level could not be interpreted (4%). In these instances families had marked more than one description. One indicated a level III 'on a good day' and level IV 'on a bad day'. Another family selected items from descriptions of levels II to IV without any comments and without indicating a discrete choice. In answer to the first question, about the interpretability of the questionnaire,

it would appear that for the majority of families who responded the questionnaire was appropriately interpreted and completed.

Whilst no child was 'misclassified' by more than one level, there was not perfect agreement between professionals and families. The overall measure of chance corrected agreement was high enough to indicate good agreement but was lower than that reported by the developers between therapists. The disagreements appear to be random for children in levels III to V, however, several children whom professionals had rated at level I were rated by their families as level II, versus only one instance of the reverse where a child was rated as level I by the family and rated level II by a professional. Thus it would appear that some families tended to rate their child as more limited functionally than did professionals and this observation may represent more systematic difference. The interesting issue here is whether families who spend more time with their child are in fact aware of their child's performance in more varied situations than health professionals. Health professionals will tend to see children in clinic or school environments which are often specifically designed to enable children to function to the best of their ability. Furthermore, it has been shown that children within the same GMFCS level use different mobility methods at school, home and outdoors in the community (Palisano et al. 2003). Therefore when parents or family carers make a judgement about which description best represents the child's function, they are almost certainly doing so with knowledge of the child across a broader spectrum of environments than is true for health professionals. If one accepts that this is likely, then health professional ratings cannot be considered a 'gold standard' even though work on developing the GMFCS to date has depended on professional ratings. Without a known true level for each child only the reliability between assessments can be examined and not whether families are more or less reliable than health professionals.

By their nature, reliability coefficients are specific to the variability within the population in which they are measured. It was fortunate that there were a reasonable number of children in each of the five levels among our respondents to provide the required variability. If all the children had been in only one GMFCS Level there would be no variance for the observers and subsequent analyses to detect. The sample in the present study was part of a longitudinal study of motor development of children with cerebral palsy. To anyone who was aware of the GMFCS it may have been evident what was being studied. For this reason families were asked to indicate whether they had discussed the classification system with a health professional. Indeed, as expected, there was a slightly higher correlation for the families who indicated that they had discussed the GMFCS with a health professional. However there was also excellent reliability between professionals and families who indicated that a health professional had not discussed the GMFCS with them.

4.3.5 Conclusions

Although there was not perfect agreement, there was excellent reliability between GMFCS ratings made by families and professionals. This suggests that family report for the GMFCS collected using a newly piloted questionnaire provides a reliable method for measuring gross motor functional abilities of children between 6 and 12 years old. To address the potential confounding effect of prior knowledge of the GMFCS in this study population, and the fact that reliability coefficients are not readily generalisable because of their dependence on the characteristics of the population in which they are assessed, these issues were subsequently re-examined in a UK population.

4.4 CHAPTER SUMMARY

The systematic search and structured review described in this chapter identified seven health status instruments that measure aspects of children's activities and participation as defined by the ICF for Children and Youth. Given the recent publication of the ICF, it was perhaps not surprising that no instrument provided coverage of all the domains in the ICF. The LIFE-H offered the broadest coverage but is relatively long, potentially complex and the reliability of family assessment without a trained interviewer had not been assessed. At the time when the search was first conducted in autumn 2002, and when review was concluded in the summer of 2004, the ASK and LAQ instruments, between them, appeared to cover most domains of the ICF and are both appropriate for use in postal surveys (Morris et al. 2005) (Appendix B).

A survey was conducted with families of children with cerebral palsy in Canada for whom a physiotherapist had previously assessed their GMFCS Level. There was excellent reliability between GMFCS ratings made by families and professionals. This preliminary work suggests that family report for the GMFCS collected using a newly piloted questionnaire provides a reliable method for measuring gross motor functional abilities of children between 6 and 12 years old (Morris et al. 2004) (Appendix B). These two pieces of preparatory research enabled the main study in the UK to proceed. The family-assessed instruments identified, and piloted in the case of the GMFCS, were employed. The next chapter describes the methods used to conduct the main survey.

CHAPTER 5

THAMES VALLEY SURVEY

METHODS

5.1 CHAPTER OVERVIEW

This chapter describes the methods of the main study in which a survey was undertaken with families of children with cerebral palsy in the UK. The survey gathered families' ratings of the severity of their child's movement disability using the same questionnaire developed in the pilot study, corresponding to the 6-12 years of age-band of the Gross Motor Function Classification System (GMFCS). A subsequent survey of health professionals nominated by the families enabled an analysis of the reliability of family assessment for the GMFCS to be evaluated. Families were also invited to complete two other questionnaires: the Activities Scale for Kids (ASK) and the Lifestyle Assessment Questionnaire (LAQ-CP). These tools were identified in the structured review as the best available instruments for measuring children's activities and participation using family assessment. After the study had commenced a new measure of manual ability for children with cerebral palsy became available. Only families who had already participated in the survey at that point were invited to assess their child using the Manual Ability Classification System (MACS).

5.2 DEFINING AND TRACING THE STUDY POPULATION

The population eligible for this study were families of children with cerebral palsy born to a mother who was resident at the time of the child's birth in the defined geographical area known as the Thames Valley Strategic Health Authority (TVSHA), which comprises Berkshire, Buckinghamshire and Oxfordshire. The children were born between 1 January 1991 and 31 December 1997 and were between 6 and 12 years old when the survey commenced in January 2003.

5.2.1 Source of the study population

The source of the study population was the *4Child*, Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children, formerly the Oxford Register of Early

Childhood Impairment (ORECI). *4Child* monitors the prevalence of cerebral palsy, hearing and visual impairments of children born to mothers who were resident at the time of birth within the four English counties of Berkshire, Buckinghamshire, Northamptonshire and Oxfordshire. The database was established in 1984 and there is evidence of a high level of case ascertainment from capture-recapture analysis (A. da Silva, personal communication). To date, the information on the database has been collected without gaining the consent of parents, as is common practise for public health surveillance registers. The present study focused only on children from the counties of Berkshire, Buckinghamshire and Oxfordshire which together make up the Thames Valley Strategic Health Authority (TVSHA). This was partly for pragmatic reasons as the initial ethical approval from the Local Research Ethics Committee (LREC) in Oxford covered the whole of the Thames Valley area but not Northamptonshire. The findings from a single strategic health authority might also have more relevance for public health and health services policy.

5.2.2 Ethics Committee approval

To conduct the study approval was required from the *4Child* Advisory Group, local (LREC) and multi-centre research ethics committees (MREC). Following approval from the *4Child* Advisory Group the proposal was further refined into a set of procedures and the survey documentation was prepared. The survey of families was conducted in two phases. As families had not given their consent for the names of their children to be included on the *4Child* database, the first phase involved inviting families to take part and then only families who consented were sent questionnaires. The LREC approved the study on the basis that only families still resident in the Thames Valley Strategic Health Authority would be contacted. However, families who had migrated outside the district were not able be identified until after the tracing process. Ethics approval was subsequently sought and

granted by the Manchester MREC to include those families residing in the UK outside the Thames Valley. Details of the approved study protocol and all the subsequent protocol amendments were posted on the study website hosted by the National Perinatal Epidemiology Unit (www.npeu.ox.ac.uk).

5.2.3 Defining the study population

Details of the study population were extracted from *4Child* with the help of the Database Manager. A dataset was created that included only children with a confirmed diagnosis of cerebral palsy born to mothers resident within the TVSHA within the specified timeframe (1991-1997). Children known to have died were excluded; children with cerebral palsy thought to be of post-neonatal origin were retained in the study. The number of children in the study dataset was 338.

5.2.4 Tracing the study population

4Child staff complete the final routine data collection on children when they are aged 5 years. Details of family addresses and General Practitioner (GP) details were therefore not necessarily up to date and needed to be checked. The National Health Service (NHS) Information Authority maintains the NHS Strategic Tracing Service (NSTS) for England and Wales. The NSTS database holds details indexed by the NHS number unique to each person who is eligible to receive health services including patients' home addresses and their General Practitioner (GP). The NSTS is updated by all NHS organisations whenever patients have contact with their services. Permission to use the NSTS for this study was granted by the Caldicott Guardian and Information Officer at the Nuffield Orthopaedic Centre NHS Trust where the researcher has a clinical contract.

The *4Child* Database Manager provided a dataset for the study population that included the child's name, NHS Number, date of birth and gender, last known address, and the unique *4Child* identifying number. The relevant personal information was individually entered into the online version of the Person Tracing Service of the NSTS for each child. The search returned the name of each child together with details last recorded with the NHS for: home address, GP Practice Code and name of their GP. The GP addresses were then obtained from a separate NHS database with the help of hospital information management staff. The tracing process indicated that one child had died, and a current address was not available for a further five children. One had migrated to Scotland and one to the Republic of Ireland; the details of another three had been removed by the Health Authority without a given reason. Forty families had migrated outside the study area but remained within England. *4Child* obtained permission to use the NSTS directly while the study was in progress. Any changes to current addresses were checked and noted by the *4Child* Computer Programmer after six and twelve months.

Before families were contacted, their GPs was offered the opportunity to decline access if they felt it would have been inappropriate to contact the family as part of the survey. The GP for each child was sent a letter that briefly described the aim of the study and identified that the family of the named child would be invited to take part; copies of the family and child information sheets were also enclosed (Appendix A). The GP was requested to inform the researcher only if they felt it would be inappropriate for the family to be contacted.

5.3 FAMILY SURVEY

5.3.1 Invitation

Families residing in the Thames Valley were sent an invitation package by mail that included an *Invitation letter*, *Information sheet*, and an age appropriate *Information sheet for children* and a *Consent form* (Appendix A). Different information sheets were developed for children aged 6-8 years and those aged 8-12 years. The invitation letter referred to each of the enclosed documents, which were printed on different colour paper for ease of reference and identification.

The survey materials were prepared following as closely as possible the published recommendations for maximising response rates from mailed questionnaires (Edwards et al. 2002; McColl et al. 2002 ; Streiner & Norman 2003). Each invitation letter was addressed to the 'parent or carer' of the child and stated the name of the child for whom we were requesting information and was personally signed by the researcher. Families who wished to take part were asked to sign and return the consent form. Children who were capable and who wanted to could sign to indicate their consent to take part. Families could indicate if they preferred not to take part in which case reminder letters were not mailed. A third option was for families to indicate if they wanted more information and the researcher's contact details were provided. Families were assured that they would receive feedback of the findings from the study and newsletters were subsequently sent out by post at six month intervals. Return envelopes were individually stamped with first class postage and addressed to the NPEU. For families from whom a response was not received, a first reminder was mailed after four weeks and a second reminder after two more weeks.

With the agreement of the MREC, families who were residing outside the Thames Valley were provided with the questionnaires at the same time as their invitations. This made it clearer what was being asked of them. For these families, return of the questionnaires was taken as evidence of their consent to take part. Reminder letters were sent after four weeks and if indicated a second questionnaire package was mailed after a further two weeks.

5.3.2 Questionnaire Design

Questionnaire packages included several sections. The GMFCS family questionnaire for 6 to 12 year old children used in the Canadian survey had been found to perform satisfactorily (Morris et al. 2004). This questionnaire was therefore included in the UK survey without changes. A second single sheet questionnaire asked families to nominate some of their child's health professionals providing examples such as physiotherapists, paediatrician or orthopaedic surgeon. These were used for the subsequent survey of health professionals to obtain the professional assessment of the GMFCS level for the same child.

The Activities Scale for Kids (ASK) questionnaire was developed in Canada and contained a small number of North American expressions, for example 'pants' for 'trousers'. These were edited with the permission of the developers. The version of the ASK that was provided by the developers in Portable Document Format (PDF) was formatted for a size of paper (*Letter*) not commonly used in the UK. The ASK was modified to fit A4 and age-appropriate pictures were added to make it more colourful. The Lifestyle Assessment Questionnaire (LAQ-CP) was developed in the UK with children up to 11 years old but is routinely administered by the North of England Cerebral Palsy Survey when children are aged 4 years. The LAQ-CP therefore required only minimal modification to ensure all the questions were relevant to school age children, mainly by omitting references to preschool.

The LAQ-CP questionnaire was reformatted into a booklet style, including a few pictures, that matched the appearance of ASK questionnaire (Appendix A). All four data collection questionnaires were individualised using the identifying number unique to each child in the study.

5.3.3 Administration of Questionnaires

The survey package was sent using the UK standard first class postal service for families in the Thames Valley, and using the 'Recorded Signed for' service for families living elsewhere. A personalised letter, addressed to the person who had signed the consent form, or otherwise to the child's 'Parent or Carer', thanked families for agreeing to take part and provided instructions about the questionnaires. All letters were again printed with the colourful research unit and study-specific logos, and individually signed by the researcher. A return envelope was individually stamped with first class postage and addressed to the NPEU. Two reminder packages were mailed: a letter that was sent after four weeks expressing how important it was to return the questionnaires. If no response had been received two weeks later then a second package, including a full complement of questionnaires, was sent. With the approval of the LREC, non-responders residing in the Thames Valley were then mailed only the GMFCS questionnaire alone in an effort to obtain family-assessed ratings of children's movement ability for as many of the children as possible.

5.3.4 Test-Retest Study of family-assessment of the GMFCS

A second small survey was conducted with the families who participated. This included only the family assessment of the GMFCS to enable a test-retest reliability analysis between ratings made on different occasions. As a child's GMFCS Level would not be expected to

change over time then neither should the family-assessed rating. Families were contacted in a single mailing that was between three and six months after they had returned their original batch of questionnaires. The reason for the repeated survey was described in the introductory letter to families and another copy of the GMFCS questionnaire was supplied. No reminders were used in the test-retest part of the study.

5.3.5 Assessment of Children's Manual Ability

At the time when the survey was largely completed and most of the data had been collected, a group in Sweden were piloting their Manual Ability Classification System (MACS) for children with cerebral palsy. The MACS aims to describe children's manual ability when they are "handling objects in daily activities" (Eliasson et al. 2005). The structure of MACS was purposely styled on the GMFCS in that the distinctions between Levels are intended to be clinically meaningful (P. Rosenbaum, personal communication). The developers initially focused on children between 8-12 years old and used an expert group of health professionals to achieve a consensus on the descriptions of manual ability in each MACS Level. Further validity was established using interviews with therapists and parents of children with cerebral palsy to explore the content of each description. MACS assessments have been undertaken by pairs of therapists for 72 children between 8-12 years in Sweden and Australia indicating an excellent inter-observer reliability between health professionals (ICC = 0.96) (Eliason et al. 2005). The reliability between professional and family assessments for the MACS for 25 children, although a small number, was also excellent (ICC = 0.96). The Swedish group wanted to conduct larger reliability analyses and approached the researcher for help following the publication of the article on the reliability of family report for the GMFCS (Morris et al. 2004). A key incentive for gathering valid and reliable data regarding children's manual ability for the main UK study was that aspects of handling objects are

included in many of the items of the ASK and LAQ-CP questionnaires. The MACS therefore provided the serendipitous opportunity for collecting a further family-assessed explanatory variable for the analyses examining what factors influence children's activities and participation.

The survey of families asking about children's manual ability was approved by the MREC and LREC. One child was excluded as the family had identified them as having no hands at the time of the initial survey. Although, in principle, this would not preclude them from some adapted form of manipulation of objects, it was not possible to judge how the questions would be received by the family. An introductory letter with the colourful logos matching the paperwork of the main survey and related documents about the MACS were sent to all the families who had already participated in the main study. The developers of the MACS preferred that families were provided with the same instructions leaflet as were professionals (Appendix A). Families were therefore asked to read the standard instructions and report the MACS Level they felt best represented their child's manual ability. Families were again asked for permission to approach the child's health professionals and asked if the names of any therapists had changed.

5.4 HEALTH PROFESSIONAL SURVEY

Part of the family survey requested the names of the child's health professionals and where they worked, such as the name of the hospital or school (Appendix A). Each health professional was sent a letter that informed them that the family was taking part in the research, had identified them as working with their child and consented for the researcher to contact them. The child was named in the letter along with their date of birth and address. The letters were supplemented with a copy of the standard GMFCS 'user guide' (Appendix

B) from which health professionals were asked to provide an assessment of the GMFCS level for that child. The nominated professionals were later surveyed again by post to obtain their assessment of the child's manual ability, using the standard MACS instructions leaflet. For both assessments health professionals were asked to indicate on the data collection form whether they had made the assessment by 'direct observation' or by 'reviewing the child's records' or a 'mixture of both'. Prepaid return envelopes were provided for both surveys and reminder letters were mailed if no response was received after four weeks.

All the health professionals who took part in the survey were sent the same study newsletters mailed to families. This was important, not only as a courtesy, but also to make them aware of what the study achieved. Copies of the study newsletters that were mailed in May and October 2004 were added to the study website and are included in the Appendix A.

5.5 SOCIO-ECONOMIC INDICES

Socio-economic factors may firstly influence whether or not people participate in survey research and therefore potentially bias the findings, and secondly socio-economic factors may also influence children's activities and participation. Personal indicators such as occupation, financial income, or level of educational attainment are commonly used as proxies to the socio-economic status of individuals or households as they are brief and easy to collect. No personal socio-economic data were gathered in the survey and would not in any case have provided information about families who did not respond. Area deprivation measures are conceptually different to personal measures but offer an alternative approach for assessing the influence of socio-economic factors when personal information is unavailable (Spencer et al. 1999; Sundrum et al. 2005; Pattenden et al. 1999; Dolk 2001). It is also likely however that the smaller the area under consideration the closer the findings

will correlate with individual data (Adams et al 2005). Data relating to three socio-economic indices were obtained for this study. The Townsend scores (Townsend et al. 1988) and Carstairs scores (Carstairs and Morris, 1990) for England and Wales were provided by the Cathy Marsh Centre for Census and Survey Research at the University of Manchester. The UK government's revised English Indices of Deprivation (IMD 2004) (Noble et al. 2004) were provided by the Office of the Deputy Prime Minister (ODPM).

The Townsend index is derived from four variables from the UK national census; these are unemployment, overcrowded households, households without a car, and non-homeowners. The Carstairs index uses similar but slightly different census variables: the proportions of male unemployment, overcrowded households, households without a car, and households in the lowest social class. The proportions of households affected were calculated from the census, with all households in the defined designated area as the denominator. The areas for which these data were available corresponded to census wards from the 2001 census. The resulting individual variables had been standardised as z-scores and added together to create the index score for each area. Finally, quintiles were created for the national dataset so that the relevant national quintile could be referenced for each ward. Look-up tables provided by the Census Dissemination Unit with the national quintiles from the Cathy Marsh Centre for Census and Survey Research enabled the postcode of each family to be assigned to a ward area and thus a Townsend score, a Carstairs score and national quintile.

The IMD 2004 comprises seven domains of area deprivation that were derived from 37 routinely collected indicators: (i) income; (ii) employment; (iii) health and disability; (iv) education skills and training; (v) barriers to housing and services; (vi) living environment; and (vii) crime. The indicators are available for areas called Super Output Areas (SOA). The

smallest SOA is the Lower Layer SOA which typically comprises 5 output areas with a minimum population of 1000 and mean of 1500 people. The IMD is a weighted sum of deprivation domain scores in which the greatest weights were credited to the income and employment domains (22.5% each); health and education domains were less weighted (13.5% each); barriers to housing and services, crime and living environment were weighted least (9.3%). The IMD scores for England were divided into national quintiles by the *4Child* computer programmer. Look-up tables provided by the ODPM were then used to assign the postcode of each family in the complete study population, including non-responders, to a SOA and an IMD score and national quintile.

The IMD was selected to be the primary index of area deprivation for this study as the IMD was the most recently updated, in 2004 versus 2001 for the Townsend and Carstairs' indices. The IMD has also been rigorously developed to be a more comprehensive measure of area deprivation. One limitation of the IMD, due to the recent publication, was that no reference to its performance in public health research was available. The more established Townsend and Carstairs quintile data were therefore retained in the database for later reference and comparison.

5.6 DATA MANAGEMENT

All questionnaires were returned to the NPEU and their arrival recorded in the study database by the researcher. Packages from families were first inspected to see whether all four completed questionnaires were enclosed. If any documents were missing then a letter and duplicate questionnaire were sent to the family. Each individual questionnaire was then checked for completeness and clarity. For instance, a range of answers had been offered in response to many of the questions; the document was therefore inspected to ensure only one

answer had been selected. In the case of any uncertainty the coding was undertaken by the researcher prior to data entry.

The developers of the LAQ-CP provided a copy of their customised database (Microsoft Access) to facilitate data entry and expedient calculation of domain and summary scores. A custom database was written in Microsoft Visual Basic by the computing co-ordinator at the research unit to enable data from the ASK questionnaire and the family and health professional ratings of the GMFCS and MACS to be entered and stored securely. Data were entered once by the researcher and then again by administrative colleagues. The first and second data entry were compared for discrepancies. Any differences were listed and checked against the original questionnaire to determine the source of the anomaly and corrected in both datasets and rechecked. Details of each child's impairment were extracted from the *4Child* database by the Computer Programmer and likewise added to the final dataset for analyses. Impairment data included type and distribution of cerebral palsy, and the presence of seizures, hearing or visual impairments. Measures of children's oromotor function, intellectual delay, and upper and lower limb function from *4Child*, and the area deprivation indices were also merged into the dataset

5.7 STATISTICAL ANALYSES

The cleaned and coded data were imported into SPSS (Version 11) and for some analyses STATA (Version 8). A comparison of the known characteristics of participating and non-participating families and children was undertaken to gauge the extent of any respondent bias. The statistical significance of any differences in the proportion of children with various impairments amongst full and non-participants was compared using the chi-squared (χ^2) test for proportions.

5.7.1 Reliability of family report for the GMFCS and MACS

The agreement and reliability of classification made by families for the GMFCS and MACS was assessed by comparing these with classifications provided by the child's key health professionals. The GMFCS and MACS are ordinal scales with five levels. Absolute agreement was assessed as a percentage of all paired comparisons for the GMFCS and MACS overall and for each level. Cohen's kappa coefficient was used to determine the agreement corrected for chance for each individual GMFCS and MACS Level and for the overall system.

Then, as in the Canadian study described in chapter 4, the Intraclass Correlation Coefficient (ICC) was used to measure reliability as, like the weighted kappa statistic, it takes partial agreement into account (Fleiss and Cohen, 1973). The ICC was again determined using repeated measures analyses of variance (ANOVA) in SPSS to calculate the mean squares (MS) due to the children's ratings ($MS_{subjects}$) and the mean square error (MS_{error}). The calculation was then performed as shown in the following equation where the symbol σ represents variance.

$$ICC = \frac{\sigma_{subjects}}{\sigma_{subjects} + \sigma_{error}} = \frac{MS_{subjects} - MS_{error}}{MS_{subjects} + MS_{error}}$$

For each ICC reliability coefficient, 95% confidence intervals were determined by hand using Fisher's *Z* transformation (Fisher 1925; Streiner and Norman 2003). Classifications of children for whom two assessments of their GMFCS Level had been provided by families three months apart were used to determine the test-retest reliability. The ICC and 95% confidence intervals were again calculated as detailed above.

5.7.2 Reliability of the of the ASK and LAQ-CP instruments

The reliability of the ASK and LAQ-CP instruments was examined as they performed in this study. At an item or individual question level, the frequency that each response option was selected was examined; the mean score for each item was inspected to see if the mean was located central to the item scale. The same process was conducted for the summary scores for the ASK and each domain of the LAQ-CP, which is aggregated from the performance of all the items in a domain. The internal consistency of the ASK scale and each of the domains of the LAQ-CP was checked using the item-total correlation (Nunnally 1978) and Cronbach's alpha (Cronbach 1951) statistics.

5.7.3 Construct validity of instruments

The construct validity of the GMFCS, MACS, ASK and LAQ-CP scores were assessed to determine to what extent they correlated. The Pearson correlation was used to assess associations between the quasi-continuous scores, and Kendall's tau-b was used to examine correlation between the ordinal classification systems. Correlations between the GMFCS and MACS and the corresponding measures of lower and upper limb functioning recorded by *4Child* were assessed using Kendall's tau-b.

5.7.4 Factors affecting children's activities and participation

The ASK is a unidimensional health status measure which produces a single summary score which can be considered as quasi-continuous data. The LAQ-CP produces six domain scores and a total summary score as quasi-continuous data. Impairment data from *4Child* were coded with dummy variables where '0' indicated no impairment and '1' indicated, for example, 'vision loss'. Univariate categorical regression analyses were undertaken first to explore the association between each of the main outcome scores and the explanatory

variables. Variables that were significant at $p \leq 0.10$ in univariate analyses were retained for the subsequent multivariate analysis. Box plots were created for ordinal explanatory variables to examine the distribution of the outcome data and the linearity of any effect across categories. The variables found to be significant in univariate analyses were then all included in backwards stepwise regression analyses. The explanatory variable showing the least significant association with the outcome was dropped in sequence until only those variables significant at the $p < 0.05$ level remained in the model.

The robustness of the fit of the final regression models was checked using the following diagnostic procedures:

1. Examining the proportion (%) of variance explained overall by the model.
2. Plotting a histogram of the standardised residual values to see whether these were normally distributed. The residuals being the amount of score that remains unexplained by the model; the residuals were then standardised to have a mean of zero.
3. Plotting the standardised residuals against the fitted residual values predicted by the model and examining their distribution for outlying cases that were unusually distant from zero.
4. The Breusch-Pagan / Cook-Weisberg test for heteroskedasticity was used to test whether the variance in the residuals varied for particular values of the explanatory variables (Breusch & Pagan 1979; Cook & Weisberg 1983). Such variation would indicate that the model was not a good fit.

5.7.5 Sample size and power

Given that this study recruited from what was presumed to be a complete geographically defined population, the magnitude of the study sample was directly dependent upon achieving an adequate response to the survey. To determine how representative the final

study sample was, the known characteristics of those who fully participated in the survey were compared with those who did not fully take part, including those who were for whatever reason excluded from partaking.

The power to detect real differences between groups when they existed was assessed in sample size calculations using Stata 8 for a range of differences between mean scores of 20 to 50, with the alpha level set to 0.05 (Table 5.1). Assuming that there are 15, 20 or 25 children in each group, for instance in different levels of the GMFCS or MACS, and the within-group standard deviations were 35, the power to detect differences in mean scores of 20 to 50 points is listed. Greater differences in mean score and larger numbers of children in each group increase the power of the analyses. For instance, when comparing the mean scores for two GMFCS Levels with 15 children in each group there will be 88% power to detect a difference of 40 points. Similarly, groups with 20 children will have 95% power to detect a difference of 40 points. The shaded area in Table 5.1 indicates where the power would be within an acceptable range.

Table 5.1 The statistical power to detect real differences in outcome mean scores of 20 to 50 (standard deviation of 35) with varying numbers of subjects in each group, for example, in different GMFCS Levels

Difference in mean score	Number of individuals in each group		
	15	20	25
20	0.35	0.44	0.52
30	0.65	0.77	0.86
40	0.88	0.95	0.98
50	0.97	0.99	0.99

In considering the sample size for the multiple regression analyses the 'rule of thumb' advocated by several eminent authors is that the number of cases should be greater than ten times the number of explanatory variables (Altman 1991; Norman and Streiner 2000). As up to eleven explanatory variables were identified for inclusion in these analyses then data from at least 110 children were required to meet this criterion.

Reliability analyses such as the ICC calculate the ratio of true variance due to the subjects compared to that true variance plus measurement error; sometimes referred to as the signal to noise ratio. However if there is no variation in the sample there is consequently no variance to detect. For the analyses of reliability for family assessment for the classifications systems, the GMFCS and MACS, it was therefore as important to have a sample that included children with a wide range of abilities as well as consider the magnitude of the sample. Nonetheless, given that adequate reliability would be ICC of 0.9 and at least 0.85, which is equivalent to the lower bound 95% confidence interval (assuming a standard error of the reliability coefficient of 0.05), a sample size of at least 23 subjects is required, and for a narrower confidence interval of 0.88 (assuming a standard error of the reliability coefficient of 0.025) then around 72 subjects are required (Streiner and Norman 2003).

5.8 CHAPTER SUMMARY

This chapter has described how the study population from the Thames Valley was identified, traced and contacted. The study was initially approved by the Oxfordshire LREC permitting the tracing and inclusion of families who were residing within the Thames Valley area. These families were invited to take part in a postal survey and those that returned a signed consent form were mailed the package of questionnaires. The package contained the family assessment for the GMFCS, request for details of the child's health professionals, the Activities Scale for Kids (ASK) and Lifestyle Assessment Questionnaire (LAQ-CP). Once the main survey in Thames Valley was completed, a second survey of families enabled a test-retest analysis to be carried out on the reliability of family assessment of the GMFCS. With the approval of the LREC, non-responders were mailed only the GMFCS questionnaire in an effort to obtain family-assessed ratings of the GMFCS for as many of the population as possible. Following MREC approval, the families who were residing outside the Thames Valley were mailed invitations and the questionnaires for the full study. A further survey asking families about children's manual ability using the Manual Ability Classification System (MACS) was conducted with all families that had fully participated in the main survey. The sequence of key events in the survey is summarised in Figure 5.1

Completed questionnaires were returned to the researcher and the data were cleaned, and for quality control purposes double entered into custom databases. These duplicated data were compared to check for errors, and any errors found were amended. Statistical analyses examined the reliability of the instruments and estimated what factors affected children's activities and participation using multiple regression analyses.

Figure 5.1 Summary of the procedures undertaken in the Thames Valley study methods, represented by a timeline of key events.

	2002							2003											2004											2005										
	J	J	A	S	O	N	D	J	F	M	A	M	J	J	A	S	O	N	D	J	F	M	A	M	J	J	A	S	O	N	D	J	F	M	A	M	J	J	A	S	O	N	D
Ethical approval (LREC)																																										
Preparatory work																																									
Tracing																																									
GP survey																																									
Family survey (within Thames Valley)																																									
Health professional survey - GMFCS																																									
Retest - GMFCS																																									
Non-responders - GMFCS																																									
Data management																																									
Newsletters																																									
Ethical approval (MREC)																																									
Family survey (outside Thames Valley)																																									
Family survey (all participants) - MACS																																									
Health professionals survey - MACS																																									
Socio-economic indices gathered																																									
Statistical analyses																																									

CHAPTER 6

THAMES VALLEY SURVEY RESULTS I:

RESPONSE AND

PARTICIPANT CHARACTERISTICS

6.1 CHAPTER OVERVIEW

This chapter describes recruitment to the study and compares the known characteristics of children and families who participated in the survey with those who did not. Details of children's demographic and impairment characteristics were available from the *4Child* database. Area deprivation characteristics for families' area of residence were derived by linking families' postcode with information from the Indices of Multiple Deprivation 2004 (IMD). The chapter concludes with a summary of the characteristics of the children and families who participated and hence whose abilities are represented in the results.

6.2 SURVEY PARTICIPANTS

6.2.1 Response and recruitment

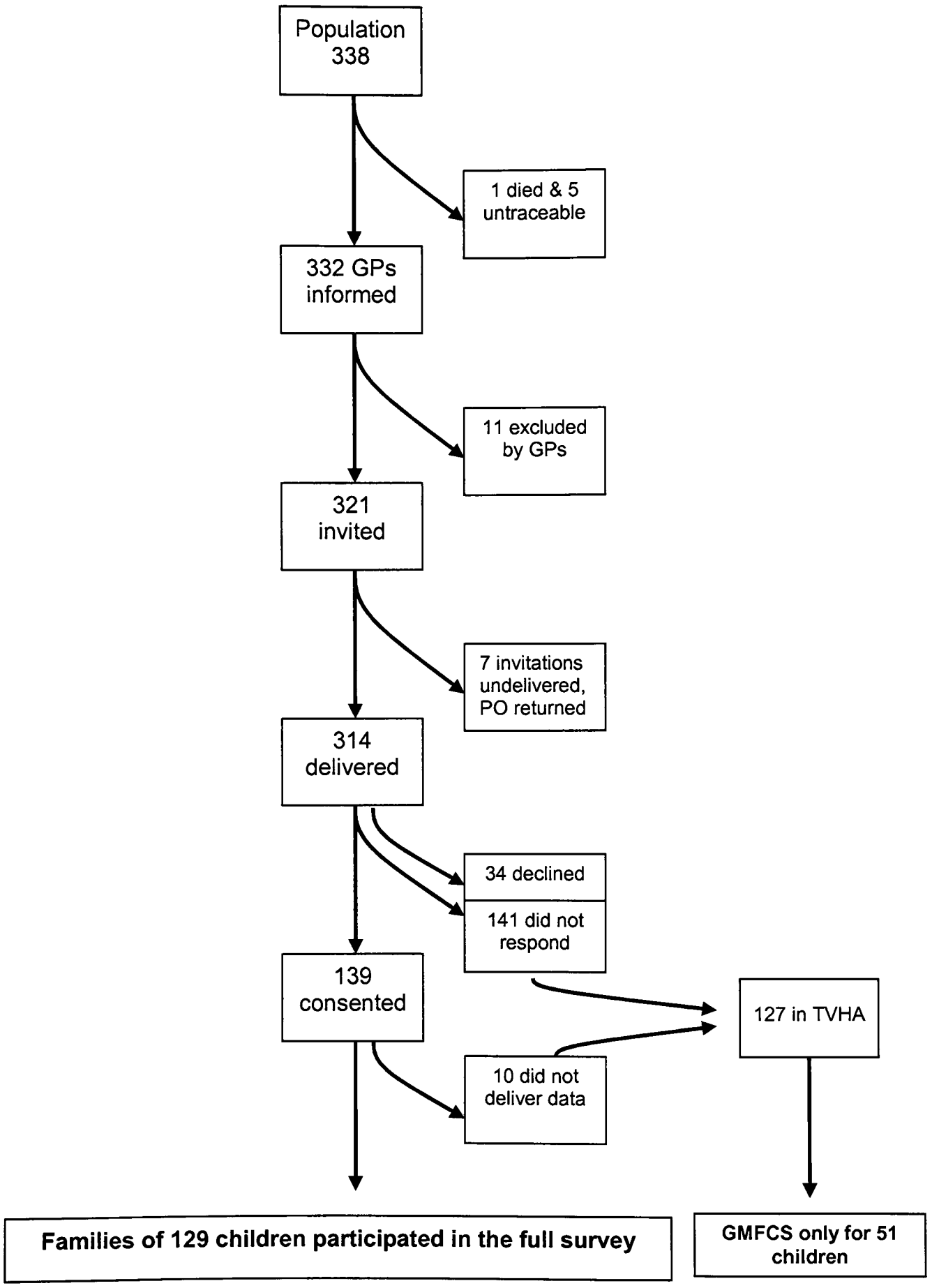
The *4Child* Database Manager identified 338 children who were born between 1991 and 1997 and eligible for inclusion in the study. Following the tracing procedure there were five children for whom a current address could not be identified and one child was deceased. GPs excluded the families of eleven children whom they felt it would be inappropriate to contact at the time of the survey. This included one child who was acutely unwell in hospital, one child who had been placed on the 'at-risk' register, and one who was residing in Australia. No reasons were given for the exclusion of eight other children. Some GPs suggested that the family were unlikely to respond although they did not stipulate their exclusion.

Invitations were mailed to the families of 321 children. Seven of these were returned by the postal service as undeliverable. Of the 314 families who could therefore have received the invitation, the families of 173 children responded (response 55%). The families of 139 children consented to take part (44%, or 80% of responders) and 34 actively declined using

that option on the consent form (11%, or 20% of responders). The families of 141 children did not respond after two reminder letters. The families of ten children who consented to take part did not return questionnaires. Therefore, in total, the families of 129 children fully participated in the survey, which is 41% of the potential survey population (Figure 6.1). Of these 112/272 children were residing within the Thames valley (response 41%) and 17/42 were scattered around England (response 40%). Among the participants there was one pair of siblings of different ages living in the same family. A subsequent mailing to the families of 127 children who were resident in the Thames Valley and had not specifically declined resulted in a further 51/127 responses (40%). These families provided only an assessment of their child's GMFCS Level. Families who fully participated were invited to take part in two further surveys to gather information for this study. First, a test-retest study of the GMFCS for participating families residing within the Thames Valley area achieved 86/112 responses (77%). Secondly, a survey asking about children's manual ability using the MACS with all participants elicited 91/128 responses (71%). A child identified by their family as having no hands in the first survey was not included in the survey of manual ability.

Full survey participants nominated between none and five of their child's health professionals depending on what clinical services the children were receiving. The survey of health professionals requesting their assessment of children using the GMFCS achieved 100/104 responses from physiotherapists (96%), 80/83 responses from paediatricians (96%), and 53/58 responses from orthopaedic surgeons (91%). The survey asking health professionals to classify children's manual ability using the MACS received 60/71 responses from physiotherapists (85%), 55/58 responses from paediatricians (93%), 21/24 responses from occupational therapists (88%).

Figure 6.1: Summary of recruitment of survey participants from study population.



6.2.2 Characteristics of participants

The known characteristics of the children from families who participated fully and returned all the questionnaires were compared with the characteristics of non-participants in order to determine whether the participants were representative of the study population. The non-participants included families of children who were either (i) not invited or declined to take part, or (ii) did not respond to the main survey, which included families who returned only GMFCS ratings.

Age and gender

There was no difference in the children's mean age between the full and non-participating groups: 9.9 and 9.8 years old ($p = 0.13$), or the proportion of males (56% and 54%, $p = 0.24$) (Table 6.1). There was a small number of children among both full and non-participating groups that had cerebral palsy presumed to have resulted from a post-neonatal insult. There were five children in the full participant group (4%) and thirteen among the non-participants (6%); the difference was not significant ($p = 0.35$).

Table 6.1 Comparison of full and non-participants by age and gender.

	Full participants N=129		**Non-participants** N=209		***p* value**
	n	**%**	**n**	**%**	
Male	72	*56*	113	*54*	0.13
Mean age, in years (sd)	9.9 (1.9)		9.8 (2.1)		

Type and distribution of cerebral palsy

As shown in Table 6.2 there was no difference between the full and non-participating groups in terms of the type of cerebral palsy defined using the SCPE classification ($p = 0.52$).

Table 6.2 Comparison of full and non-participant by type of cerebral palsy.

SCPE classification	Full participants N=129		Non-participants N=209		p value
	n	%	n	%	
Spastic	108	84	171	82	0.52
Dyskinetic	15	12	22	11	
Ataxic	3	2	4	2	
Unclassifiable	3	2	12	6	

In terms of the topographical distribution of the motor impairment, there was a slightly higher proportion of children with hemiplegia among the full participants and slightly fewer with bilateral cerebral palsy. However, the overall difference between the proportions of children with bilateral cerebral palsy and hemiplegia among full and non-participants was not statistically significant ($p = 0.14$). A more detailed analysis comparing the subtypes of cerebral palsy between the full and non-participating groups is shown in Table 6.3.

There was no significant difference between the full and non-participating groups the proportion of children with bilateral spastic cerebral palsy ($p = 0.28$), or spastic hemiplegia ($p = 0.13$); nor was there a difference in terms of the number of limbs affected by bilateral cerebral palsy ($p = 0.72$) or the proportions with left or right hemiplegia ($p = 0.57$). There

was no significant difference in the proportions of children with dyskinetic cerebral palsy (p = 0.75) or with different subtypes of dyskinesia (p = 0.49).

Table 6.3 Comparison of full and non-participants by subtype of cerebral palsy.

Subtype of CP	Full participants N=129		Non-participants N=209		*p* value
	n	**%**	**n**	**%**	
Spastic bilateral	62	*48*	113	*54*	0.28
Two-limbs	27	*21*	46	*22*	
Three-limbs	3	*2*	9	*4*	0.72
Four-limbs	32	*25*	58	*28*	
Spastic hemiplegia	46	*36*	58	*28*	0.13
Right	24	*19*	27	*13*	0.57
Left	22	*17*	31	*15*	
Dyskinetic	15	*12*	22	*9*	0.75
Choreo-athetotic	4	*3*	9	*4*	
Dystonic	5	*4*	8	*3*	0.49
Unclassifiable	6	*5*	5	*2*	
Ataxia	3	*2*	4	*2*	
Unclassifiable	3	*2*	12	*6*	

Sensory impairments

Table 6.4 shows that in terms of the sensory impairments recorded by *4Child*, the difference between the proportion of children in the full and non-participant groups with a defined level of hearing loss was not significant (p = 0.61), nor was there a significant difference in the proportion of cases with vision loss (p = 0.34).

Table 6.4 Comparison of full and non-participants with sensory impairments.

Sensory impairments	Full participants N=209		Non-participants N=209		Full participants
	n	%	n	%	
Hearing loss*	5	*4*	6	*2*	0.61
Vision loss**	25	*19*	32	*15*	0.34

***Hearing loss** was sensorineural loss of 50dB or more averaged across the range 0.5 to 4 kHz in the better ear. In the absence of a pure-tone audiogram, all children with a hearing aid fitted for sensorineural loss were included.
****Vision loss** referred to visual acuity in the better eye of 6/18 or less; if visual acuity could not be measured, an assessment of the degree of visual impairment was made on the behavioural responses of the child.

Seizures

The proportion of children who were recorded as suffering from seizures in the year they were age 5 years old was identical in both groups (23%). There were 13 children about whom the reporting paediatrician had been uncertain whether the child suffered seizures and four cases missing these data (missing data about seizures accounted for 4% of full participants and 6% of non-participants).

Intellectual delay

There was a smaller proportion of children recorded as having any intellectual impairment among the full participants than the non-participants, 38% versus 47% , the difference was however not statistically significant ($p = 0.11$). There were 43 children for whom the severity of intellectual delay was uncertain or unknown, accounting for 11% of full participants and 13% of non-participants. Information about the IQ level was recorded for 139 children with intellectual delay. Ten cases were missing these data, accounting for 6%

of full participants and 7% of non-participants. The difference between the proportions within defined ranges of IQ levels was marginally significant ($p = 0.07$) (Table 6.5). There was a greater proportion of children with no delay among participants and a slightly smaller proportion with minor and moderate delay, while the proportions with severe delay were similar.

Table 6.5 Comparison of full and non-participants by the severity of intellectual delay.

Intellectual impairment	Full participants N=113		Non-participants N=174		*p* value
	n	%	n	%	
IQ < 50	26	*23*	37	*21*	0.07
IQ 50-69	13	*11*	27	*16*	
IQ 70-84	8	*7*	28	*16*	
No delay	66	*58*	82	*47*	

Oromotor function

In terms of oromotor function, there was a smaller proportion of children recorded as having a speech problem among the full participants, 33% versus 45% among the non-participants ($p = 0.07$). There were 28 cases which were either uncertain or missing these data items (accounting for 6% of full participants and 10% of non-participants). The proportions of children with swallowing problems were similar, 14% among participants and 17% among non-participants ($p = 0.62$); 39 cases were missing these data and are not included in the χ^2 test (accounting for 9% of full participants and 13% of non-participants). Likewise for the proportions of children with drooling problems, 21% among participants and 27% among

non-participants ($p = 0.20$), 44 cases were missing these data and are not included in the χ^2 test (accounting for 12% of full participants and 15% of non-participants).

Upper and lower limb function

Using indicators of children's movement ability recorded by *4Child*, a third of children in both the full and non-participant groups were not walking at age five years, 31% among participants and 33% among non-participants. However, using the complete four-level 'Evans' classification, shown in Table 6.6, the difference between the groups was marginally significant ($p = 0.05$).

Table 6.6 Comparison of children's lower limb function among full and non-participants.

Lower limb function	**Full participants** N=122		**Non-participants** N=200		***p* value**
	n	***%***	**n**	***%***	
Walks fluently	14	*11*	11	*5*	0.05
Functional, not fluent	52	*40*	74	*35*	
Restricting lifestyle	16	*12*	45	*21*	
Not walking	40	*31*	70	*33*	

Using *4Child*'s four-level classification of upper limb function the difference between full and non-participant groups was also marginally significant ($p = 0.06$) (Table 6.7). There was a greater proportion of children with 'some difficulty' among participants, and a smaller proportion 'able to feed themselves'. However, it was notable that the differences appear in the middle strata, whilst the proportions were similar at each end of the scale. The proportion

of children with no difficulty was 18% among participants and 19% among non-participants, and the proportion of children unable to feed themselves was 22% in both groups. There were 10 cases missing these data accounting for 3% of children in both groups who were not included in the χ^2 test.

Table 6.7 Comparison of children's upper limb function among full and non-participants

Upper limb function	Full participants N=125		Non-participants N=203		*p* value
	n	%	n	%	
No problem	22	*18*	39	*19*	0.06
Some difficulty	53	*42*	60	*30*	
Able to feed self	23	*18*	59	*29*	
Unable to feed self	27	*22*	45	*22*	

Deprivation in area of residence

Using their last known address, full and non-participating families were also compared using postcode derived national quintiles of the Indices of Multiple Deprivation for England (IMD). There were 16 children for whom a unique IMD quintile could not be derived, of which two were among the participants. These included the five children for whom an address could not be traced and eleven that did not yield a single quintile due to the overlapping composition of 'super output areas'. These 16 children were therefore excluded from analyses involving area deprivation data. Overall the majority of families were living in areas corresponding to the least deprived areas relative to all of England. For the total survey population: 44% of families were in quintile I, 22% in quintile 2, 16% in quintile 3, 14% in quintile 4, and only 4% in the areas with greatest deprivation, quintile V. A

comparison of the proportions of full and non-participating families resident in each area quintile is shown in Table 6.8.

Table 6.8 Comparison of full and non-participants by deprivation in area of residence using national quintiles of the Indices of Multiple Deprivation 2004

IMD 2004 national quintiles	Full participants N=129		Non-participants N=198		*p* value
	n	%	n	%	
Quintile I (least deprived)	67	*53*	76	*38*	
Quintile II	23	*18*	49	*25*	
Quintile III	17	*13*	36	*18*	0.11
Quintile IV	17	*13*	27	*14*	
Quintile V (most deprived)	3	*2*	10	*5*	

A greater proportion of fully participating families were resident in areas corresponding to the least deprived quintile, 53%, versus 38% in the non-participants. Overall the difference between the proportions in each quintile of the IMD was not statistically significant (p = 0.11). However, the relatively small number of families residing in the most deprived quintile limits the power of this analysis. The quintiles were therefore recalibrated using only the indices from the IMD for families of children within the study population. The comparison of full and non-responders was then repeated as shown in Table 6. 9, and was still found to be non-significant (p = 0.12). The trend persisted for a higher proportion of families who fully participated to be residing in the least deprived area corresponding to quintile I (27% versus 16%). Similarly, the trend for a marginally lower response from

families residing in more deprived areas, quintile III to V, was also observed. Overall, however, the difference between full and non-respondents by deprivation in their area of residence during the study was not significant ($p = 0.12$).

Table 6.9 Comparison of deprivation in area of current residence using quintiles of the Indices of Multiple Deprivation 2004, derived from within the study population.

	Full participants N=127		Non-participants N=198		p value
IMD 2004 within study quintiles	**n**	**%**	**n**	**%**	
Quintile I (least deprived)	34	27	31	16	
Quintile II	27	21	38	19	
Quintile III	22	17	43	22	0.12
Quintile IV	21	17	44	22	
Quintile V (most deprived)	23	18	42	21	

The trend for a greater response from families residing in the least deprived national quintile (quintile I) was also observed in the test-retest survey for the GMFCS, the MACS survey and the survey of non-responders living in the Thames Valley. Just over half of the 51 responses to the survey of non-responders in the Thames Valley were from families living in quintile I areas and the overall pattern of difference across all quintiles was then marginally significant ($p = 0.06$). The comparison of full and non-participants was repeated with the more established Townsend and Carstairs' area deprivation indices as shown in Tables 6.10 and 6.11. These analyses indicate that whichever area deprivation index is used the response was

greater from families residing in the least deprived national quintile. Notably, fewer families were included in Quintile I, the least deprived, using the Townsend system than the IMD, and the Carstairs system included fewer again than the Townsend.

Table 6.10 Comparison of deprivation in area of current residence using national quintiles of the Carstairs index derived from the 2001 Census.

Carstairs index (2001 Census)	Full participants N=127		Non-participants N=198		*p* value
	n	%	n	%	
Quintile I (least deprived)	51	*40*	66	*33*	
Quintile II	28	*22*	49	*25*	
Quintile III	22	*17*	26	*13*	0.31
Quintile IV	17	*13*	32	*16*	
Quintile V (most deprived)	9	*7*	25	*13*	

Table 6.11 Comparison of deprivation in area of current residence using national quintiles of the Townsend index derived from the 2001 Census.

Townsend index (2001 Census)	Full participants N=127		Non-participants N=198		*p* value
	n	%	n	%	
Quintile I (least deprived)	43	*34*	39	*20*	
Quintile II	31	*24*	61	*31*	
Quintile III	25	*20*	41	*21*	0.07
Quintile IV	19	*15*	38	*19*	
Quintile V (most deprived)	9	*7*	19	*10*	

By linking the GP practice code from the NSTS to a database of Primary Care Trusts (PCT), 120 families that fully participated in the survey were receiving their health services from one of 30 PCTs (Table 6.12), nine cases were missing these data.

Table 6.12 Primary Care Trusts providing services to fully participating families.

Primary Care Trust	n=120
Chiltern And South Bucks*	11
Milton Keynes *	11
South East Oxfordshire*	11
Wycombe *	11
Vale Of Aylesbury*	9
Reading*	9
Bracknell Forest*	6
Oxford City*	6
Windsor, Ascot And Maidenhead*	6
Wokingham*	6
Cherwell Vale*	5
South West Oxfordshire*	4
North East Oxfordshire*	4
Newbury And Community*	4
Slough*	2
1. North Hampshire	1
2. North Surrey	1
3. Nottingham City	1
4. Central Cheshire	1
5. Shropshire County	1
6. South Hams And West Devon	1
7. South Wiltshire	1
8. Southern Norfolk	1
9. Suffolk West	1
10. Teignbridge	1
11. Coventry Teaching	1
12. Daventry And South Northamptonshire	1
13. East Elmbridge And Mid Surrey	1
14. Maldon And South Chelmsford	1
15. Mid-Sussex	1

*indicates within Thames Valley Strategic Health Authority

6.3 Summary of survey response and participants' characteristics

The Thames Valley survey received a moderate response. The families of 129 children fully participated in the survey, which is equal to 41% of the population who could have received invitations, and 38% of the surviving complete geographically defined study population. Families of a further 15% of the study population subsequently provided assessments of their child's movement ability by GMFCS Level in a secondary survey of families who did not respond to the initial invitation, which was carried out to describe the abilities of as many of the complete population as possible. Therefore family-assessed GMFCS ratings were available for 53% of the total population. The two supplementary surveys of families who had participated fully in the main survey received a better response from families, 77% for the GMFCS retest and 70% for the MACS. The response from health professionals was excellent, exceeding 90% for the request for GMFCS assessments and 85% for MACS assessments.

The families who participated fully in the survey only represent approximately 40% of the children in eligible population. Therefore, a series of analyses was conducted to determine whether the children of families who fully participated were different from non-participants. Characteristics of children's demography and impairments were available from the *4Child* database. The key characteristics of the sample who fully participated in the survey and whose family provided a GMFCS rating are shown in Table 6.13. In general, there were no statistically significant differences between the children of families who fully participated and those that did not take part. However full participants had relatively less severe intellectual delay and slightly better upper and lower limb function in the mid range of the scales used by *4Child*, and half of families lived in areas corresponding to the least deprived national quintile of the Indices of Multiple Deprivation 2004.

Table 6.13 Summary of key characteristics of the full participants and children whose families provided a GMFCS rating

	Full survey participants N=129		**Family-assessed GMFCS** N=175	
Mean age, years *(sd)*	9.9 (1.9)		9.9 (2.0)	
	n	***%***	**n**	***%***
Female	57	*44*	74	*42*
Male	72	*56*	101	*58*
Type of cerebral palsy				
Spastic	108	*84*	143	*82*
▪ **Bilateral**	62	*48*	81	*57*
▪ **Hemiplegia**	46	*36*	62	*43*
Dyskinetic	15	*12*	22	*13*
Ataxic	3	*2*	3	*2*
Unclassified	3	*2*	7	*4*
Associated impairments				
Seizures	28	*23*	40	*24*
Hearing loss	5	*4*	5	*3*
Vision loss	25	*19*	34	*19*
▪ **IQ < 50**	26	*23*	32	*21*
▪ **IQ 50-69**	13	*11*	19	*13*
▪ **IQ > 70**	74	*65*	98	*66*

Children with hemiplegia were represented in a greater proportion amongst the total number of families who provided an assessment of the GMFCS compared to the children of families who fully participated and the complete geographically defined population.

CHAPTER 7

THAMES VALLEY SURVEY RESULTS II:

PERFORMANCE

OF THE SURVEY INSTRUMENTS

7.1 CHAPTER OVERVIEW

This chapter reviews the performance of the instruments used in the survey to measure children's abilities, activities and participation using family-assessed methods. Building on the previous section, these data describe the activities and participation of the children and families that took part, as well as enabling the reliability and validity of the instruments themselves to be reviewed. The reliability of family assessment for the GMFCS and MACS was examined by comparing agreement between classifications made by families and health professionals, and between different professions. The reliability of the ASK and LAQ-CP instruments was evaluated by examining item response frequencies and the internal consistency of each domain and summary scale. The construct validity of the instruments was assessed by examining the correlation between the scores produced by each instrument

7.2 GROSS MOTOR FUNCTION CLASSIFICATION SYSTEM (GMFCS)

GMFCS assessments were provided by 126 eligible fully participating families for 127 children, one family returned a separate form for two children who were siblings and both had cerebral palsy. Two further assessments did not indicate a single GMFCS Level and were excluded. The major professions nominated by families as people to report their child's GMFCS level were physiotherapists, paediatricians and orthopaedic surgeons and some clinicians provided classifications for more than one child. Subsequently, 58 physiotherapists provided between one and six GMFCS assessments for 100 children, 31 paediatricians provided between one and 12 assessments for 80 children, 10 orthopaedic surgeons provided between one and 33 assessments for 53 children; in addition there were assessments provided for 28 children by a mixture of paediatric neurologists, neurosurgeons, occupational therapists and orthotists. The number of ratings made in each GMFCS Level by families and the larger professional groups is shown in Table 7.1.

Table 7.1 GMFCS ratings made by families and key health professional groups

	Families		Physiotherapists		Paediatricians		Orthopaedic surgeons	
GMFCS	**n**	***%***	**n**	***%***	**n**	***%***	**n**	***%***
Level I	32	*25*	36	*36*	24	*30*	4	*8*
Level II	43	*34*	19	*19*	15	*19*	18	*34*
Level III	15	*12*	16	*16*	13	*16*	11	*21*
Level IV	14	*11*	15	*15*	11	*14*	13	*25*
Level V	23	*18*	14	*14*	17	*21*	7	*13*
Total	127		100		80		53	

Notably, families and orthopaedic surgeons reported more children in Level II and physiotherapists and paediatricians reported more children in Level I, whereas orthopaedic surgeons reported relatively few children in Level I.

7.2.1 Agreement

Families and physiotherapists agreed for 64% of the children's GMFCS, both families and paediatricians and families and orthopaedic surgeons agreed for 59% of the children. The number and proportion of perfect agreements between families and the professional groups for each GMFCS Level is summarised in Table 7.2, chance-corrected agreement is indicated by the kappa statistic (κ). The chance-corrected agreement for each GMFCS Level ranged from 0.34 to 0.82; the lowest value was for agreement between families and paediatricians for GMFCS Level III and was highest for all professions for GMFCS Level V. Overall, the chance-corrected agreement between families and physiotherapists was $\kappa = 0.54$, between families and paediatricians $\kappa = 0.50$, and between families and orthopaedic surgeons $\kappa = 0.47$.

Table 7.2 Number of perfect agreements between families and each of the professional groups, showing chance corrected agreement (κ) for each GMFCS Level.

Family assessed GMFCS	Physiotherapists (N=99)			Paediatricians (N=79)			Orthopaedic surgeons (N=51)		
	n	%	κ	n	%	κ	n	%	κ
Level I	17	17	0.53	9	11	0.46	3	6	0.45
Level II	16	16	0.44	13	16	0.42	11	22	0.47
Level III	8	8	0.48	5	6	0.34	4	8	0.56
Level IV	8	8	0.48	6	8	0.44	6	12	0.51
Level V	14	14	0.82	14	18	0.81	6	12	0.60
Total	63	64	0.54	47	59	0.50	30	59	0.47

In general, these statistics rate as 'fair to good' agreement according to the recommendations by Landis and Koch (1977).

7.2.2 Disagreement

The number of disagreements between families and each of the larger professional groups about GMFCS Level is shown in Table 7.3. Disagreements were most frequent between Levels I and II and occurred by more than one level for only three assessments. Children whom physiotherapists or paediatricians rated as Level I were frequently rated by their families at Level II, whilst the converse was rarer. Only one child who had been classified as Level II by a physiotherapist was classified by their family as Level I, and no children classified as Level II by paediatricians were classified as Level I by their families. In contrast to the physiotherapists and paediatricians, orthopaedic surgeons classified six children at Level II that families had classified as Level I. There was a tendency for some families to classify their children one level lower than physiotherapists or paediatricians but one level higher than orthopaedic surgeons, however the trend was not indicative of

systematic bias. Disagreement between health professionals of more than one GMFCS Level was also rare. One child classified by a paediatrician as Level I was classified by an orthopaedic surgeon as Level III, and similarly, one child classified as Level III by a paediatrician was classified by the surgeon as Level V. One child classified as Level I by a physiotherapist was classified as Level III by a paediatrician, and one classified as Level III by a physiotherapist was classified as Level V by the paediatrician. Physiotherapists classified two children in Level I who were classified by orthopaedic surgeons as Level III.

Table 7.3 Number of disagreements between families and each of the professional groups for the GMFCS.

Disagreement with families	Physiotherapists (N=99)		Paediatricians (N=79)		Ortho surgeons (N=51)	
	n	%	n	%	N	%
Level I / II	19	*19*	15	*19*	6	*12*
Level I / III	1	*1*	-	-	-	-
Level II / III	4	*4*	5	*7*	6	*12*
Level III / IV	7	*7*	7	*9*	3	*6*
Level III / V	-	-	1	*1*	1	*2*
Level IV / V	5	*5*	4	*5*	5	*9*
Total	36	*36*	32	*41*	21	*41*

The direction of disagreement between observers is shown more clearly in the tables in the following section.

7.2.3 Reliability

Tables 7.4 to 7.6 display the agreement between families and each of the professional groups. The reliability of family assessment of the GMFCS relative to each group was assessed using the Intraclass Correlation Coefficients (ICC) and is shown at the bottom of each table with 95% confidence intervals. Tables 7.4 to 7.6 show that, although there was not complete agreement between families and professionals about every child, and by taking partial agreement into account, the correlation between observers was very high. The reliability of family assessment relative to physiotherapists was ICC = 0.92 (95% CI from 0.90 to 0.93), relative to paediatricians was ICC = 0.90 (95% CI from 0.88 to 0.92), and relative to orthopaedic surgeons was ICC = 0.87 (95% CI from 0.82 to 0.90).

Table 7.4 Comparison of GMFCS ratings by Families and Physiotherapists

		Families					
		I	**II**	**III**	**IV**	**V**	**Total**
Physiotherapists	**I**	17	18	1			36
	II	1	16	2			19
	III		2	8	5		15
	IV			2	8	5	15
	V				-	14	14
	Total	18	36	13	13	19	99

ICC = 0.92 (95% CI from 0.90 to 0.93)

Table 7.5 Comparison of GMFCS ratings made by Families and Paediatricians

		Families					
		I	**II**	**III**	**IV**	**V**	**Total**
Paediatricians	**I**	9	15				24
	II	-	13	2			15
	III		3	5	5		13
	IV			2	6	2	10
	V			1	2	14	17
	Total	9	31	10	13	16	79

ICC = 0.90 (95% CI from 0.88 to 0.92)

Table 7.6 Comparison of GMFCS ratings made by Families and Orthopaedic Surgeons

		Families					
		I	**II**	**III**	**IV**	**V**	**Total**
Orthopaedic Surgeons	**I**	3	-				3
	II	6	11	1			18
	III		5	4	2		11
	IV			1	6	5	12
	V			1	-	6	7
	Total	9	16	7	8	11	51

ICC = 0.87 (95% CI from 0.82 to 0.90)

The absolute and chance-corrected agreement, and reliability, between assessments for the GMFCS made by the different professional groups was also examined (Tables 7.7 to 7.9). Paediatricians and physiotherapists agreed exactly on children's GMFCS Level 72% of the time (κ =0.65); orthopaedic surgeons and physiotherapists agreed precisely for 50% of children (κ =0.37); and orthopaedic surgeons and paediatricians agreed exactly for 50% for children (κ =0.38). The coefficients of reliability (ICC) between professional assessments ranged from 0.82 to 0.92. There were 26 children who had been classified by all three key professions for which the reliability was ICC=0.91 (95% CI from 0.90 to 0.96).

Table 7.7 Comparison of GMFCS ratings made by paediatricians and physiotherapists

		Paediatricians					
		I	**II**	**III**	**IV**	**V**	**Total**
Physiotherapists	**I**	14	2	1			17
	II	3	8	3			14
	III		2	6	3	1	12
	IV			1	8	2	11
	V					11	11
	Total	17	12	11	11	14	65

ICC=0.92 (95% CI from 0.89 to 0.94)

Table 7.8 Comparison of GMFCS ratings made by paediatricians and orthopaedic surgeons

		Paediatricians					
		I	**II**	**III**	**IV**	**V**	**Total**
Orthopaedic Surgeons	**I**	1					1
	II	5	5	1			11
	III	1	1	2	2		6
	IV			1	4	3	8
	V			1	1	4	6
	Total	7	6	5	7	7	32

ICC=0.83 (95% CI from 0.67 to 0.90)

Table 7.9 Comparison of GMFCS ratings made by physiotherapists and orthopaedic surgeons

		Physiotherapists					
		I	**II**	**III**	**IV**	**V**	**Total**
Orthopaedic Surgeons	**I**						0
	II	9	7				16
	III	2	1	5	2		10
	IV			3	5	3	11
	V				2	4	6
	Total	11	8	8	9	7	43

ICC=0.82 (95% CI from 0.75 to 0.86)

The means by which different health professionals made their assessments was noteworthy. The majority of physiotherapists made their assessments by direct observation (34%) or used a mixture of observation and record review (54%) while only 8/99 (8%) of ratings were made solely by reviewing the records. A smaller proportion of paediatricians used direct observation (24%) and the mixed method (48%) and hence more of them were dependent on reviewing the child's records (28%). Orthopaedic surgeons depended almost entirely on reviewing children's records (84%) or the mixed method (10%), with only 2/51 ratings (4%) made by direct observation.

The reliability coefficients for family versus professional report using the GMFCS were higher when a greater proportion of the professionals indicated using direct observation and lower when professionals depended on information from the child's medical records. In contrast to the previous study in Canada, very few families were aware of the GMFCS: only 13/129 (10%) indicated on the questionnaire that they had discussed the system with a health professional.

7.2.4 Test-retest survey with families

In the retest survey 7/86 families did not indicate a discrete GMFCS Level. A comparison of the first (test) and second (retest) assessments for the remaining 79 children is shown in Table 7.10.

Table 7.10 Comparison of first and second family assessments of GMFCS

		First family assessment					
		I	**II**	**III**	**IV**	**V**	**Total**
Retest after 3 months	**I**	19	1				20
	II	2	25	1			28
	III			8	1		9
	IV				9		9
	V					13	13
	Total	21	26	9	10	13	79

ICC = 0.98 (95% CI from 0.97 to 0.98)

There was very good agreement between first and second assessments three months apart; 94% of classifications concurred exactly and the chance-corrected agreement was $\kappa = 0.92$. Of the five classifications that disagreed, three were between Levels I and II, one each between Levels II and III, and III and IV. The reliability of family assessment for the GMFCS on two separate occasions was extremely high ICC=0.98 (95% CI from 0.97 to 0.98).

7.2.5 Summary of GMFCS performance

Overall family and professional assessments of the GMFCS were well correlated. There was not absolute agreement on the GMFCS Level for every child although disagreements by more than one level were extremely rare. In general there was better agreement with families when professionals indicated they made their assessment using some degree of direct observation of the child rather than exclusively reviewing the child's medical records. The reliability of family assessment for the GMFCS, indicated by the ICC, and the agreement corrected for chance, expressed using the kappa coefficient, were comparable to the indices of inter-observer agreement and reliability between professionals. The reliability coefficients for family assessment of the GMFCS relative to each professional group are summarised in Table 7.11.

Table 7.11 Summary of indices of agreement and reliability between family and professional classifications using the GMFCS

	Number of children	Absolute and chance corrected agreement		Reliability
Observers	**n**	**%**	**κ**	**ICC**
Families / Physiotherapists	99	64	0.54	0.92
Families / Paediatricians	79	59	0.50	0.90
Families / Orthopaedic Surgeons	51	59	0.47	0.87
Physiotherapists / Paediatricians	65	72	0.65	0.92
Paediatricians / Orthopaedic Surgeons	43	50	0.37	0.82
Physiotherapists / Orthopaedic Surgeons	32	50	0.38	0.83

7.3 MANUAL ABILITY CLASSIFICATION SYSTEM (MACS)

Families provided MACS assessments for 88 children; three further assessments did not indicate a single MACS Level and these were excluded. The physiotherapists, paediatricians and occupational therapists who had been nominated by families were surveyed for their assessment of the MACS for these children. Orthopaedic surgeons were not contacted as they rarely record information on manual ability for children with cerebral palsy. Subsequently, 36 physiotherapists classified between one and six of 60 children, 21 paediatricians classified between one and eight of 55 children, 17 occupational therapists provided between one and two assessments for 23 children. The number of ratings made in each MACS Level by families and the larger professional groups is shown in Table 7.12.

Table 7.12 Summary of MACS ratings made by families and health professional groups

	Families		Physiotherapists		Paediatricians		Occupational therapists	
MACS	**n**	***%***	**n**	***%***	**n**	***%***	**n**	***%***
Level I	14	*16*	17	*28*	9	*16*	2	*9*
Level II	30	*34*	18	*30*	16	*29*	13	*57*
Level III	18	*21*	8	*13*	13	*24*	4	*17*
Level IV	13	*15*	7	*12*	6	*11*	3	*13*
Level V	13	*15*	10	*17*	11	*20*	1	*4*
Total	88		60		55		23	

7.3.1 Agreement

The agreement between families and the professional groups for each MACS Level is summarised in Table 7.13, where absolute agreement is shown as a percentage and chance-corrected agreement is indicated by the kappa statistic (κ). Families and physiotherapists

agreed exactly for 51% of the children's MACS level, both families and paediatricians and families and occupational therapists agreed for 58% of the children.

Table 7.13 Number of perfect agreements between families and each of the professional groups for the MACS, showing chance corrected agreement (κ) for each MACS Level.

Family-assessed MACS	Physiotherapists (N=59)			Paediatricians (N=52)			Occupational therapists (N=21)		
	n	%	κ	n	%	κ	n	%	κ
Level I	5	*8*	0.37	5	*10*	0.74	1	*5*	0.45
Level II	10	*17*	0.30	8	*15*	0.41	7	*33*	0.45
Level III	4	*7*	0.28	4	*8*	0.22	1	*5*	0.04
Level IV	4	*7*	0.42	4	*8*	0.25	2	*10*	0.49
Level V	7	*12*	0.64	9	*17*	0.83	0	-	-
Total	30	*51*	0.38	30	*58*	0.46	11	*53*	0.32

The chance-corrected agreement between families and occupational therapists for MACS Levels III and V was poor, in part because so few children were classified. The chance corrected agreement between families and physiotherapists and paediatricians, respectively, ranged from κ=0.22 to 0.83. The lowest agreement was for MACS Level III and the highest agreement for MACS Level V, and between families and paediatricians also for Level I. Overall, the chance-corrected agreement between families and physiotherapists for the MACS was κ=0.38, between families and paediatricians κ=0.46, and between families and occupational therapists κ=0.32; not that good and only rated as 'fair' agreement according to Landis and Koch (1977).

7.3.2 Disagreement

The number of disagreements between families and each of the larger professional groups about MACS Level is shown in Table 7.14.

Table 7.14 Number of disagreements between families and each of the professional groups for the MACS.

Disagreement with families	Physiotherapists (N=59)		Paediatricians (N=52)		Occupational therapists (N=21)	
	n	%	n	%	n	%
Level I / II	11	*18*	1	*2*	2	*10*
Level I / III	1	*2*	2	*4*	-	-
Level II / III	8	*13*	10	*19*	4	*19*
Level II / IV	1	*2*	2	*4*	-	-
Level III / IV	2	*3*	2	*4*	2	*10*
Level III / V	1	*2*	-	-	1	*5*
Level IV / V	5	*8*	3	*6*	1	*5*
Total	29	*48*	20	*39*	10	*49*

Disagreements between professionals and families were most frequent for MACS Levels II and III and particularly between physiotherapists and families for Levels I and II. There were disagreements of more than one level between families and physiotherapists for three children and between families and paediatricians for four children. In general, when families and professionals disagreed, families had classified their child one MACS Level lower than the professionals, which indicated the family thought the child had poorer manual ability than thought by professionals. Disagreement between health professionals by more than one MACS Level was also rare. One child classified by a physiotherapist at MACS Level I was classified by a paediatrician as Level III, and similarly, one child classified as MACS Level

II by a physiotherapist and occupational therapist was rated by the paediatrician as Level IV. There were no disagreements between occupational and physiotherapists.

7.3.3 Reliability

The reliability of family assessment of the MACS relative to each group was assessed using the Intraclass Correlation Coefficients (ICC) and is shown at the bottom of Tables 7.15 to 7.17 with 95% confidence intervals (CI). Tables 7.15 to 7.17 indicate that, although there was not complete agreement between families and professionals about the MACS level for every child, and by taking partial agreement into account, the correlation was very high.

Table 7.15 Comparison of MACS rating by Families and Physiotherapists

		Families					
		I	**II**	**III**	**IV**	**V**	**Total**
Physiotherapists	**I**	5	11	1			17
	II		10	6	1		17
	III		2	4	2		8
	IV				4	3	7
	V			1	2	7	10
	Total	5	23	12	9	10	59

ICC = 0.85 (95% CI from 0.80 to 0.88)

Table 7.16 Comparison of MACS ratings made by Families and Paediatricians

		Families					
		I	**II**	**III**	**IV**	**V**	**Total**
Paediatricians	**I**	5	1	2			8
	II		8	6	1		15
	III		4	4	2		12
	IV		1		4	1	6
	V				2	9	11
	Total	7	14	12	9	10	52

ICC = 0.79 (95% CI from 0.71 to 0.83)

Table 7.17 Comparison of MACS ratings made by Families and Occupational therapists

		Families					
		I	**II**	**III**	**IV**	**V**	**Total**
Occupational therapists	**I**	1	1				2
	II	1	7	4			12
	III			1	2		3
	IV				2	1	3
	V			1			1
	Total	2	8	6	4	1	21

ICC = 0.73 (95% CI from 0.60 to 0.82)

The reliability of family assessment relative to physiotherapists was ICC = 0.85 (95% CI from 0.80 to 0.88), relative to paediatricians was ICC = 0.79 (95% CI from 0.71 to 0.83), and relative to occupational therapists was ICC = 0.73 (95% CI from 0.60 to 0.82). The absolute and chance-corrected agreement, and reliability, between the different professional groups was also examined and is presented in Tables 7.18 to 7.20. Paediatricians and physiotherapists agreed exactly on 22/40 children's MACS Level (55%, κ =0.43); paediatricians and occupational therapists agreed exactly for 6/13 children (46%, κ =0.30), and occupational therapists and physiotherapists agreed for 14/14 children (100%, κ =1). The reliability of the professional assessments when compared to each other ranged from ICC=0.66 to 1. There were 10 children that had been classified by all three professions; however, there was insufficient variation in children's MACS Levels to make estimation of the reliability viable.

Table 7.18 Comparison of MACS ratings made by paediatricians and physiotherapists

		Paediatricians					
		I	**II**	**III**	**IV**	**V**	**Total**
Physiotherapists	**I**	4	1	1			6
	II	2	8	2	1		13
	III		3	3	2		8
	IV			1	1	4	6
	V				1	6	7
	Total	6	12	7	5	10	40

ICC=0.85 (95% CI from 0.78 to 0.89)

Table 7.19 Comparison of MACS ratings made by paediatricians and occupational therapists

		Paediatricians					
		I	**II**	**III**	**IV**	**V**	**Total**
Occupational therapists	**I**	1					1
	II	2	2	2	1		7
	III			3	1		4
	IV					1	1
	V						0
	Total	3	2	5	2	1	13

ICC=0.66 (95% CI from 0.33 to 0.77)

Table 7.20 Comparison of MACS ratings made by occupational therapists and physiotherapists

		Physiotherapists					
		I	**II**	**III**	**IV**	**V**	**Total**
Occupational therapists	**I**	2					2
	II		8				8
	III			2			2
	IV				1		1
	V					1	1
	Total	2	8	2	1	1	14

ICC=1

The means by which different health professionals made their classification of the MACS was monitored. Physiotherapists made 29/60 classifications by direct observation (48%), 3/60 used a mixture of observation and record review (5%) and 24/60 by reviewing the child's records (40%). Paediatricians used direct observation to classify 9/55 children (16%), the mixed method for 22/55 children (40%) and 24/55 classifications were made by reviewing the child's records (44%). Occupational therapists used direct observation to classify 5/23 children (22%), the mixed method for 4/23 children (17%) and 14/23 by reviewing the child's records (61%). The reliability between families versus professional report using the MACS was higher for physiotherapists, of whom a greater proportion indicated making their classification using direct observation. The reliability was slightly lower for paediatricians and occupational therapists, a greater proportion of whom had depended on the medical records to make their decision.

7.3.4 Summary of MACS performance

Overall family and professional assessments of the MACS were reasonably well correlated. There was not perfect agreement on the MACS Level for every child although disagreements by more than one level were rare. The reliability of family assessment for the MACS, indicated by the ICC, and the agreement corrected for chance expressed using the kappa coefficient, were comparable to the indices of inter-observer agreement and reliability between professionals. The reliability coefficients for family assessment of the MACS relative to each professional group are summarised in the Table 7.21 on the next page.

Table 7.21 Summary of indices of agreement and reliability of family and professional assessments for the MACS

Observers	n	% agreed	κ	ICC
Families / Physiotherapists	59	*51*	0.38	0.85
Families / Paediatricians	52	*58*	0.46	0.79
Families / Occupational therapists	21	*53*	0.32	0.73
Physiotherapists / Paediatricians	40	*55*	0.43	0.85
Paediatricians / Occupational therapists	13	*46*	0.30	0.66
Physiotherapists / Occupational therapists	14	*100*	1	1

The preceding sections have demonstrated the reliability of classifications made by families of children's GMFCS and MACS Levels. In effect, these were 'summary items' in the survey. The following sections examine aspects of the acceptability and reliability of the two longer questionnaires, the Activities Scale for Kids (ASK) and the Lifestyle Assessment Questionnaire (LAQ-CP). For these instruments the type of reliability of interest is not comparison between different observers, but whether the scales are internally consistent; that is, whether all the items in the scale are and measuring the same different aspects of the same attribute by which the scale is labelled.

7.4 ACTIVITIES SCALE FOR KIDS

The performance of the ASK was assessed in terms of how acceptable the instrument was to families, based on any comments made on the questionnaires, the frequency of responses to individual items and the internal consistency of the overall scale in this survey population. The ASK scale includes 38 items with the same five ordinal response categories used for all items, ranging from 'none of the time', which scores zero, to 'all of the time', which scores four. There were several children who scored zero for all items meaning they were unable to perform any of the activities listed in the questionnaire. Indeed, some of the families of those children commented that, in their opinion, the questionnaire was unsuitable for their child, for instance:

> "This was a really depressing thing to do. [He] is a great kid but he can't do any of the things listed except sitting unassisted."

> "Why does your questionnaire not have a box to tick at the beginning saying unable to do anything unaided to stop a parent having to face ticking 38 boxes with "*none of the time*" - not nice!"

However the majority of families did complete and return the questionnaire and many were completed by the children themselves albeit with some assistance; one child noted:

> "I found it fun filling in the questions hope it helps you with your study".

A non-scoring descriptive item in the ASK questionnaire requests children to state how much help they had when completing the form and a summary of responses to this question are shown in Table 7.22.

Table 7.22 Assistance given to children completing the Activities Scale for Kids

Amount of assistance given	n	%
Did all the questions themselves	22	*17*
Someone read them all the questions	23	*18*
Someone helped with some answers	22	*17*
Someone helped with most answers	21	*17*
Someone else completed all the questions	39	*31*

Respondents were able to indicate that some items were not applicable for them, in which case the item did not contribute to the child's ASK scale score and those children do not contribute to the item mean score. Most items yielded a small proportion of *not applicable* responses however a few items were particularly notable for the high proportion of *not applicable* responses such as 'running in a race' (47%), 'kept balance when playing rough games' (39%), 'climbing activities' (39%) and 'playing sport with competitive clubs' (52%). Notably, all the response options were use by participants and the frequency that responses were selected for each ASK item is shown in Table 7.23.

Table 7.23 Response frequencies to items in the Activities Scale for Kids (ASK)

	None of the time 0		Once in a while 1		Sometimes 2		Once in a while 3		All of the time 4		Mean score (sd)
	n	%	n	%	n	%	n	%	n	%	
Brushed teeth unaided	39	*31*	5	*4*	16	*13*	19	*15*	48	*38*	2.2 (1.7)
Used toilet unaided	42	*33*	2	*2*	1	*1*	17	*13*	64	*50*	2.4 (1.8)
Washed unaided	51	*40*	8	*6*	9	*7*	13	*10*	46	*36*	1.9 (1.8)
Put on shirt unaided	38	*30*	7	*6*	12	*9*	27	*21*	42	*33*	2.2 (1.7)
Put on trousers unaided	41	*32*	7	*6*	9	*7*	19	*15*	51	*40*	2.2 (1.7)
Fastened own clothes	51	*40*	5	*4*	11	*9*	33	*26*	24	*19*	1.7 (1.6)
Put on shoes unaided	61	*48*	9	*7*	13	*10*	11	*9*	32	*25*	1.5 (1.7)
Made a snack unaided	58	*45*	18	*14*	20	*16*	9	*7*	4	*3*	0.8 (1.1)
Did usual chores	38	*30*	22	*17*	18	*14*	8	*6*	10	*8*	0.9 (1.3)
Took care of medical needs	58	*45*	8	*6*	10	*8*	8	*6*	8	*6*	0.6 (1.2)
Did handwriting unaided	30	*24*	8	*6*	12	*9*	18	*14*	59	*47*	2.5 (1.7)
Did individual sports	41	*32*	8	*6*	11	*9*	23	*18*	42	*33*	2.1 (1.7)
Walked without any support	43	*34*	5	*4*	12	*9*	6	*5*	60	*47*	2.3 (1.8)
Got around inside unaided	24	*19*	2	*2*	10	*8*	15	*12*	76	*60*	2.9 (1.6)
Got around in crowded areas	29	*23*	3	*2*	13	*10*	17	*13*	59	*47*	2.5 (1.7)
Got around outside unaided	46	*36*	4	*3*	10	*8*	9	*7*	51	*40*	2.0 (1.8)
Negotiated a slope or hill	39	*31*	13	*10*	4	*3*	19	*15*	43	*34*	1.9 (1.8)
Negotiated rough ground	33	*26*	12	*9*	13	*10*	16	*13*	38	*30*	1.8 (1.7)
Kept up with friends outside	51	*40*	8	*6*	23	*18*	19	*15*	17	*13*	1.4 (1.5)
Carried a drink without spilling	37	*29*	6	*5*	20	*16*	30	*24*	21	*17*	1.7 (1.6)

Table 7.23 continued	**None of the time 0**		**Once in a while 1**		**Sometimes 2**		**Once in a while 3**		**All of the time 4**		**Mean score (sd)**
Carried objects in two hands	44	*35*	16	*13*	22	*17*	14	*11*	18	*14*	1.3 (1.5)
Stood still for 10 minutes	63	*50*	11	*9*	16	*13*	8	*6*	24	*19*	1.3 (1.6)
Stretched to reach a high shelf	29	*23*	18	*14*	21	*17*	7	*6*	24	*19*	1.4 (1.5)
Negotiated heavy doors unaided	44	*35*	6	*5*	21	*17*	22	*17*	31	*24*	1.8 (1.6)
Negotiated stairs unaided	43	*34*	5	*4*	9	*7*	16	*13*	45	*35*	1.9 (1.8)
Got in and out of a car unaided	43	*34*	4	*3*	11	*9*	22	*17*	45	*35*	2.1 (1.7)
Got in and out of a chair unaided	26	*21*	2	*2*	9	*7*	13	*10*	75	*59*	2.8 (1.7)
Sat on the floor	24	*19*	2	*2*	16	*13*	15	*12*	65	*51*	2.6 (1.6)
Got in and out bed unaided	29	*23*	2	*2*	1	*1*	14	*11*	79	*62*	2.8 (1.7)
Got down to the ground and up again	38	*30*	3	*2*	3	*2*	22	*17*	57	*45*	2.3 (1.8)
Did climbing activities	45	*35*	6	*5*	13	*10*	4	*3*	9	*7*	0.6 (1.1)
Played team sports	27	*21*	7	*6*	16	*13*	12	*9*	34	*27*	1.6 (1.7)
Played sports with friends	22	*17*	15	*12*	29	*23*	14	*11*	30	*24*	1.8 (1.5)
Played sports with competitive clubs	43	*34*	4	*3*	4	*3*	2	*2*	61	*48*	0.4 (1.1)
Kept balance when playing games	40	*32*	5	*4*	13	*10*	8	*6*	11	*9*	0.8 (1.3)
Did not tire during activities	26	*21*	5	*4*	26	*21*	30	*24*	16	*13*	1.7 (1.5)
Ran in a race	30	*24*	10	*8*	11	*9*	16	*5*	11	*9*	0.7 (1.3)
Worked carefully with hands	27	*21*	21	*17*	27	*21*	14	*11*	22	*17*	1.6 (1.5)

Approximately a third of the children performed most of the activities *none of the time*. Very few children were taking care of their own medical needs and half of the children never put their shoes on or made a snack unaided and could not stand still for ten minutes. However, more than half the children did get around the house unaided *all of the time* and were also able to get out of bed, in and out of chairs, and use the toilet unaided *all of the time*. A third of the children never managed to negotiate stairs or heavy doors. Less than a fifth of children carried a drink without spilling or carried an object with two hands *all of the time* while a third never did these activities. A smaller proportion of children were able to get around outside unaided than inside *all of the time* and this reduced to a third for activities where a slope or rough ground was encountered. Very few children participated in climbing activities. A third of children were taking part in sports they enjoyed on their own and half were participating in sports with competitive clubs, although, aside from the physical problems, one family also noted other difficulties in performing this particular activity:

> "We can only find one disabled athletics club within a 50 mile radius, which is what she wants to do"

Individual item scores ranged from zero to four and the mean score for items ranged from 0.4 to 2.9. The items with the lowest mean scores, indicating that children in this survey rarely participated in these activities, were 'sport with competitive clubs' (mean = 0.4), 'took care of my medical needs' (mean = 0.6) and 'climbing activities' (mean = 0.5). The items with the highest mean scores, indicating that more children in the survey participated in these activities more of the time were 'getting around inside' (mean = 2.9), and 'getting in and out of bed' (mean = 2.8).

Children's individual scores ranged across the standardised scale from zero to 92; the scale mean was 67 with a standard deviation of 46. In terms of the homogeneity of the scale, that is whether all the items were measuring the same construct, items with the lowest item-total (I-T) correlations were 'playing sport with competitive clubs' (I-T = 0.40) and 'took care of my medical needs' (I-T = 0.38). The Cronbach's alpha statistic was 0.98, indicating that the scale was highly internally consistent, although the figure is naturally inflated by the large number (38) of items in the scale.

ASK summary

Based on completion rates and comments added by respondents at the end of the ASK questionnaire was generally acceptable to families who participated in this survey, except for those children with the least physical abilities who are totally dependent on assistance for all of the activities listed. All the available response options in the item scales were utilised by participants. The scale was therefore able to discriminate between children's different activity performance and participation resulting in a broad range and distribution of scores. Descriptively, information from the ASK indicates that most children were independent for typical activities in the house but fewer children achieved independence all the time outside the house, while about a third were dependent for most activities.

7.5 LIFESTYLE ASSESSMENT QUESTIONNAIRE (LAQ-CP)

Based only on the comments added at the end of the questionnaire, the LAQ-CP appeared generally acceptable to families, although one family thought the overall tone of the questionnaire was pessimistic:

> "It all comes across as so negative - we love [her] and I chose to care for her myself. These qualitative aspects are important for really understanding what its like for families"

The LAQ-CP includes 16 items that have five incremental scores (0 to 4), 16 items have three increments (0, 2 or 4), and five items are scored dichotomously (0 or 4). However the scaling is opposite to the ASK, such that higher scores for the LAQ-CP indicate increasing activity limitations and participation restrictions. The descriptions used to represent different response categories varied as appropriate for each item. The performance of each item and each of the six domains of the LAQ-CP was examined separately as described below.

7.5.1 LAQ-CP Physical Independence

The physical independence domain comprises 12 items and the distribution of responses to each item is shown in Table 7.24. As children scoring zero are the most independent, the table shows that around 60% of children were able to get out of bed and eat cereal unaided, and just less than half could wash, dress and go to the toilet without assistance. Children needed most help to fasten buttons or zips and to get out of the bath; over a quarter of the children needed all the activities in the items to be done for them. Approximately half of the children needed to be lifted for some activities. As well as frequent assistance in the daytime, around a third of the children also needed regular assistance at night.

The average mean score for items in the physical independence domain was 1.7, range from 1.2 to 2.7. The item with the lowest mean was "getting out of bed" (mean = 1.2) indicating, as did the ASK, that this activity is completed by most of the survey population although a quarter were completely dependent on assistance for this activity. The item with the highest mean was "doing up buttons or buckles" (mean 2.7) indicating that most children required help with this activity.

Table 7.24 Response frequencies to items in LAQ-CP Physical Independence domain.

	Least limitation				**Item score**			*Most limitation*			**Mean score**
	0		**1**		**2**		**3**		**4**		
Physical Independence items	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**(sd)**
Washing hands	59	*46*	x	*x*	37	*29*	x	*x*	33	*26*	1.6 (1.6)
Eating cereal	77	*60*	x	*x*	24	*19*	x	*x*	28	*22*	1.2 (1.6)
Putting on T-shirt	62	*48*	x	*x*	27	*21*	x	*x*	40	*31*	1.7 (1.8)
Doing up buttons	20	*16*	x	*x*	44	*34*	x	*x*	65	*50*	2.7 (1.5)
Getting out of bed	81	*63*	x	*x*	16	*12*	x	*x*	32	*25*	1.2 (1.7)
Getting out of bath	29	*23*	x	*x*	48	*37*	x	*x*	52	*40*	2.4 (1.6)
Going to toilet	56	*43*	x	*x*	34	*26*	x	*x*	39	*30*	1.7 (1.7)
Climbing stairs	55	*43*	x	*x*	40	*31*	x	*x*	34	*26*	1.7 (1.6)
Getting in/out of car	48	*37*	x	*x*	43	*33*	x	*x*	38	*30*	1.8 (1.6)
Lifting needs	60	*47*	6	*5*	22	*17*	18	*14*	23	*18*	1.5 (1.6)
Night time assistance	47	*36*	37	*29*	22	*17*	8	*6*	15	*12*	1.3 (1.3)
Dependence in day	43	*33*	12	*9*	14	*11*	40	*31*	20	*16*	1.9 (1.5)

x = response option not available

The standardised scale scores ranged from zero to 100 and the mean was 43 (standard deviation, 34). The item with the lowest item-total correlation was 'dependence in day' which asked families to "list areas where your child needs assistance" (I-T = 0.5), however this value suggests that the item is consistent with the construct measured by this scale. The Cronbach alpha statistic of 0.96 indicated the scale was highly internally consistent.

7.5.2 LAQ-CP Mobility

The mobility domain includes seven items and the distribution of responses to each item is shown in Table 7.25. Nearly 70% of children accessed most rooms in their houses and a half could get around at home without help; half of the children also needed no assistance to get in or out of the house. Despite these abilities around 70% of children did not go out unaided.

The average item mean was 2.1, range from 0.8 to 3.5. The item with the lowest mean score asked "how many rooms in the house the child had accessed in the past week" (mean = 0.8). The item with the highest mean score asked "how many times your child has been out of the house by themselves" indicating that fewer children participated in this activity. The standardised scale scores ranged from seven to 93 out of 100 and the mean was 52 (standard deviation, 25). The item asking "how many times your child has been on a longer outing requiring transport" produced an item-total correlation of minus 0.12 suggesting that the item was least associated with the construct measured by this scale. However the Cronbach alpha statistic of 0.83 indicates that overall the scale is highly internally consistent.

Table 7.25 Response frequencies to items in LAQ-CP Mobility domain.

	Least limitation				**Item score**			*Most limitation*			**Mean score**
	0		**1**		**2**		**3**		**4**		
Mobility items	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**(sd)**
Mobile hand function	31	*24*	23	*18*	29	*23*	18	*14*	28	*22*	1.9 (1.5)
Getting around home	72	*56*	23	*18*	27	*21*	7	*5*	-	-	0.7 (1.0)
Accessing rooms at home independently	89	*69*	1	*1*	5	*4*	3	*2*	31	*24*	1.1 (1.7)
Assistance to go outside	69	*54*	x	*x*	x	*x*	x	*x*	60	*47*	1.9 (2.0)
Distance travelled outside unaided	27	*21*	7	*5*	17	*13*	23	*18*	55	*43*	2.6 (1.6)
Frequency outside unaided	5	*4*	4	*3*	5	*4*	28	*22*	87	*67*	3.5 (1.0)
Outings requiring transport	-	-	6	*5*	21	*16*	70	*54*	32	*25*	3.0 (0.8)

x = response option not available

7.5.3 LAQ-CP Clinical Burden

The clinical burden domain includes 12 items and the distribution of responses to each item is shown in Table 7.26. In the previous year half of the children had been seen by up to five doctors apart from their school doctor, a quarter had to spend time in hospital, a fifth had an operation of some kind. No children had been in hospital for more than 26 weeks or plaster for more than 12 weeks, and none had undergone more than two operations. A fifth of the children had spent a few weeks with some part of their body in plaster and half of children in this study regularly used some form of orthosis. A quarter of children were taking up to four medicinal doses in one day and a fifth were taking more. Around 15% of the children had experienced seizures occasionally or had regular fits and nearly a fifth had been seen by a

behavioural specialist. Half of the children saw a physiotherapist once a month and a quarter of families were receiving other types of personal support from health or social services.

Table 7.26 Response frequencies to items in LAQ-CP Clinical Burden domain.

	Least burden				**Item score**			*Most burden*			**Mean score**
	0		**1**		**2**		**3**		**4**		
Clinical Burden items	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**(sd)**
Seen a doctor	10	*8*	32	*25*	70	*54*	12	*9*	5	*4*	1.8 (0.9)
Stay in hospital	95	*74*	18	*14*	15	*12*	1	*1*	-	-	0.4 (0.7)
Number of operations	104	*81*	17	*13*	8	*6*	-	-	-	-	0.3 (0.6)
Time spent in plaster	106	*82*	18	*14*	5	*4*	-	-	-	-	0.2 (0.5)
Use of orthoses	50	*39*	8	*6*	4	*3*	15	*12*	52	*40*	2.1 (1.8)
Number of medicines	85	*66*	29	*23*	7	*5*	6	*5*	2	*2*	0.5 (0.9)
Seizures per year	110	*85*	10	*8*	6	*5*	2	*2*	1	*1*	0.2 (0.7)
Behavioural specialist	106	*82*	x	*x*	x	*x*	x	*x*	23	*18*	0.7 (1.5)
Physiotherapy	16	*12*	9	*7*	63	*49*	31	*24*	10	*8*	2.1 (1.1)
Other services	91	*71*	x	*x*	34	*26*	x	*x*	4	*3*	0.7 (1.1)
Any special equipment	40	*31*	47	*36*	30	*23*	7	*5*	5	*4*	1.1 (1.0)

x = response option not available

The average mean score for items in the clinical burden domain was 1.0, range from 0.2 to 2.1. The items with the lowest means asked "how many weeks your child has spent in plaster this year" (mean = 0.2) and "how many fits or blackouts has your child suffered" (mean = 0.2). The higher means were associated with the items asking about frequency of

"using body or leg support" (orthoses) and "seeing a therapist" (both means = 2.1) indicating these were relatively common interventions. The standardised scale scores ranged from zero to 55 out of 100, hence the scale mean was low at 23 (standard deviation, 12). The item asking whether the child was being "seen by a specialist about their behaviour" had the lowest item-total correlation (I-T = 0.02) indicating this item was least associated with the construct measured by this scale. The Cronbach alpha statistic of 0.69 indicated that the scale has adequate internal consistency.

7.5.4 LAQ-CP Schooling

The schooling domain has only two items which are shown with the distribution of responses in Table 7.27. With only two items the domain was not suitable for analysis as a scale but does provide useful descriptive information. The first item shows that a third of the survey population (33%) are attending special schools for children with physical or learning disabilities; the majority attend mainstream schools either with a special support unit (23%) or without (41%). There were a few children that attended boarding schools (4%) but none were being taught at home.

Table 7.27 Response frequencies to the items in LAQ-CP Schooling domain.

	Least exclusion				**Item score**				*Most exclusion*		**Mean score**
	0		**1**		**2**		**3**		**4**		
Schooling items	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**(sd)**
School attended	53	41	29	23	42	33	-	-	5	4	1.0 (1.0)
Time to get to school	64	50	35	27	14	11	9	7	7	5	0.9 (1.2)

x = response option not available

The second item shows that it takes half of the survey population less than 15 minutes to travel to school (50%); another quarter take less than 30 minutes (27%) while 18% spend between 30 minutes to an hour travelling. The families spending more than one hour travelling to school (5%) include those children using boarding schools and three other families: one uses a mainstream school with a special support unit and two children are in schools for children with physical or learning disabilities.

7.5.5 LAQ-CP Economic Burden

The economic burden domain includes six items which are shown with the distribution of responses in Table 7.28. Half of the families had incurred costs purchasing special equipment and other commodities not covered by grants or allowances in the previous year. Nearly a half of carers had made major and permanent changes to their employment, predominantly to reduce working hours in order to care for the child. A fifth of families had made changes to their houses and a third planned to make adaptations in the next year. A fifth of children were eating some form of special diet.

The average item mean was 1.0, range from 0.3 to 1.7. The items with the lowest means asked "how many adaptations need to be made to the house because of your child", either completed in the past year (mean = 0.3) or planned (mean = 0.4). The highest item mean in the economic burden domain corresponded to whether anyone had changed their employment situation to care for their child (mean = 1.7). The children's standardised scale scores ranged from zero to 83 out of 100, however the scale mean was low, 24 (standard deviation, 23). The item asking "is your child is receiving any special diet" showed the lowest item-total correlation (I-T = 0.31) suggesting it was the item least associated with the

construct measured by this scale. The Cronbach alpha statistic of 0.68 indicated that the scale has good internal consistency.

Table 7.28 Response frequencies to items in LAQ-CP Economic Burden domain.

	Least burden				**Item score**			*Most burden*			**Mean score**
	0		**1**		**2**		**3**		**4**		
Economic Burden items	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**(sd)**
Any special diet	105	81	x	x	x	x	x	x	24	19	1.2 (1.6)
Cost of equipment	68	53	27	21	3	2	4	3	27	21	1.5 (1.7)
Other costs	62	48	18	14	10	8	2	2	37	29	0.2 (0.6)
Housing adaptations	105	81	20	16	1	1	2	2	1	1	0.4 (0.7)
Planned adaptations	91	71	33	26	2	2	1	1	2	1	0.7 (1.6)
Change to employment	72	56	x	x	4	3	x	x	53	41	1.7 (2.0)

x = response option not available

7.5.6 LAQ-CP Social Integration

The social integration domain includes 8 items which are shown with the distribution of responses in Table 7.29. Nearly three quarters of children were residing with both birth parents and a quarter with at least one birth parent, and four children were not with either birth parent, note that these are not the children at boarding school. Of the five children at boarding school, four were 'residing' with both parents and one with one birth parent. A third of children did not see any friends outside of school hours but this was not related to

how far they had to travel to school. A quarter of carers reported they did not have anyone locally they could turn to for help and only a half of families felt that local people were always understanding about their child. Two thirds of respondents felt that their social lives had been adversely affected and a half said that family holidays were difficult to organise because of having a child with a disability. The majority of families indicated that the child with cerebral palsy had increased stress on the carers and two thirds also indicated that it had also increased stress on the child's siblings.

Table 7.29 Response frequencies to items in LAQ-CP Social Integration domain.

	Least limitation				**Item score**			*Most limitation*			**Mean score**
	0		**1**		**2**		**3**		**4**		
Social Integration items	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**	**(sd)**
Number of friends seen	2	2	3	2	13	10	67	52	44	34	3.1 (0.8)
Local help available	100	76	x	x	x	x	x	x	29	23	0.9 (1.7)
Local people understanding	66	51	x	x	51	40	x	x	12	9	1.2 (1.3)
Effect on social life	44	34	x	x	37	29	x	x	48	37	2.1 (1.7)
Effect on holidays	72	56	x	x	x	x	x	x	57	44	1.8 (2.0)
Residing with birth parents	93	72	x	x	32	25	x	x	4	3	0.6 (1.1)
Stress on parents	20	16	x	x	71	55	x	x	38	30	2.3 (1.3)
Stress on siblings	42	33	x	x	63	49	x	x	24	19	1.7 (1.4)

x = response option not available

The average item mean was 1.7, and ranged from 0.6 to 3.1. The item with the lowest mean asked "is the child living with their natural parents". The item that asked "is there someone locally to whom you can turn for help" also had a relatively low mean (0.9) as most

respondents did have someone locally to help. The standardised scale score ranged from three to 88 out of 100, the mean scale score was 43 (standard deviation, 21). The item about whether the carers were the natural parents showed the lowest item-total correlation (I-T = 0.01) suggesting this item was least associated with the construct measured by this scale. The Cronbach alpha statistic of 0.73 indicates that overall the scale has good internal consistency.

The questions asking about any increased stress on parents and siblings were the final items on the questionnaire and placed before a space inviting any further comments. Most of the comments noted by families therefore related to these last items. The response options were 'none, mild or severe' and at least one respondent found these difficult terms to interpret:

> "Severe is an overstatement and an unkind word for a parent to use when discussing their child, however slight is also not accurate"

Some respondents emphasised how dependent they were for support from extended family and respite centres, while some expressed that the support they had received was inadequate:

> "There needs to be much more help given to parents of sick, disabled children - radical help, not superficial stuff. The burden of responsibility needs to be shared more widely as early in the child's life as possible so that the parent and sibling do not end up crushed under it"

Other families identified their strategies for managing stress on siblings by giving them protected time, but this can have downsides too as one respondent identified:

> "I make sure [sibling] has special time without his brother to enable him to do normal things a little boy would do. This often means we have to split up as a family rather than doing all these things together"

One parent noted that having a sibling with a disability had positively influenced another of their children towards a career in education, and especially for children with special needs.

7.5.7 LAQ-CP summary

In general, by the high completion rates for all items and comments added by respondents, the LAQ-CP appeared acceptable to families although the items measuring stress on parents or siblings created some criticism. Only a few response options remained unused by any respondents in the survey. These were predominantly in the clinical burden domain because no children had been in hospital for more than 26 weeks or plaster for more than 12 weeks, and none had undergone more than two operations in the previous year. There were also no children in the survey who were being educated at home rather than at a school, or who had gone on more than 10 outings requiring transport in the previous week. All the domain scales performed acceptably in terms of reliability although they were skewed towards lower mean scores in the clinical and economic burden domains. The performance of the LAQ in this survey is summarised in Table6.39.

Table 7.30 Summary of the reliability of the LAQ-CP domains

Domain	**Number of items**	**Standardised mean *(sd)***	**α**
Physical independence	12	43 *(34)*	0.96
Mobility	7	52 *(25)*	0.83
Clinical burden	12	23 *(12)*	0.69
Schooling	2	-	-
Economic burden	6	24 *(23)*	0.68
Social integration	8	42 *(21)*	0.73

7.6 CONSTRUCT VALIDITY

The construct validity of the instruments used in the survey was assessed by examining correlations between the GMFCS, MACS, ASK score and each of the LAQ-CP domain and summary scores, as shown in Tables 7.31. Negative correlations occurred between the ASK and the other variables as higher ASK scores indicate better functioning whereas higher scores on the LAQ-CP, and levels of the GMFCS and MACS indicate greater limitations. High levels of correlation were found between the GMFCS, ASK and the LAQ-CP domains for physical independence and mobility (all $r \geq 0.9$, $p<0.01$). There was also a high correlation between these variables and the LAQ-CP summary score, as more weight is given to the physical independence and mobility domain scores in its derivation. These variables all represent constructs related to coordination and movement and there is strong evidence for convergent validity.

The MACS was most highly correlated with the LAQ-CP domain for physical independence ($r = 0.83$, $p<0.01$), and also moderately with the ASK ($r = -0.76$, $p<0.01$). As both of these scales include several items that involve handling objects there is evidence of convergent validity. The correlation between the MACS and GMFCS was moderate (tau-b=0.62, $p<0.01$).

The lowest correlations were between clinical burden and social integration and all the other variables. Social integration and clinical burden scores were not strongly correlated with any of the other variables (all $r \leq 0.60$) except social integration and LAS ($r = 0.72$). Therefore, there is divergent validity that clinical burden and social integration scores are not necessarily related to each other, to economic burden or to the severity of a child's movement or manual disability.

Table 7.31 Correlations between the products of family-assessed instruments.

	GMFCS	MACS	ASK	LAQ-CP Physical Independence	Mobility	Clinical Burden	Economic Burden	Social Integration
MACS	0.62[a]							
ASK	-0.90	-0.76						
LAQ-CP Physical Independence	0.89	0.83	-0.95					
Mobility	0.87	0.72	-0.92	0.88				
Clinical Burden	0.53	0.46	-0.60	0.59	0.60			
Economic Burden	0.67	0.66	-0.70	0.70	0.65	0.52		
Social Integration	0.53	0.50	-0.58	0.56	0.56	0.42	0.61	
Summary score	0.90	0.80	-0.96	0.93	0.93	0.62	0.77	0.72

All values Pearson correlations except [a]Kendall's tau-b; all correlations $p<0.01$

GMFCS *Gross Motor Function Classification System*; **MACS** *Manual Ability Classification System;* **ASK** *Activities Scale for Kids;* **LAQ-CP** *Lifestyle Assessment Questionnaire*

Classifications of the GMFCS in this survey were compared with children's 'Lower limb function' recorded by *4Child*; and likewise classifications of the MACS were compared with children's 'Upper limb function' recorded by *4Child*. As it is the paediatricians who provide *4Child* with their information the comparison of the paediatrician's classifications made using each system are shown in Tables 7.32, and 7.33. The association between the GMFCS and Lower limb function was reasonably good for 73 children (tau-b=0.77, $p<0.001$)

however the association between the MACS and Upper limb function for 49 children was moderate (tau-b=0.54, $p<0.001$).

Table 7.32 Comparison of classifications made by paediatricians using the GMFCS and 4Child's Lower limb function variable.

	GMFCS (paediatrician)					
Lower limb Function (*4Child*)	I	II	III	IV	V	Total
Walks fluently	4					4
Functional, not fluent	16	9	2	1		28
Restricting lifestyle	1	2	7	1		11
Not walking		2	2	9	17	30
Total	21	13	11	11	17	73

Kendall's tau-b = 0.77 ($p<0.001$)

Table 7.33 Comparison of classifications made by paediatricians using the MACS and 4Child's Upper limb function variable.

	MACS (paediatrician)					
Upper limb Function (*4Child*)	I	II	III	IV	V	Total
No problem	2	2	1	1		6
Some difficulty	6	5	3	1		15
Able to feed self	1	5	7	1	1	15
Unable to feed self		1	2	2	8	13
Total	9	13	13	5	9	49

Kendall's tau-b = 0.54 ($p<0.001$)

Comparisons of classifications made by families and the *4Child* variables for upper and lower limb function are shown in Tables 7.34, and 7.35. The association between classifications made by families using the GMFCS and *4Child's* Lower limb function for 168 children was again fairly good (tau-b=0.76, $p<0.001$), and the association between the MACS and Upper limb function for 84 children was less good (tau-b=0.62, $p<0.001$). Note the large number of children who were classified by families using the GMFCS available from the survey for this analysis.

Table 7.34 Comparison of classifications made by families using the GMFCS and 4Child's Lower limb function variable.

	GMFCS (family)					
Lower limb Function (*4Child*)	I	II	III	IV	V	Total
Walks fluently	13	5				18
Functional, not fluent	26	38	3	1		68
Restricting lifestyle	2	9	8	4	1	24
Not walking		1	6	12	39	58
Total	41	53	17	17	40	168

Kendall's tau-b = 0.76 ($p<0.001$)

Table 7.35 Comparison of classifications made by families using the MACS and 4Child's Upper limb function variable.

	MACS (family)					
Upper limb Function (*4Child*)	I	II	III	IV	V	Total
No problem	5	7	1			13
Some difficulty	7	15	7	1		30
Able to feed self		7	8	4	1	20
Unable to feed self	1	1	1	7	11	21
Total	13	30	17	12	12	84

Kendall's tau-b = 0.62 ($p<0.001$)

7.7 SUMMARY OF PERFORMANCE OF THE SURVEY INSTRUMENTS

The family-assessed instruments used in the survey to measure children abilities, activities and participation were the GMFCS, MACS, ASK and LAQ-CP. Agreement between classifications made by families and health professionals was examined using indices of absolute and chance-corrected agreement (κ), and the Intraclass Correlation Coefficient (ICC) as an appropriate index of reliability for ordinal data. Family assessment for both the GMFCS and MACS showed good correlation with professional assessments, equivalent to that found between professionals in this study. Although there was not always perfect agreement between observers the indices of agreement corrected for chance were fair to good and the indices of reliability were excellent. In general there was better concordance with the classifications made by families when professional assessments were made by direct observation rather than using the child's records. The reliability between the initial and retest family-assessed GMFCS classifications three months apart were exceptionally high.

Families of children who were dependent on assistance for all the activities listed in the ASK indicated strongly that the questionnaire was not appropriate for them to complete. However all the other respondents appeared to find the ASK acceptable, and some of the children even stated that they had enjoyed answering the questions. The scale was found to be internally consistent although the high number of items naturally inflates the way this parameter is estimated. All the response options for all the items were utilised indicating that the items and resulting scale clearly discriminated between children with different activities and participation. In general, most children were able to get around and complete activities in the house, while only a third of the children were independent for activities outside, and especially when the terrain was more challenging.

The LAQ-CP appeared generally acceptable to families, however, two items that assess the severity of any stress on parents or siblings from having a child with cerebral palsy were commented upon by several families. Some comments were critical of the descriptions used for the response options, while others commented generally on the importance of support for families. Only a few response options to items in the LAQ-CP remained unused by any participants in the survey; most domains had a centrally positioned mean score and a broad distribution of scores across the scales. However, the mean scores for clinical and economic burden were shifted towards the lower end of the scales, indicative of lower burden in this survey. All the domain scales were found to have good internal consistency.

There was strong evidence of convergent validity between the classifications and scores representing aspects of co-ordination and movement. There was some evidence of divergent validity in that clinical burden and social integration scores were not associated with each other or with the severity of children's disability, as described by the GMFCS and MACS. The GMFCS and MACS were only moderately associated with each other. There was a fairly good association between *4Child's* measure of children's lower limb function and the GMFCS and but less so between the MACS and *4Child's* measure of upper limb function.

In summary, all the family-assessed instruments have been shown to be valid and reliable, and information from the ASK and LAQ-CP suggests that children and families experience a wide range of activity limitations and participation restrictions. The following section examines to what extent the indices of children's activities and participation can be explained using information available about their impairments and environment.

CHAPTER 8

THAMES VALLEY SURVEY RESULTS III:

FACTORS AFFECTING

CHILDREN'S ACTIVITIES AND PARTICIPATION

8.1 CHAPTER OVERVIEW

This chapter describes statistical analyses examining associations between the family-assessed outcomes of children's activities and participation and the available explanatory variables, children's abilities and impairments related to their cerebral palsy. The adequacy of each of the derived models was assessed by examining the distribution of plots of the predicted and observed residual values. The chapter concludes with a summary of what these models tell us about the effect of disability on children's activities and participation.

8.2 OUTCOME AND EXPLANATORY VARIABLES

Outcome variables

The outcome variables represent aspects of children's activities and participation ascertained using the ASK and LAQ-CP. Most of the outcome variables are presented as standardised scale scores ranging from zero to 100. Whilst these scales do not provide truly continuous data, it has been recommended that they can be assumed to approximate continuous data for the purpose of statistical analysis (quasi-continuous) (Streiner and Norman 2003). Note that higher scores for the ASK indicate greater participation whereas higher scores for the LAQ scales represent reduced participation. The schooling domain of the LAQ did not have enough items to function reliably as a scale. These data were therefore recoded to describe whether the child attended a mainstream school or some other type, including schools for children with profound physical or learning disability and boarding schools. A summary of the outcome variables is shown in Table 8.1 on the next page.

Table 8.1 Outcome variables representing children's activities and participation

Outcome variables	Type	Range	Source
ASK score	Continuous	0 – 100	Family survey
LAQ-CP Physical Independence	Continuous	0 – 100	Family survey
LAQ-CP Mobility	Continuous	0 – 100	Family survey
LAQ-CP Clinical Burden	Continuous	0 – 100	Family survey
LAQ-CP Educational exclusion	Nominal	Mainstream / special school	Family survey
LAQ-CP Economic	Continuous	0 – 100	Family survey
LAQ-CP Social integration	Continuous	0 – 100	Family survey
LAQ-CP summary score	Continuous	0 – 100	Family survey

Explanatory variables

The following explanatory variables were selected from information on impairments recorded in the *4Child* database, from the family survey or by record linkage between databases.

(i) Type of cerebral palsy: the type of cerebral palsy was coded as hemiplegia or bilateral. The majority of children with spastic cerebral palsy (84%) were already coded in this way, however three children with three limb cerebral palsy, and twenty one children with dyskinetic, ataxic or unclassifiable types of cerebral palsy were all assumed, for pragmatic reasons, to be bilateral although this may not have actually been the case.

(ii) Hearing and vision loss: children were defined as having hearing loss or vision loss using the following definitions: Hearing loss was sensorineural loss of 50dB or more averaged

across the range 0.5 to 4 kHz in the better ear. In the absence of a pure-tone audiogram, all children with a hearing aid fitted for sensorineural loss were included. Vision loss referred to visual acuity in the better eye of 6/18 or less; if visual acuity could not be measured, an assessment of the degree of visual impairment was made on the behavioural responses of the child.

(iii) Intellectual delay: intellectual delay was recorded by *4Child* in the categories of no delay, IQ more than 70, between 50 and 69, or less than 50. However, in preliminary analyses there was little difference between the effects of 'no delay' and 'IQ more than 70' so these were merged as a single group.

(iv) Seizures and speech problems: whether the child had a speech problem or was having seizures when the child was aged 5 years was recorded on the 4Child database.

(v) Movement and manual ability: indices of children's movement and manual abilities were the families' assessments of children's GMFCS and MACS Levels reported in the survey.

(vi) Area deprivation: indices of deprivation in the area where the family were living during the study were derived by record linkage using families' current post code and the IMD national quintiles. Due the small number of children in the most deprived national quintiles, quintile IV and V were merged into a single category. A summary of all the explanatory variables is shown in Table 8.2.

Univariate correlations between outcome and explanatory variables are presented graphically as scatter plots for continuous explanatory variables (age) and in box plots for ordinal

explanatory variables. The boxes in these plots represent the middle 50% of the observations (inter-quartile range) and the median value is marked. The lines, or whiskers, extend to indicate 1.5 times the inter-quartile range. Any values outside these are shown as outliers. The box plots enable the variability of the unadjusted correlation between outcome and explanatory variables to be inspected. Explanatory variables that showed a significant correlation ($p \leq 0.1$) in univariate analyses with outcome variables were eligible for inclusion in the subsequent multivariate analysis.

Table 8.2 Explanatory variables that may affect children's activities and participation

Explanatory variables	Type	Range	Source
Age	Continuous	6 – 12 years	*4Child*
Gender	Nominal	male / female	*4Child*
Type of cerebral palsy	Nominal	hemiplegia / bilateral	*4Child*
Hearing impairment *	Nominal	yes / no	*4Child*
Vision impairment **	Nominal	yes / no	*4Child*
Intellectual delay	Ordinal	IQ <50 / 50-69 / >70	*4Child*
Seizures	Nominal	yes / no	*4Child*
Speech	Nominal	yes / no	*4Child*
GMFCS Level	Ordinal	I - V	Family survey
MACS Level	Ordinal	I - V	Family survey
IMD 2004	Ordinal	I – IV/V	Postcode/ODPM

IMD = Indices of Multiple Deprivation (Office of the Deputy Prime Minister): national quintiles. *Hearing loss was sensorineural loss of 50dB or more averaged across the range 0.5 to 4 kHz in the better ear. In the absence of a pure-tone audiogram, all children with a hearing aid fitted for sensorineural loss were included. **Vision loss referred to visual acuity in the better eye of 6/18 or less; if visual acuity could not be measured, an assessment of the degree of visual impairment was made on the behavioural responses of the child.

8.3 FACTORS AFFECTING ACTIVITIES SCALE FOR KIDS (ASK) SCORE

The crude effects of each explanatory variable are shown in Table 8.3 and Figures 8.1 to 8.4.

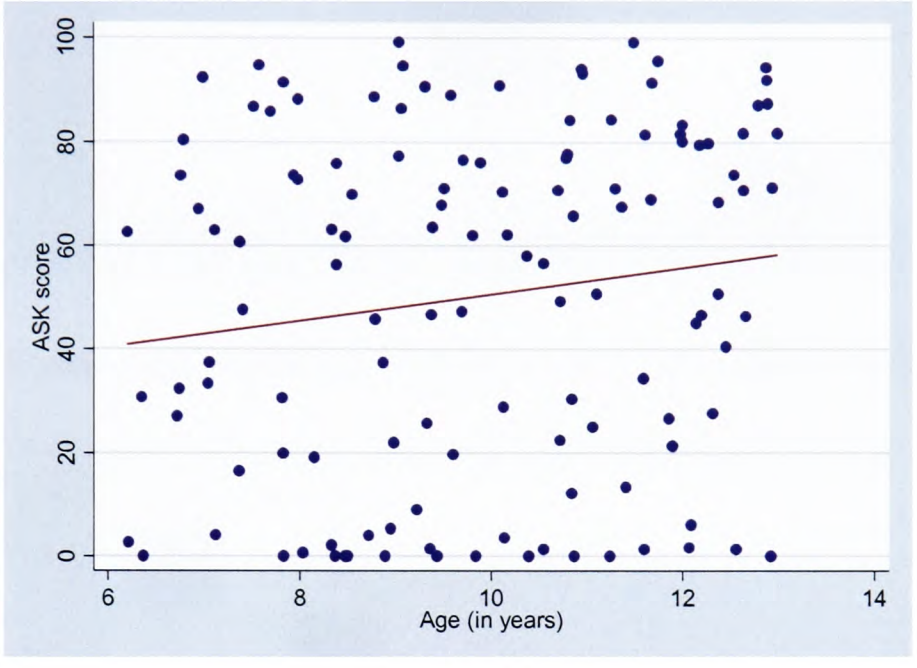
Table 8.3 Crude effect of explanatory variables on Activities Scale for Kids score

Variable	Levels	Number of cases	Crude effect	R^2_{adj}	*p*
Age	per year	129	3	0.01	0.10
Gender	female	57	*		
	male	72	-8	0.01	0.17
Type of cerebral palsy	hemiplegia	46	*		
	bilateral	83	-38	0.30	<0.01
Hearing loss	not deaf	124	*		
	Hearing loss	5	-28	0.02	0.06
Vision loss	not blind	104	*		
	Vision loss	25	-53	0.38	<0.01
Intellectual delay	no delay/IQ>70	74	*		
	IQ 50-69	13	-23	-	<0.01
	IQ<50	26	-60	0.56	<0.01
Seizures	no seizures	93	*		
	Seizures	28	-21	0.07	<0.01
Speech problem	no speech problem	78	*		
	Speech problem	43	-32	0.21	<0.01
GMFCS Level	GMFCS Level I	25	*		
	GMFCS Level II	43	-21	-	<0.01
	GMFCS Level III	15	-44	-	<0.01
	GMFCS Level IV	14	-68	-	<0.01
	GMFCS Level V	23	-84	0.82	<0.01
MACS Level	MACS Level I	14	*		
	MACS Level II	30	-3	-	0.64
	MACS Level III	18	-28	-	<0.01
	MACS Level IV	13	-59	-	<0.01
	MACS Level V	13	-72	0.60	<0.01
IMD 2004	Quintile I	67	*		
	Quintile II	23	13	-	0.10
	Quintile III	17	-1	-	0.90
	Quintiles IV/V	20	-16	0.04	0.05

*Baseline against which other levels of the same impairment are compared

There was a small increase in children's ASK score for each year as children got older (Figure 8.1) indicating, as one would expect, increasing ability with increasing age, but the small reduction in scores for males compared to females was not statistically significant.

Figure 8.1 Scatter plot showing relationship between children's age and ASK score



Children with bilateral cerebral palsy scored lower on the ASK than children with hemiplegia, and likewise hearing and vision loss, seizures and speech problems all decreased ASK scores indicating greater activity limitations with these impairments. Increasing severity of intellectual, movement and manual ability also all decreased ASK score (Figures 8.2 to 8.4) and were therefore associated with greater functional limitations. In terms of deprivation in area of residence ASK score only decreased for children residing in the most deprived areas compared to the least deprived but not in any of the other groups.

Figure 8.2 Box plots showing variation in ASK score by intellectual delay

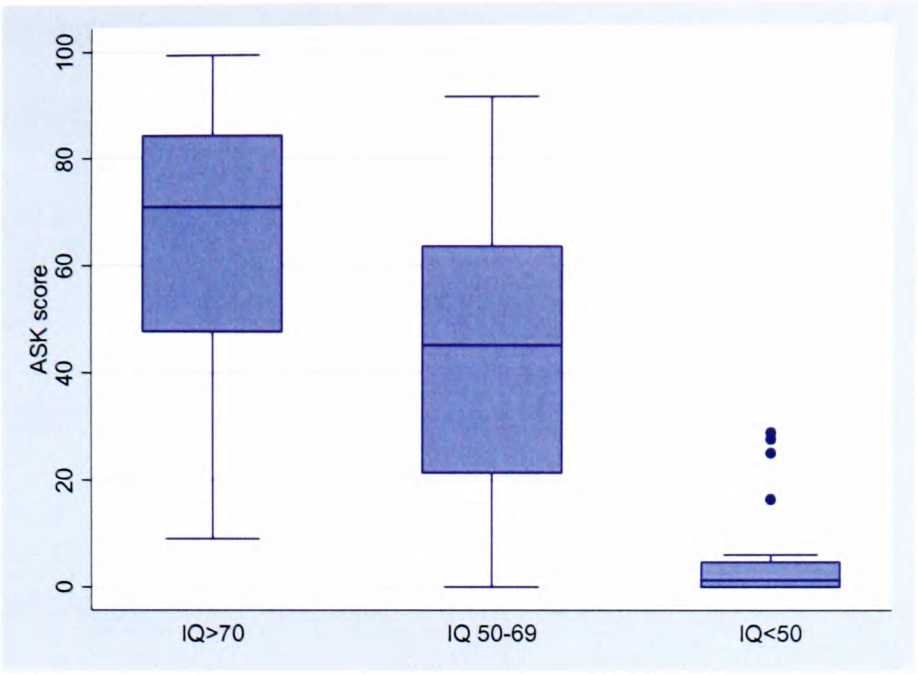


Figure 8.3 Box plots showing variation in ASK scores by GMFCS Level

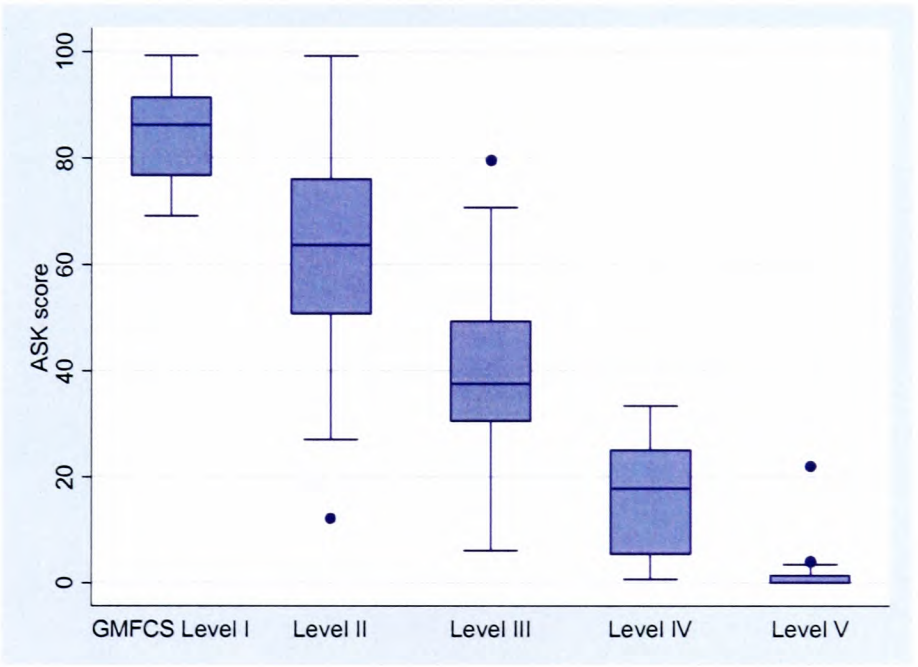
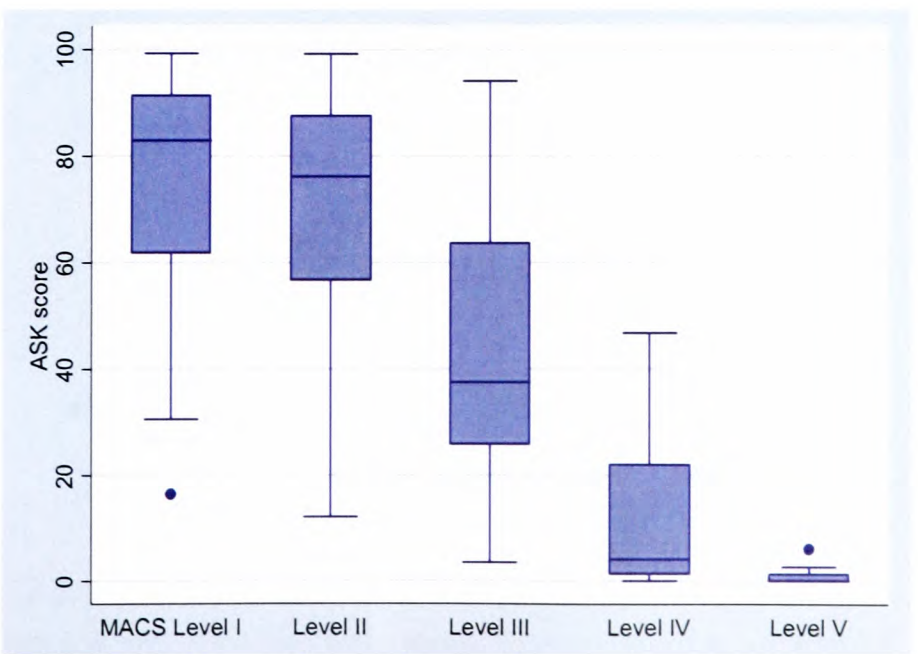


Figure 8.4 Box plots showing variation in ASK scores by MACS Level



The variables found to be significantly associated with ASK scores in the univariate analyses were subsequently included in the multiple regression analysis. After controlling for the effects of all other variables only age, movement ability (GMFCS Level), manual ability (MACS Level II to V), intellectual delay (IQ < 70) and seizures remained significant in the model explaining 89% of the variance in ASK scores ($F(12, 58) = 38.5$, $p < 0.001$). The effect on ASK score and statistical significance of each of these variables is shown in Table 8.4.

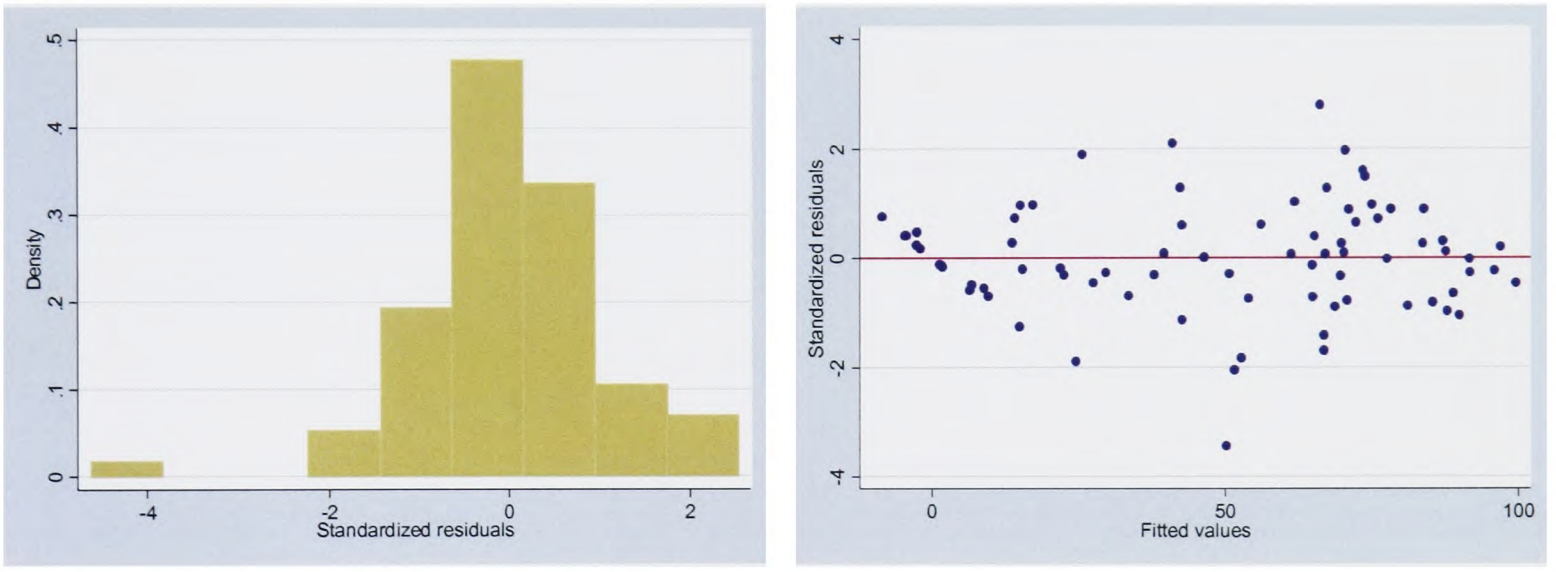
Table 8.4 Statistically significant mutually adjusted explanatory variables for the ASK score

Variable	n	Effect on ASK score		95% confidence interval		*p* value
		Crude	Adjusted			
GMFCS Level I	25	*	*			
GMFCS Level II	43	-21	-20	-29	-13	< 0.001
GMFCS Level III	15	-44	-47	- 58	-36	< 0.001
GMFCS Level IV	14	-68	-55	- 67	-42	< 0.001
GMFCS Level V	23	-82	-60	- 75	-44	< 0.001
MACS Level I	14	*	*			
MACS Level II	30	3	-6	- 15	3	0.17
MACS Level III	18	27	-13	- 23	-3	0.01
MACS Level IV	13	59	-14	- 29	-1	0.03
MACS Level V	13	72	-16	- 33	-2	0.06
IQ>70	74	*	*			
IQ 50-69	13	23	-10	-21	1	0.08
IQ<50	26	60	-15	-27	-1	0.02
no seizures	93	*	*			
Seizures	28	-21	- 9	-17	-1	0.02
Age (per year)	127	3	2	0	4	0.03
Constant		-	*77*	*58*	*97*	*< 0.001*

* Baseline against which other levels of the same impairment are compared

The variables that were not significantly associated with the ASK score when the effect of other variables was held constant were: type of cerebral palsy, hearing and vision loss, speech problems and area deprivation; forcing any of these into model failed to explain any more of the variance. The standardised residuals from the model were normally distributed and the scatter plot indicated only one far outlier indicating that the model was a reasonable fit (Figure 8.5).

Figure 8.5 Histogram of standardised residuals and scatter plot of standardised with predicted (fitted) residual values from multiple regression of ASK scores



Examination of data relating to the child represented as the far outlier at the bottom of the graph revealed a 10 year old girl with spastic hemiplegia, who was both GMFCS and MACS Level II, had seizures and an IQ between 50 and 69. The model predicts an ASK score of 52 for this child whilst her actual score was only 12, indicating that she was dependent for nearly all activities. Items in the LAQ-CP confirmed her total dependence and need for constant supervision despite being classified with relatively moderate movement and manual disability; she also had been seen by a behavioural specialist in the last year. Therefore the model is not perfect for predicting activities and participation because there is no explanatory variable for profound behavioural problems.

The Breusch-Pagan / Cook-Weisberg test for heteroskedasticity was not statistically significant ($p = 0.25$) indicating that the variance in the residuals was constant and the model was a good fit. The large amount of variance explained by the factors included in the model (89%) suggests that these factors are reasonably good for predicting ASK scores.

8.4 FACTORS AFFECTING LAQ-CP PHYSICAL INDEPENDENCE

The crude effects of explanatory variables are shown in Table 8.5 and Figures 8.6 to 8.9.

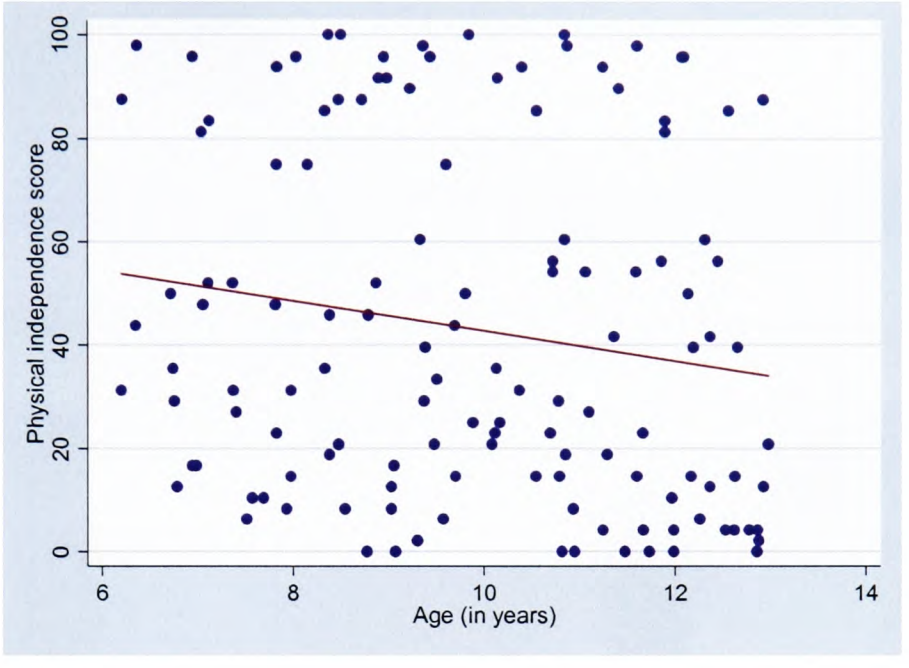
Table 8.5 Crude effects of explanatory variables on LAQ-CP Physical Independence score

Variable	Levels	Number of cases	Crude effect	R^2_{adj}	*p*
Age	per year	129	-3	0.02	0.06
Gender	female	57	*		
	male	72	6	0.00	0.28
Type of cerebral palsy	hemiplegia	46	*		
	bilateral	83	34	0.23	<0.01
Hearing loss	not deaf	124	*		
	Hearing loss	5	31	0.02	0.05
Vision loss	not blind	104	*		
	Vision loss	25	54	0.40	<0.01
Intellectual delay	no delay/IQ>70	74	*		
	IQ 50-69	13	28	-	<0.01
	IQ<50	26	59	0.55	<0.01
Seizures	no seizures	93	*		
	Seizures	28	22	0.07	<0.01
Speech problem	no speech problem	78	*		
	Speech problem	43	35	0.25	<0.01
GMFCS Level	GMFCS Level I	25	*		
	GMFCS Level II	43	17	-	<0.01
	GMFCS Level III	15	37	-	<0.01
	GMFCS Level IV	14	64	-	<0.01
	GMFCS Level V	23	82	0.79	<0.01
MACS Level	MACS Level I	14	*		
	MACS Level II	30	x	-	0.34
	MACS Level III	18	32	-	<0.01
	MACS Level IV	13	64	-	<0.01
	MACS Level V	13	78	0.71	<0.01
IMD 2004	Quintile I	67	*		
	Quintile II	23	-11	-	0.16
	Quintile III	17	-3	-	0.73
	Quintiles IV/V	20	16	0.03	0.07

*Baseline against which other levels of the same impairment are compared

Increasing age was associated with a small decrease in Physical Independence score (Figure 8.6), indicating that children became less dependent as they get older, but there was no significant difference by sex.

Figure 8.6 Scatter plot showing effect of age on in LAQ-CP Physical Independence score



Bilateral cerebral palsy was associated with higher Physical Independence scores than hemiplegia indicating reduced independence; likewise hearing and vision loss, seizures and speech problems were also associated with greater dependence. Increasing severity of intellectual, movement and manual disability were associated with increasing physical independence scores and therefore greater dependence (Figures 8.7 to 8.9). Areas with the deprivation, compared to the least deprived areas, showed an increase in Physical independence score, indicating greater dependence for children residing in areas of greater deprivation.

Figure 8.7 Box plots showing variation in LAQ-CP Physical Independence score by intellectual delay

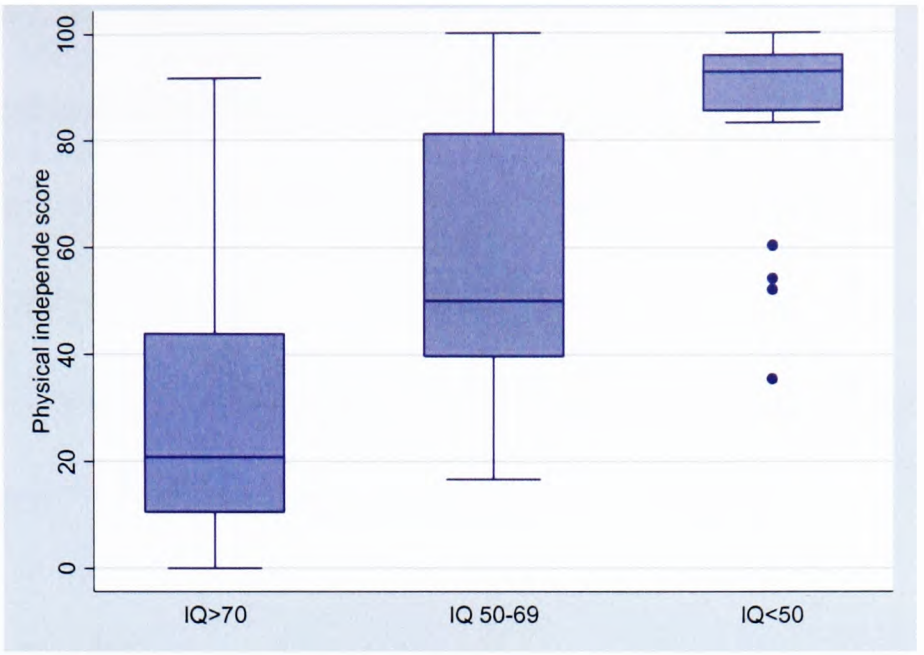


Figure 8.8 Box plots showing variation in LAQ-CP Physical Independence score by GMFCS Level

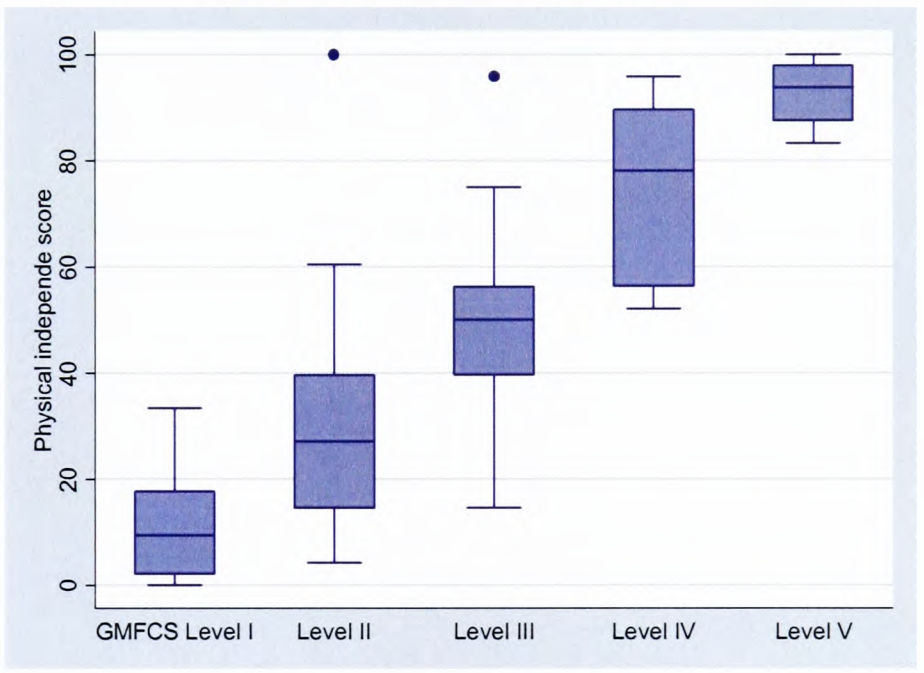
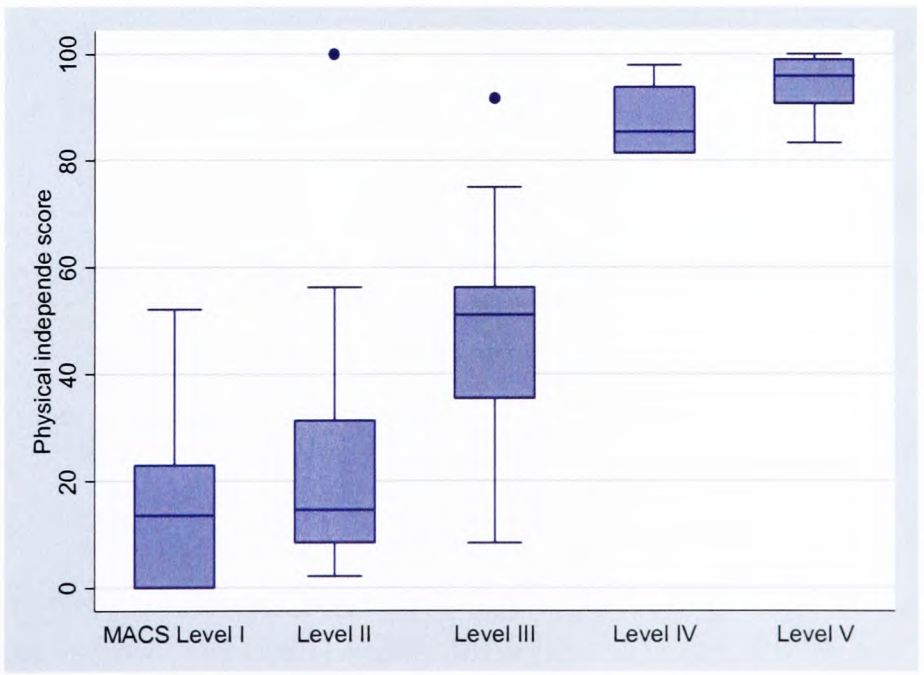


Figure 8.9 Box plots showing variation in LAQ-CP Physical Independence score by MACS Level



After controlling for the effects of other variables in the multiple regression analysis only age, intellectual delay, movement ability (GMFCS Level), manual ability (MACS Level) and speech problems remained significant in the model explaining 89% of the variance in Physical Independence score ($F(12, 59) = 39.9$, $p < 0.001$). The effects are summarised in Table 8.6. The type of cerebral palsy, hearing and vision loss, seizures and area deprivation were not significantly associated with the LAQ-CP Physical Independence score and forcing any of these into model failed to explain any more of the variance.

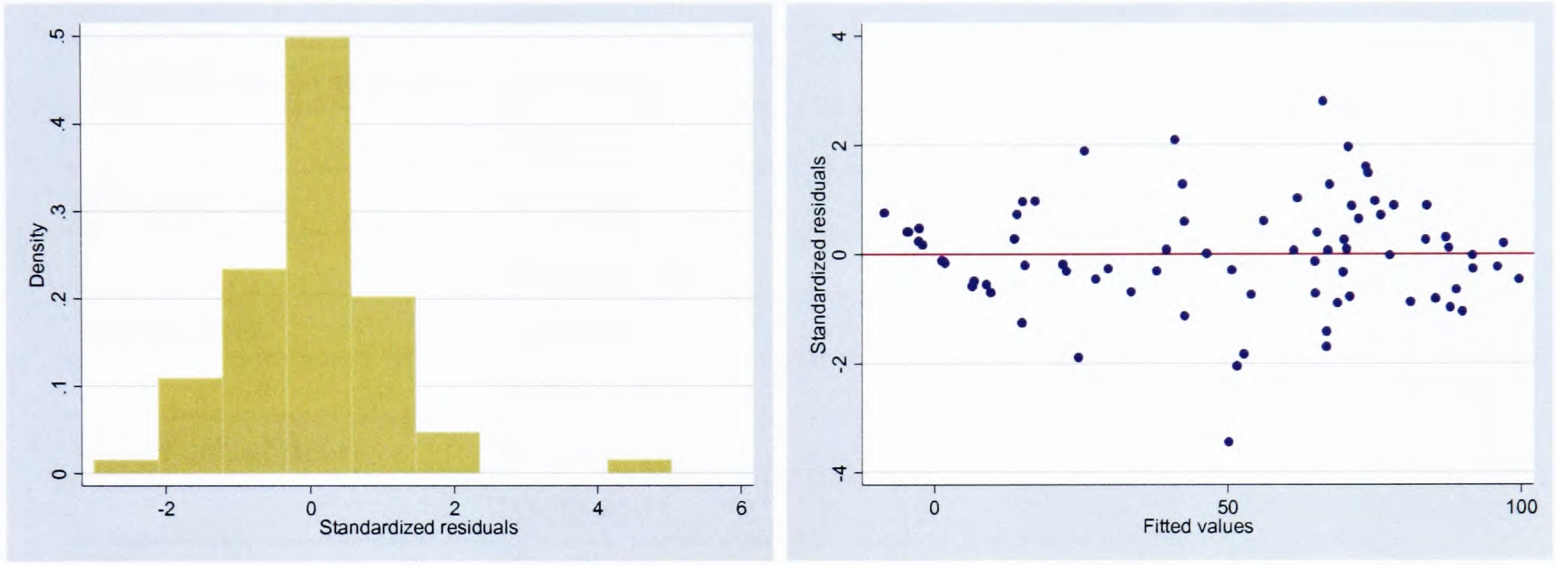
Table 8.6 Significant mutually adjusted explanatory variables for physical independence scores.

Variable	n	Effect on ASK score		95% confidence interval		*p* value
		Crude	Adjusted			
GMFCS Level I	25	*	*			
GMFCS Level II	43	17	16	8	24	< 0.001
GMFCS Level III	15	37	33	22	43	< 0.001
GMFCS Level IV	14	63	44	32	57	< 0.001
GMFCS Level V	23	82	54	39	69	< 0.001
MACS Level I	14	*	*			
MACS Level II	30	6	9	0	18	0.04
MACS Level III	18	32	23	13	33	< 0.001
MACS Level IV	13	64	20	6	34	0.003
MACS Level V	13	78	31	14	48	0.002
IQ>70	74	*	*			
IQ 50-69	13	28	20	9	32	0.002
IQ<50	26	59	6	-7	18	0.37
no speech problem	78	*	*			
Speech problem	43	35	10	2	17	0.01
Age (per year)	129	-3	- 2	-4	0	0.02
Constant		-	*18*	*0*	*35*	*0.05*

*Baseline against which other levels of the same impairment are compared

The histogram shown in Figure 8.10 illustrates that standardised residuals were normally distributed and the scatter plot of standardised with predicted (fitted) residual values showed only one outlier indicating the model was a reasonable fit.

Figure 8.10 Histogram of standardised residuals, and scatter plot of standardised with predicted (fitted) residual values, from multiple regression of LAQ-CP physical independence scores.



The scatter plot in Figure 8.10 showed one outlying case which was the same child detected as an outlier in the ASK model, a 10 year old girl with spastic hemiplegia, GMFCS and MACS Level II, speech problems and IQ between 50 and 69. This child had the maximum LAQ-CP physical independence score of 100, indicating the total independence, whereas the model predicts a score of only 67. Her profound behavioural problems and need for constant supervision, which are not explicitly included as factors in the model probably explain why the model does not fit her clinical picture. The Breusch-Pagan / Cook-Weisberg test for heteroskedasticity was not statistically significant ($p = 0.38$) indicating that the variance in the residuals was constant and the model was a good fit. The large amount of variance explained by the model suggests that this is a reasonably good model for predicting the LAQ-CP Physical Independence score.

8.5 FACTORS AFFECTING LAQ-CP MOBILITY

The crude effects of each explanatory variable are shown in Table 8.5 and Figures 8.11 to 8.14.

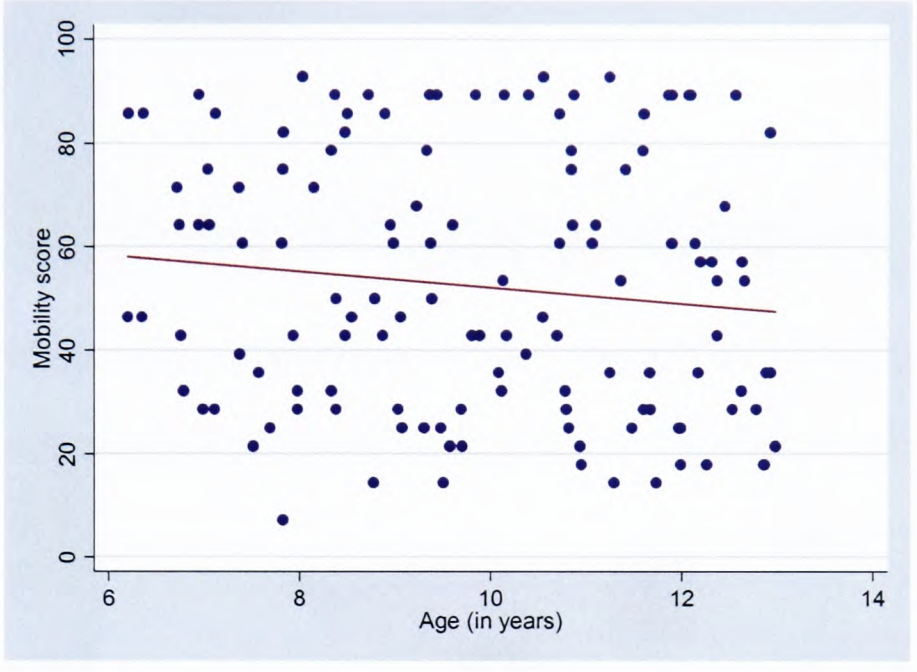
Table 8.7 Crude effects of explanatory variables on LAQ-CP Mobility score

Variable	Levels	Number of cases	Crude effect	R^2_{adj}	*p*
Age	per year	129	-2	0.01	0.17
Gender	female	57	*		
	male	72	4	0.01	0.40
Type of cerebral palsy	hemiplegia	46	*		
	bilateral	83	31	0.34	<0.01
Hearing loss	not deaf	124	*		
	Hearing loss	5	16	0.01	0.16
Vision loss	not blind	104	*		
	Vision loss	25	37	0.33	<0.01
Intellectual delay	no delay/IQ>70	74	*		
	IQ 50-69	13	13	-	0.02
	IQ<50	26	40	0.45	<0.01
Seizures	no seizures	93	*		
	Seizures	28	17	0.08	<0.01
Speech problem	no speech problem	78	*		
	Speech problem	43	23	0.19	<0.01
GMFCS Level	GMFCS Level I	25	*		
	GMFCS Level II	43	18	-	<0.01
	GMFCS Level III	15	34	-	<0.01
	GMFCS Level IV	14	49	-	<0.01
	GMFCS Level V	23	61	0.76	<0.01
MACS Level	MACS Level I	14	*		
	MACS Level II	30	4	-	0.52
	MACS Level III	18	20	-	<0.01
	MACS Level IV	13	41	-	<0.01
	MACS Level V	13	53	0.53	<0.01
IMD 2004	Quintile I	67	*		
	Quintile II	23	-4	-	0.46
	Quintile III	17	3	-	0.61
	Quintiles IV/V	20	9	0.00	0.15

*Baseline against which other levels of the same impairment are compared

Increasing age was associated with a small decrease in Mobility score (Figure 8.11), indicating that children's mobility increased as they get older; but the small decrease in Mobility score for males compared to females was not statistically significant.

Figure 8.11 Scatter plot showing effect of age on LAQ-CP Mobility score



Bilateral cerebral palsy was associated with higher Mobility scores than hemiplegia indicating restricted mobility; likewise hearing and vision loss, seizures and speech problems were also associated with restrictions in mobility. Increasing severity of intellectual, movement and manual disability were all associated with increased Mobility score and therefore greater restriction in mobility (Figures 8.7 to 8.9). There was a small increase in Mobility score for areas with greatest deprivation compared to the least deprived areas, indicating slightly greater mobility restrictions for children residing in more deprived areas.

Figure 8.12 Box plot showing variation in LAQ-CP Mobility score by intellectual delay

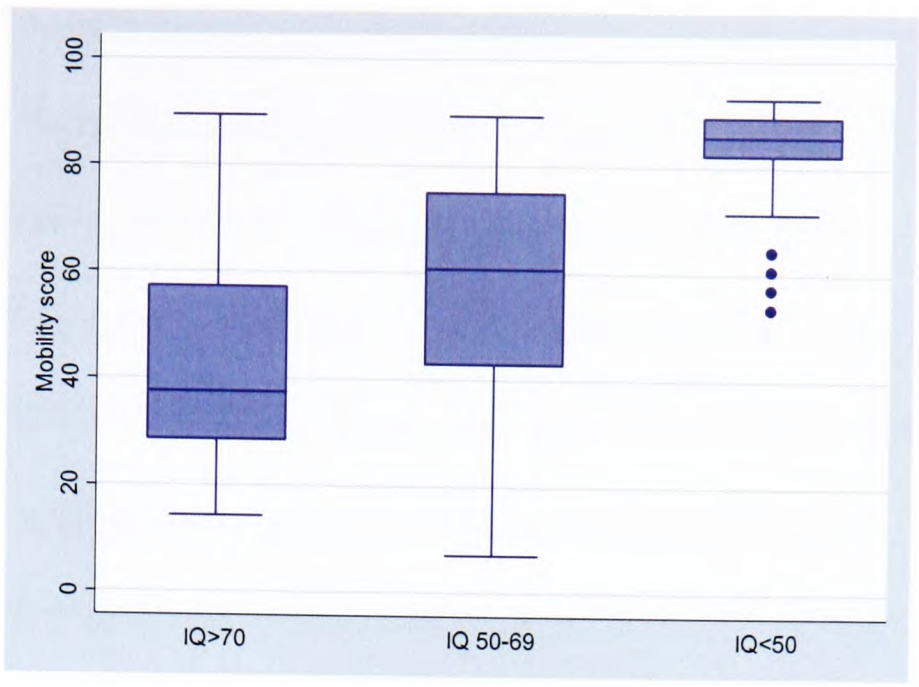


Figure 8.13 Box plot showing variation in LAQ-CP Mobility score by GMFCS

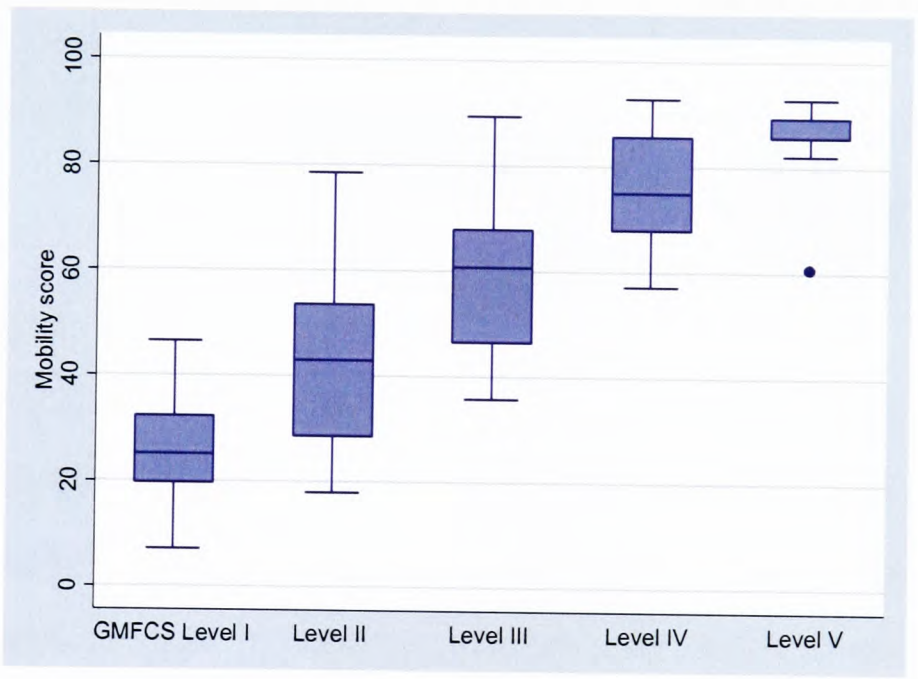
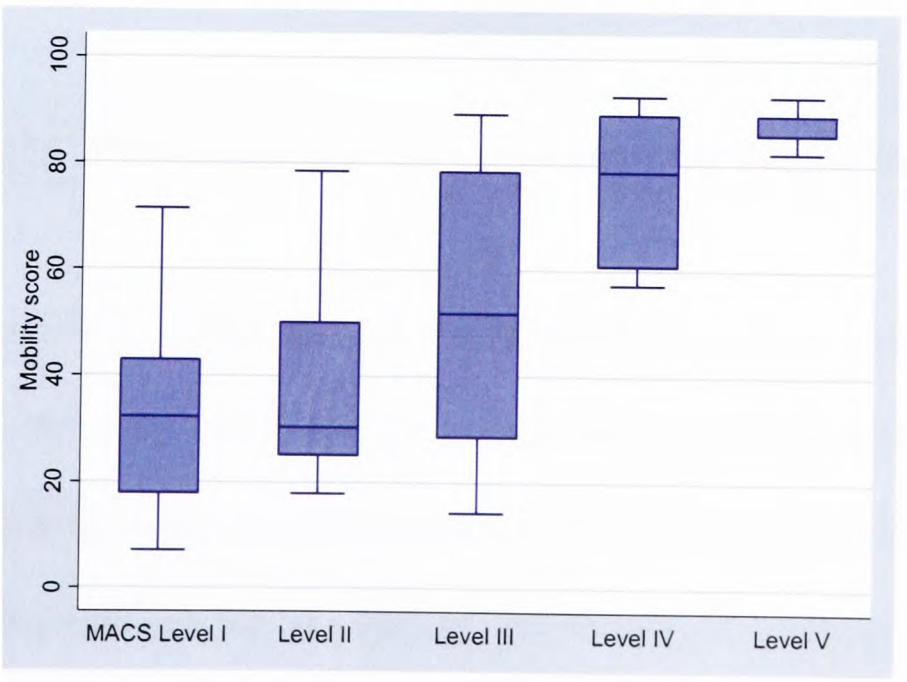


Figure 8.14 Box plot showing variation in LAQ-CP Mobility score by MACS



After controlling for the effects of other variables in the multiple regression analysis only movement ability (GMFCS Level) and seizures remained statistically significant in the model, explaining 77% of the variance in Mobility score ($F(5, 114) = 75.8$, $p < 0.001$). The effects are summarised in Table 8.8. The type of cerebral palsy, hearing and vision loss, seizures, manual and intellectual ability, and area deprivation were not significantly associated with the LAQ-CP Mobility score when the effect of other variables was held constant. However, adding manual and intellectual ability to the model, whilst not producing statistically significant effects, explained slightly more of the variance (83%).

Table 8.8 Statistically significant mutually adjusted explanatory variables for LAQ-CP Mobility score

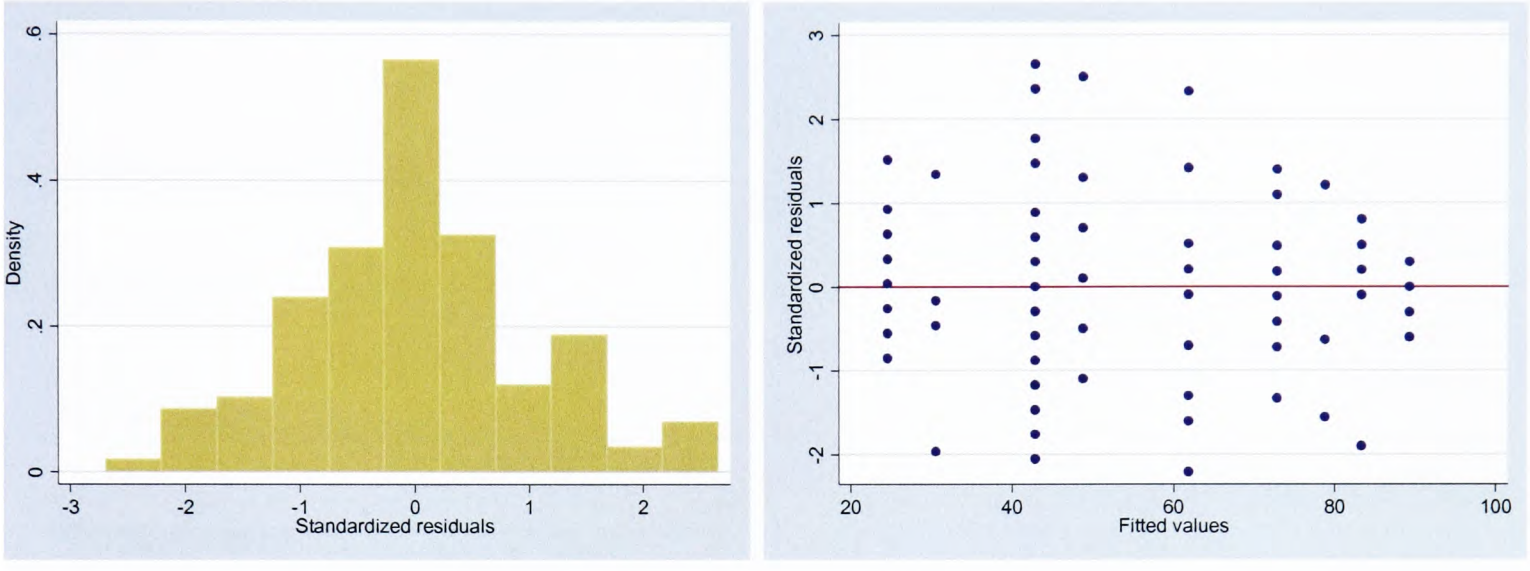
Variable	n	Effect on mobility score		95% confidence intervals		*p* value
		Crude	Adjusted			
GMFCS Level I	25	*	*			
GMFCS Level II	43	18	1	12	24	0.001
GMFCS Level III	15	34	37	29	45	<0.01
GMFCS Level IV	14	49	48	40	56	<0.01
GMFCS Level V	23	61	59	52	66	<0.01
no seizures	93	*	*			
Seizures	28	17	6	0	12	0.04
Constant		-	*18*	*20*	*29*	<0.01

*Baseline against which other levels of the same impairment are compared

The histogram in Figure 6.15 shows that the standardised residuals were normally distributed. The fact that there were only two statistically significant explanatory variables in the model means that the residuals predicted by the model can only take a limited number of fitted values and this explains the 'stacked up' distribution of standardised and predicted

(fitted) residuals and makes it difficult to identify outliers. The Breusch-Pagan / Cook-Weisberg test for heteroskedasticity was not statistically significant ($p = 0.23$) indicating that the variance in the residuals was constant and the model was a good fit.

Figure 8.15 Histogram of standardised residuals and scatter plot of standardised with predicted (fitted) residual values from multiple regression of LAQ-CP mobility



This model indicates that the LAQ-CP Mobility scale was predicted by movement ability (GMFCS) and seizures. A large amount of the variance (77%) was explained by these factors although this was mostly due to the effect of the GMFCS Level. Some ability to carry objects is included in the LAQ-CP Mobility scale and adding manual ability the MACS to the model, although not reaching statistical significance, did explain a little more of the variance (5%), adding intellectual disability explained a fraction more (1%).

8.6 FACTORS AFFECTING LAQ-CP CLINICAL BURDEN

The crude effects of each explanatory variable on Clinical Burden score are shown in Table 8.9 and Figures 8.16 to 8.19.

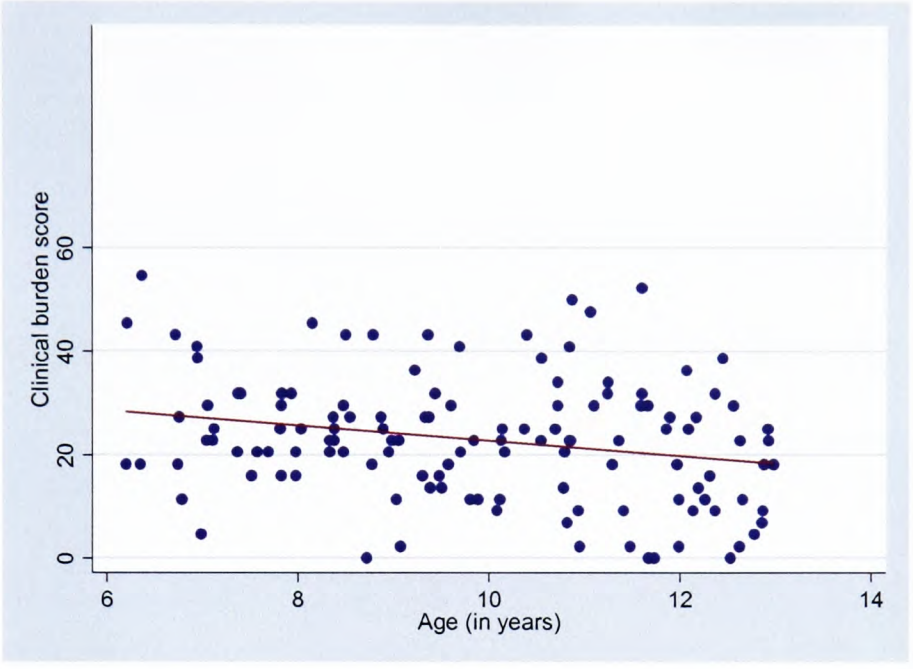
Table 8.9 Crude effects of explanatory variables on LAQ-CP Clinical Burden score

Variable	Levels	Number of cases	Crude effect	R^2_{adj}	*p*
Age	per year	129	-1	0.05	0.01
Gender	female	57	*		
	male	72	1	0.00	0.71
Type of cerebral palsy	hemiplegia	46	*		
	bilateral	83	7	0.07	<0.01
Hearing loss	not deaf	124	*		
	Hearing loss	5	6	0.00	0.25
Vision loss	not blind	104	*		
	Vision loss	25	8	0.06	<0.01
Intellectual delay	no delay/IQ>70	74	*		
	IQ 50-69	13	7	-	0.05
	IQ<50	26	11	0.13	<0.01
Seizures	no seizures	93	*		
	Seizures	28	3	0.01	0.19
Speech problem	no speech problem	78	*		
	Speech problem	43	5	0.04	0.02
GMFCS Level	GMFCS Level I	25	*		
	GMFCS Level II	43	8	-	<0.01
	GMFCS Level III	15	11	-	<0.01
	GMFCS Level IV	14	13	-	<0.01
	GMFCS Level V	23	20	0.28	<0.01
MACS Level	MACS Level I	14	*		
	MACS Level II	30	3	-	0.33
	MACS Level III	18	7	-	0.06
	MACS Level IV	13	10	-	0.02
	MACS Level V	13	18	0.22	<0.01
IMD 2004	Quintile I	67	*		
	Quintile II	23	-1	-	0.74
	Quintile III	17	0	-	0.89
	Quintiles IV/V	20	0	0.00	0.90

*Baseline against which other levels of the same impairment are compared

Increasing age was associated with a small decrease in Clinical Burden score (Figure 8.16), indicating marginally less burden for older children; but the negligible one point increase in Clinical Burden score for males compared to females was not statistically significant.

Figure 8.16 Scatter plot showing the effect of age on LAQ-CP Clinical Burden score.



Bilateral cerebral palsy was associated with greater clinical burden than hemiplegia; likewise hearing and vision loss, seizures and speech problems were also all associated with greater burden. Increasing severity of intellectual, movement and manual disability were all associated with small increases clinical burden (Figures 8.17 to 8.19). There was no effect of area deprivation on Clinical Burden scores.

Figure 8.17 Box plot showing variation in LAQ-CP Clinical Burden score by intellectual delay

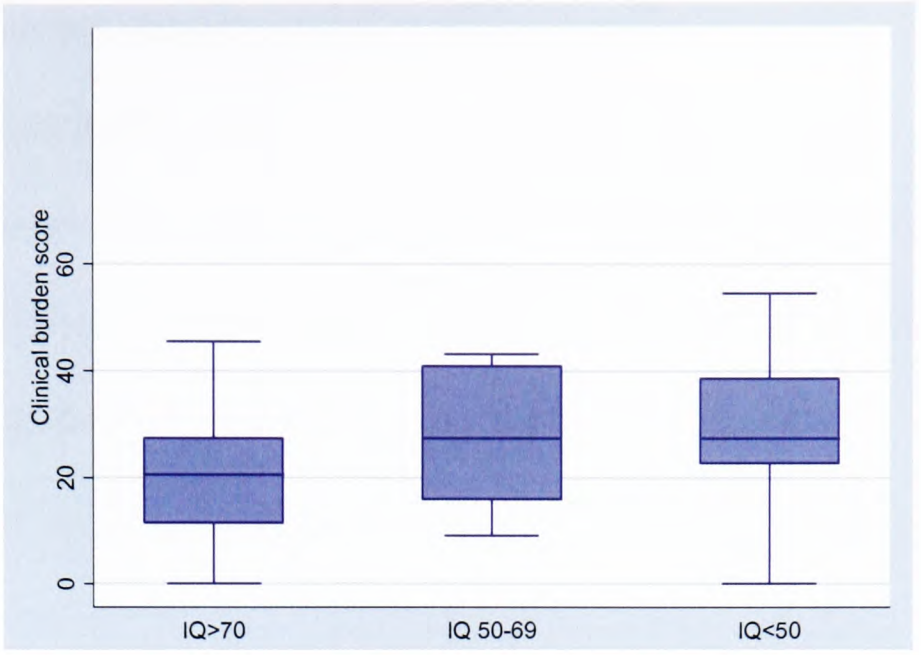


Figure 8.18 Box plot showing variation in LAQ-CP Clinical Burden score by GMFCS

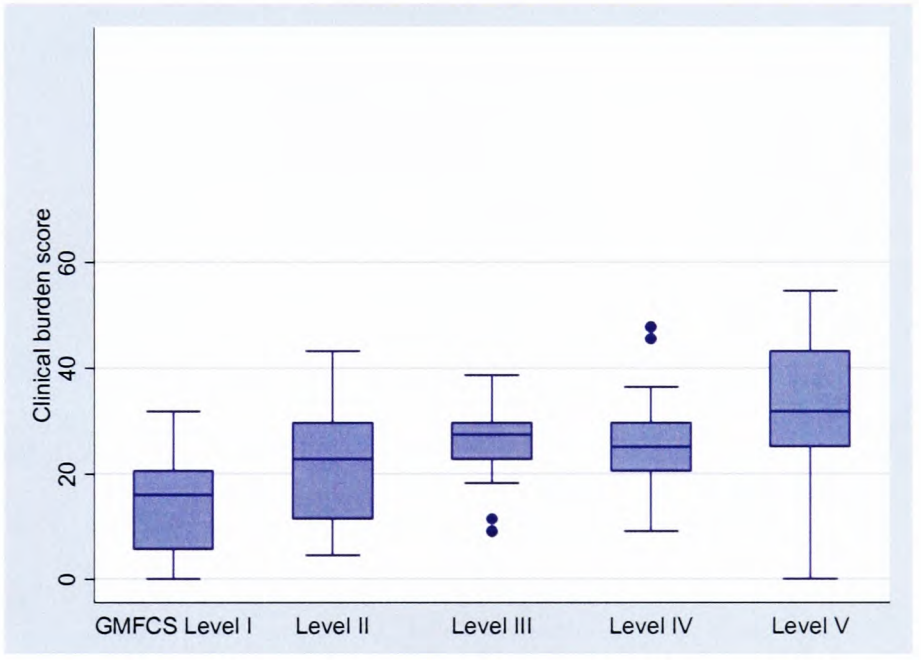
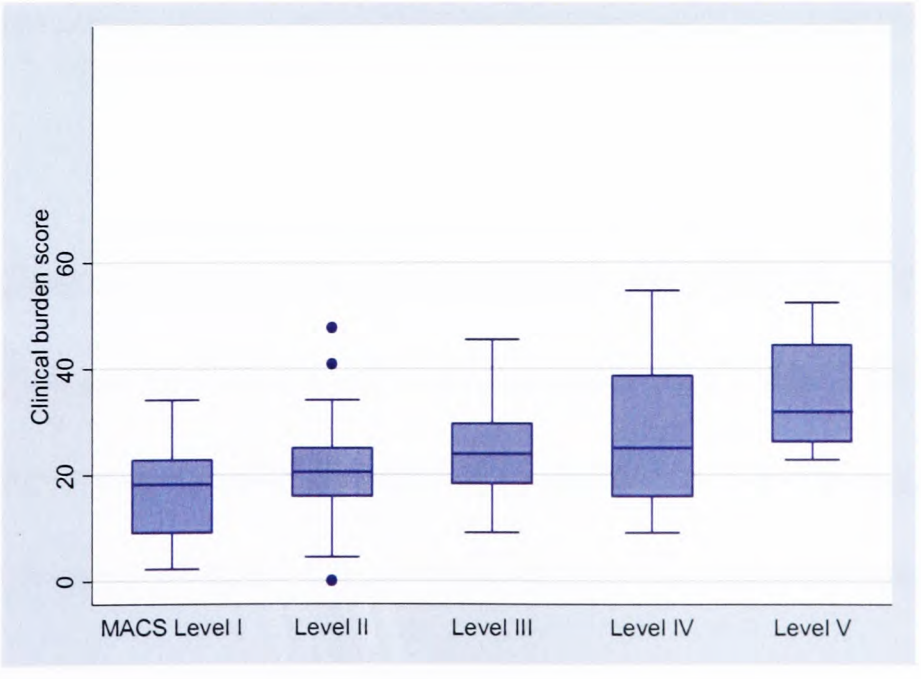


Figure 8.19 Box plot showing variation in LAQ-CP Clinical Burden score by MACS



After controlling for the effects of other variables in the multiple regression analysis, only age and movement ability remained significantly associated with clinical burden, explaining only 32% of the variance ($F(5, 121) = 12.8$, $p = < 0.001$) (Table 8.10). The type of cerebral palsy, hearing and vision loss, seizures, manual and intellectual ability, and area deprivation were not statistically significantly associated with the LAQ-CP Clinical Burden score when the effect of other variables was held constant. However, adding manual ability (MACS) to the model, whilst not producing statistically significant effects, explained slightly more of the variance (41%).

Table 8.10 Significant mutually adjusted explanatory variables for LAQ-CP Clinical Burden score

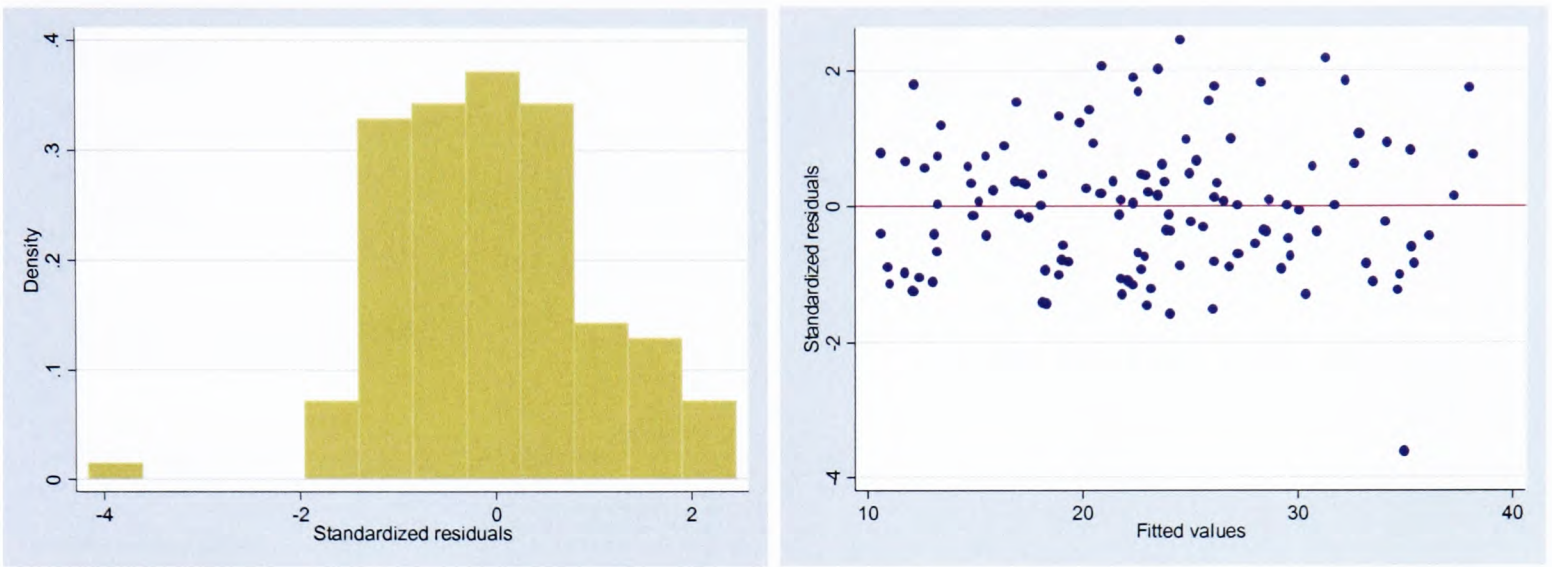
Variable	n	Effect on clinical burden score		95% confidence intervals		*p* value
		Crude	Adjusted			
GMFCS Level I	25	*	*			
GMFCS Level II	43	8	8	3	12	0.001
GMFCS Level III	15	11	11	5	18	< 0.001
GMFCS Level IV	14	12	12	5	18	< 0.001
GMFCS Level V	23	19	19	14	24	< 0.001
Age (per year)	129	-2	- 1	- 2	0	0.006
Constant		-	*27*	*17*	*37*	< 0.001

*Baseline against which other levels of the same impairment are compared

The plot of standardised and predicted (fitted) residuals identified one outlier. This 9 year old child was classified as GMFCS Level V and scored zero on the clinical burden scale for which the model estimates a score of 28 indicating the model was not a good fit using her data. In fact, as the Breusch-Pagan / Cook-Weisberg test for heteroskedasticity was

statistically significant ($p = 0.04$), this indicates that the variance in the residuals was not constant and the model was not a good fit generally.

Figure 8.20 Histogram of standardised residuals and scatter plot of standardised with predicted (fitted) residual values from multiple regression of LAQ-CP clinical burden



Only two explanatory variables remained statistically significant in the model, and little of the overall variance in the outcome measures was explained by these factors (32%); adding manual ability explained slightly more of the variance but, overall, the present model cannot be considered satisfactory for explaining Clinical Burden score.

8.7 FACTORS AFFECTING LAQ-CP ECONOMIC BURDEN SCORE

The unadjusted effect of the explanatory variables on LAQ-CP Economic Burden score are shown in Table 8.11 and Figures 8.21 to 8.24.

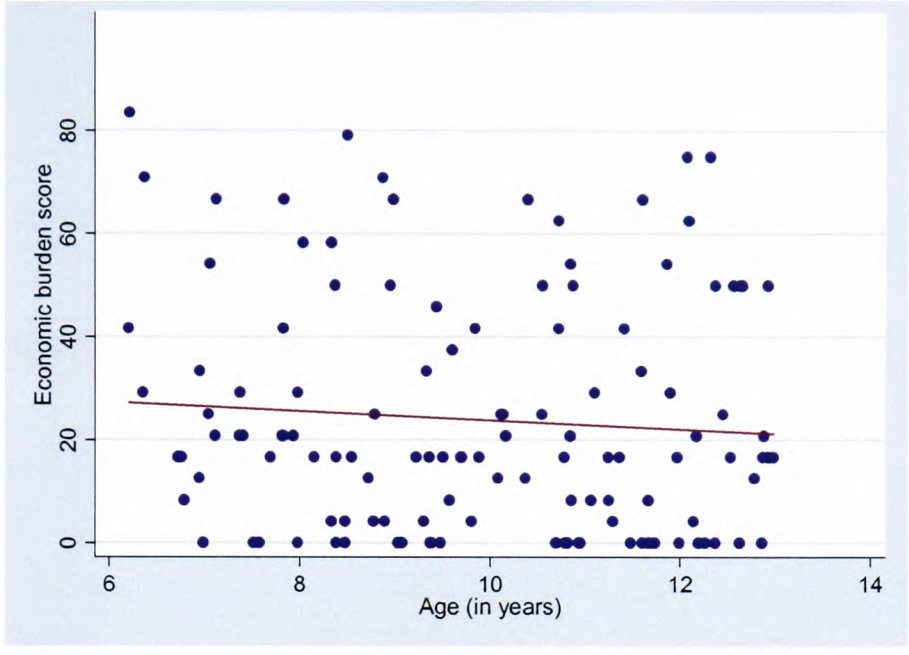
Table 8.11 Crude effects of explanatory variables on LAQ-CP Economic Burden score

Variable	Levels	Number of cases	Crude effect	R^2_{adj}	*p*
Age	per year	129	-1	0.00	0.40
Gender	female	57	*		
	male	72	6	0.01	0.11
Type of cerebral palsy	hemiplegia	46	*		
	bilateral	83	22	0.20	<0.01
Hearing loss	not deaf	124	*		
	Hearing loss	5	17	0.01	0.10
Vision loss	not blind	104	*		
	Vision loss	25	28	0.23	<0.01
Intellectual delay	no delay/IQ>70	74	*		
	IQ 50-69	13	13	-	0.03
	IQ<50	26	29	0.28	<0.01
Seizures	no seizures	93	*		
	Seizures	28	6	0.01	0.18
Speech problem	no speech problem	78	*		
	Speech problem	43	13	0.07	<0.01
GMFCS Level	GMFCS Level I	25	*		
	GMFCS Level II	43	8	-	0.05
	GMFCS Level III	15	32	-	<0.01
	GMFCS Level IV	14	34	-	<0.01
	GMFCS Level V	23	39	0.47	<0.01
MACS Level	MACS Level I	14	*		
	MACS Level II	30	2	-	0.76
	MACS Level III	18	13	-	0.04
	MACS Level IV	13	35	-	<0.01
	MACS Level V	13	42	0.44	<0.01
IMD 2004	Quintile I	67	*		
	Quintile II	23	-7	-	0.21
	Quintile III	17	-5	-	0.37
	Quintiles IV/V	20	-5	0.00	0.36

*Baseline against which other levels of the same impairment are compared

Increasing age was associated with a small decrease in Economic Burden score (Figure 8.21), indicating marginally less burden for older children; and the six point increase in Economic Burden score for males compared to females was marginally significant.

Figure 8.21 Scatter plot showing the effect of age on LAQ-CP Economic Burden score.



Bilateral cerebral palsy was associated with greater economic burden than hemiplegia; likewise hearing and vision loss, seizures and speech problems were also all associated with greater economic burden. Increasing severity of intellectual, movement and manual disability were all associated with small increases economic burden in univariate analyses (Figures 8.17 to 8.19). Economic Burden scores decreased slightly in areas of greater deprivation compared to the least deprived areas but the effect was not statistically significant.

Figure 8.22 Box plot showing variation in LAQ-CP Economic Burden score by intellectual delay

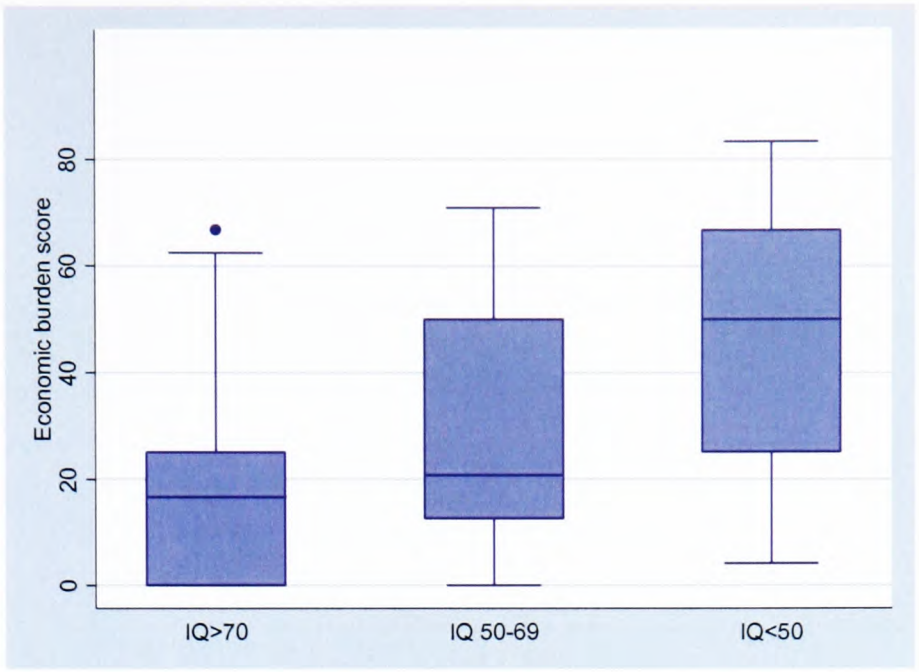


Figure 8.23 Box plot showing variation in LAQ-CP Economic Burden score by GMFCS

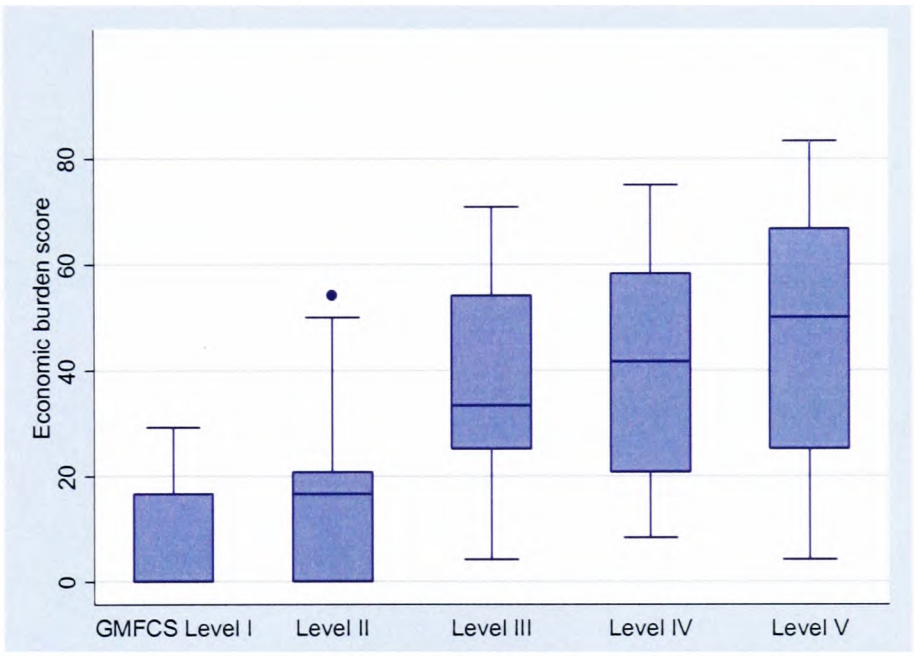
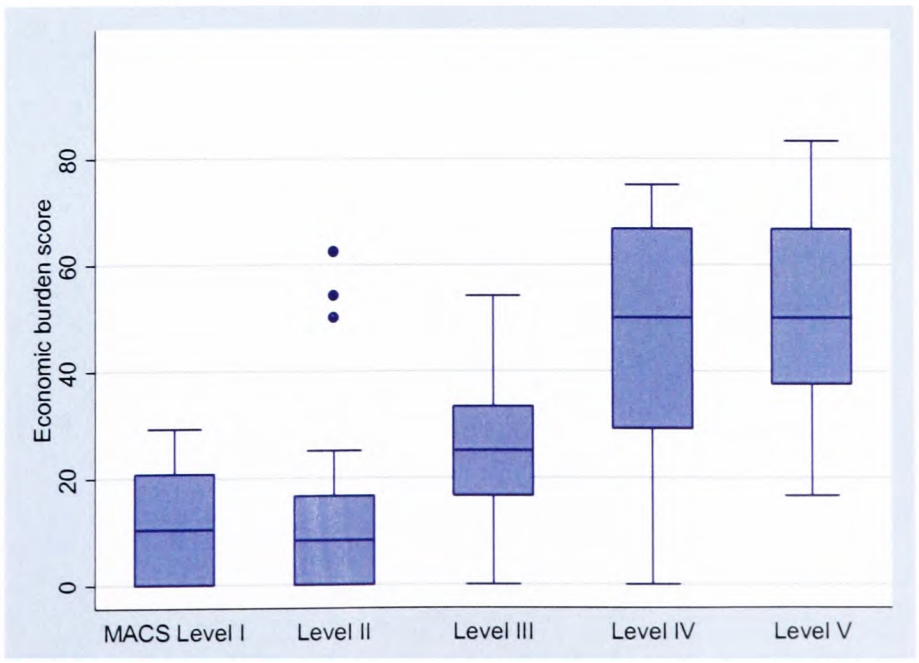


Figure 8.24 Box plot showing variation in LAQ-CP Economic Burden score by MACS



When the effect of other variables are held constant only movement and manual ability (GMFCS and MACS) remained statistically significantly associated with Economic Burden score and explained 56% of the variance ($F(8, 77) = 14.4$, $p = < 0.001$) (Table 8.12). Type of cerebral palsy, hearing and vision loss, seizures, intellectual ability and area deprivation were not statistically significantly associated with the LAQ-CP Economic Burden score when the effect of other variables was held constant.

Table 8.12 Statistically significant explanatory variable for Economic Burden Score

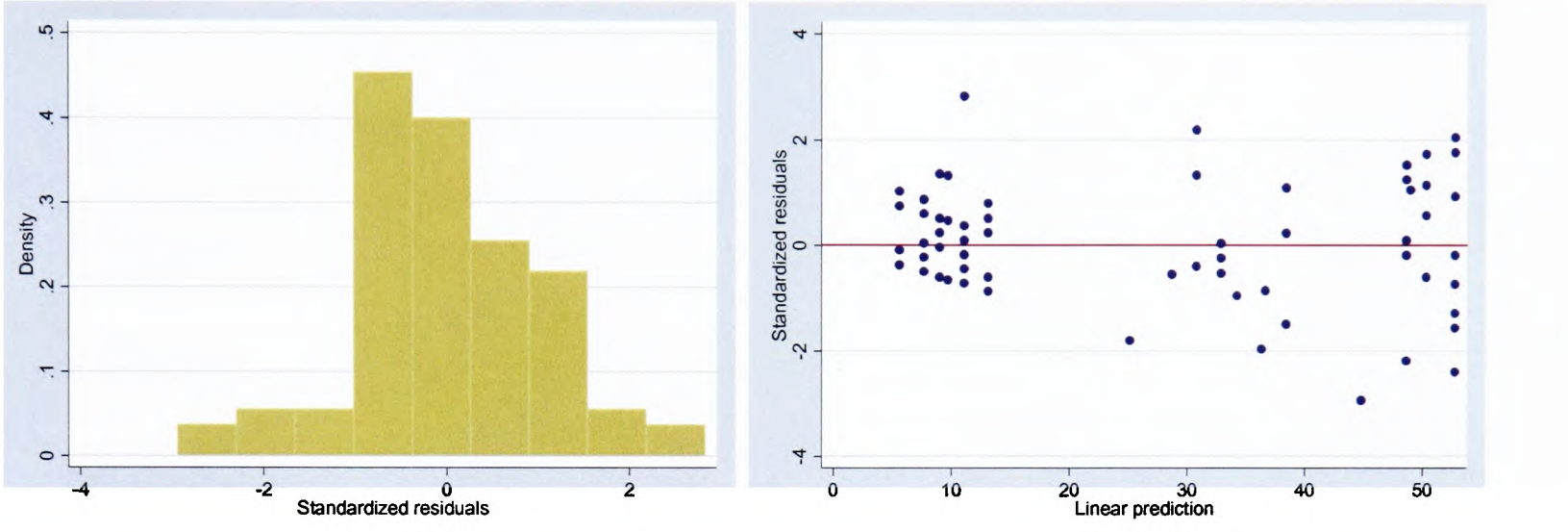
Variable	n	Effect on economic burden score		95% confidence intervals		*p* value
		Crude	Adjusted			
GMFCS Level I	25	*	*			
GMFCS Level II	43	8	3	-6	13	0.05
GMFCS Level III	15	32	23	10	46	< 0.001
GMFCS Level IV	14	34	29	15	43	< 0.001
GMFCS Level V	23	39	27	10	44	< 0.001
MACS Level I	14	*	*			
MACS Level II	30	2	2	-8	12	0.68
MACS Level III	18	13	4	-8	16	0.48
MACS Level IV	13	35	16	0	32	0.05
MACS Level V	13	42	20	1	40	0.04
Constant		-	*6*	*-4*	*15*	*0.26*

*Baseline against which other levels of the same impairment are compared

The histogram of standardised residuals showed these to be fairly normally distributed (Figure 8.25), however, the scatter plot of standardised and predicted (fitted) residuals showed a slight funnel pattern (Figure 8.25). As the Breusch-Pagan / Cook-Weisberg test

for heteroskedasticity was also highly statistically significant ($p < 0.001$), indicating that the variance in the residuals was not at all constant, taken together the evidence suggests that the model did not appear to fit these data well.

Figure 8.25 Histogram of standardised residuals and scatter plot of standardised with predicted (fitted) residual values from multiple regression of LAQ-CP Economic Burden



Although economic burden appears to be partly explained by movement and manual ability, the present model cannot be considered satisfactory for explaining Economic Burden score.

8.8 FACTORS AFFECTING LAQ-CP SOCIAL INTEGRATION

The unadjusted effect of the explanatory variables on LAQ-CP Social Integration score are shown in Table 8.13 and Figures 8.26 to 8.29.

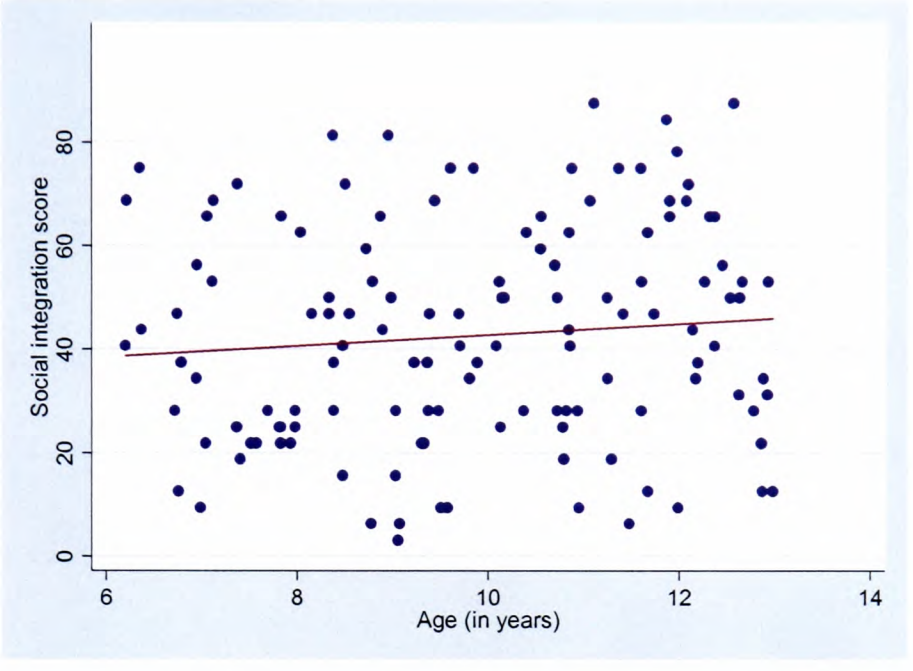
Table 8.13 Crude effects of explanatory variables on LAQ-CP Social Integration score

Variable	Levels	Number of cases	Crude effect	R^2_{adj}	*p*
Age	per year	129	1	0.00	0.27
Gender	female	57	*		
	male	72	2	0.00	0.55
Type of cerebral palsy	hemiplegia	46	*		
	bilateral	83	18	0.15	<0.01
Hearing loss	not deaf	124	*		
	Hearing loss	5	27	0.05	<0.01
Vision loss	not blind	104	*		
	Vision loss	25	20	0.13	<0.01
Intellectual delay	no delay/IQ>70	74	*		
	IQ 50-69	13	15	-	<0.01
	IQ<50	26	23	0.22	<0.01
Seizures	no seizures	93	*		
	Seizures	28	14	0.07	<0.01
Speech problem	no speech problem	78	*		
	Speech problem	43	13	0.07	<0.01
GMFCS Level	GMFCS Level I	25	*		
	GMFCS Level II	43	13	-	<0.01
	GMFCS Level III	15	28	-	<0.01
	GMFCS Level IV	14	25	-	<0.01
	GMFCS Level V	23	32	0.29	<0.01
MACS Level	MACS Level I	14	*		
	MACS Level II	30	-1	-	0.86
	MACS Level III	18	7	-	0.30
	MACS Level IV	13	20	-	<0.01
	MACS Level V	13	30	0.25	<0.01
IMD 2004	Quintile I	67	*		
	Quintile II	23	1	-	0.77
	Quintile III	17	7	-	0.20
	Quintiles IV/V	20	3	0.00	0.58

*Baseline against which other levels of the same impairment are compared

Increasing age was associated with a small decrease in Social Integration score (Figure 8.26), indicating marginally less social exclusion as children get older. The marginal addition exclusion of males compared to females was not statistically significant.

Figure 8.26 Scatter plot showing the effect of age on LAQ-CP Social Integration score.



Bilateral cerebral palsy was associated with increased social exclusion compared to hemiplegia; similarly, hearing and vision loss, seizures and speech problems were also all associated with greater social exclusion. Increasing severity of intellectual, movement and manual disability were all associated with increasing social exclusion in univariate analyses (Figures 8.27 to 8.29). Area deprivation was not statistically significantly associated with social exclusion or integration.

Figure 8.27 Box plot showing variation in LAQ-CP Social Integration score by intellectual delay

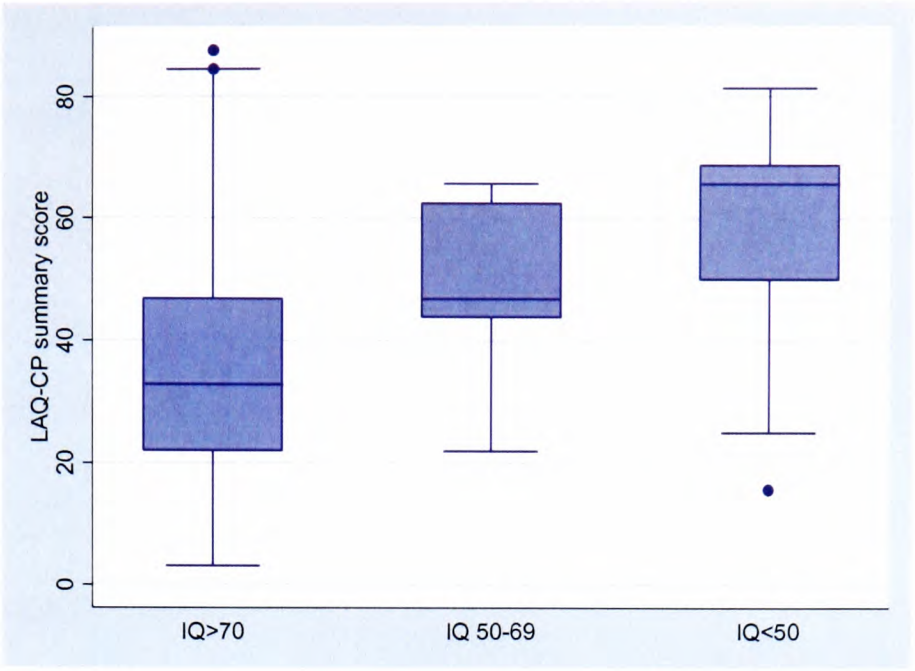


Figure 8.28 Box plot showing variation in LAQ-CP Social Integration score by GMFCS

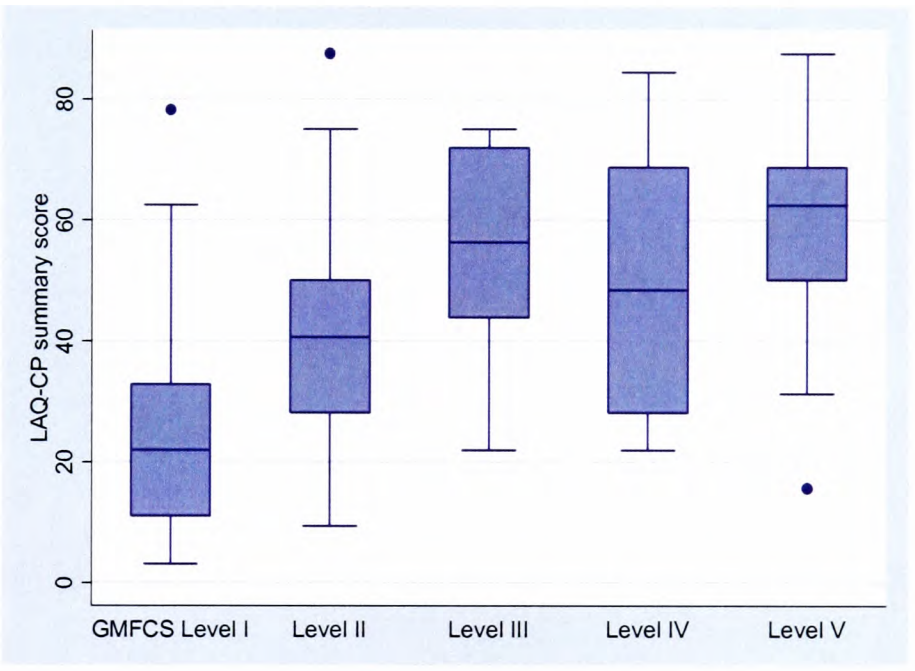
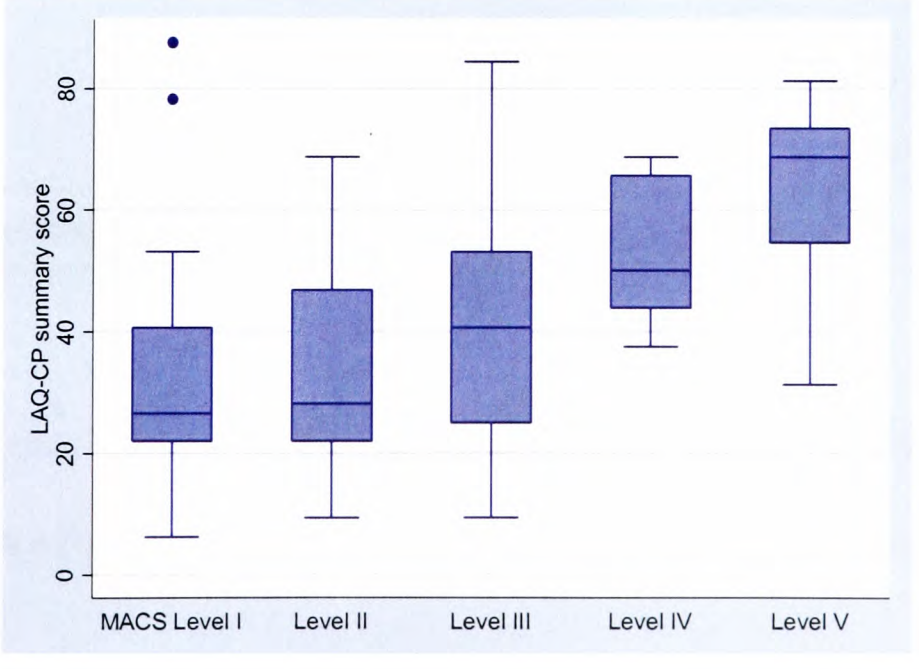


Figure 8.29 Box plot showing variation in LAQ-CP Social Integration score by MACS



After controlling for the confounding effects of other variables only movement ability (GMFCS) and intellectual delay (IQ) remained significantly associated with social integration scores, explaining 35% of the variance ($F(6, 105) = 11.0$, $p = < 0.001$) as shown in Table 8.14. Age, type of cerebral palsy, hearing and vision loss, seizures, manual ability and area deprivation were not statistically significantly associated with the LAQ-CP Social Integration score when the effect of other variables was held constant. However, including manual ability in the model, although not reaching statistically significance, did explain a little more of the variance (42%)

Table 8.14 Significant mutually adjusted explanatory variables for LAQ-CP Social Integration

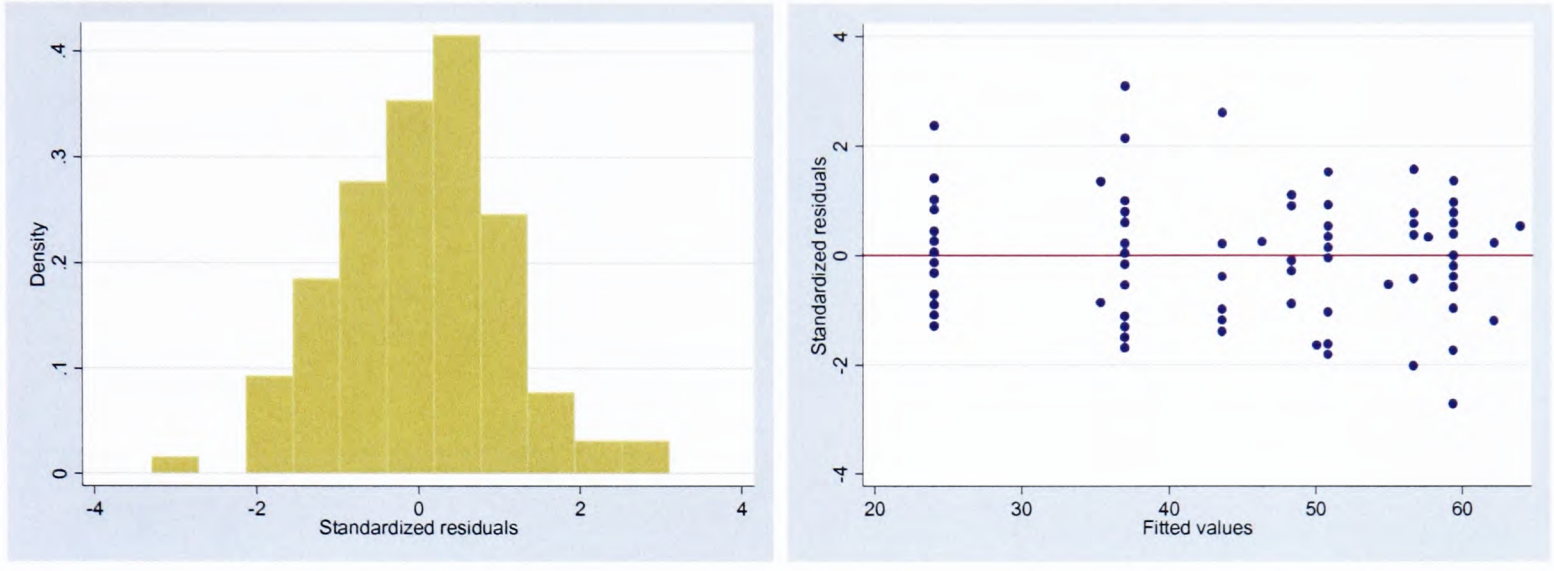
Variable	n	Effect on social integration score		95% confidence intervals		*p* value
		Crude	Adjusted			
GMFCS Level I	25	*	*			
GMFCS Level II	43	13	13	5	21	0.003
GMFCS Level III	15	28	27	16	38	<0.001
GMFCS Level IV	14	25	20	7	32	0.003
GMFCS Level V	23	33	22	7	37	0.004
IQ>70	74	*	*			
IQ 50-69	13	15	11	1	23	0.03
IQ<50	26	23	13	1	26	0.04
Constant		-	*24*	*18*	*30*	<0.001

*Baseline against which other levels of the same impairment are compared

The histogram in Figure 8.30 shows that the standardised residuals were fairly normally distributed. The plot of standardised and predicted (fitted) residuals shows a stacked pattern

which reflects the limited number of values residual take with there being only two significant explanatory variables.

Figure 8.30 Histogram of standardised residuals and scatter plot of standardised with predicted (fitted) residual values from multiple regression of LAQ-CP social integration



The Breusch-Pagan / Cook-Weisberg test for heteroskedasticity was not statistically significant ($p < 0.62$) indicating that the variance in the residuals was constant and that the model was a good fit despite only explaining a limited amount of the variance.

Although social integration appears to be partly explained by a child's movement ability and intellectual delay, as only 36% (or 42% if manual ability is included) of the variation is explained by these factors, the present model cannot be considered entirely satisfactory for explaining LAQ-CP Social Integration score

8.9 FACTORS AFFECTING LAQ-CP SUMMARY SCORE

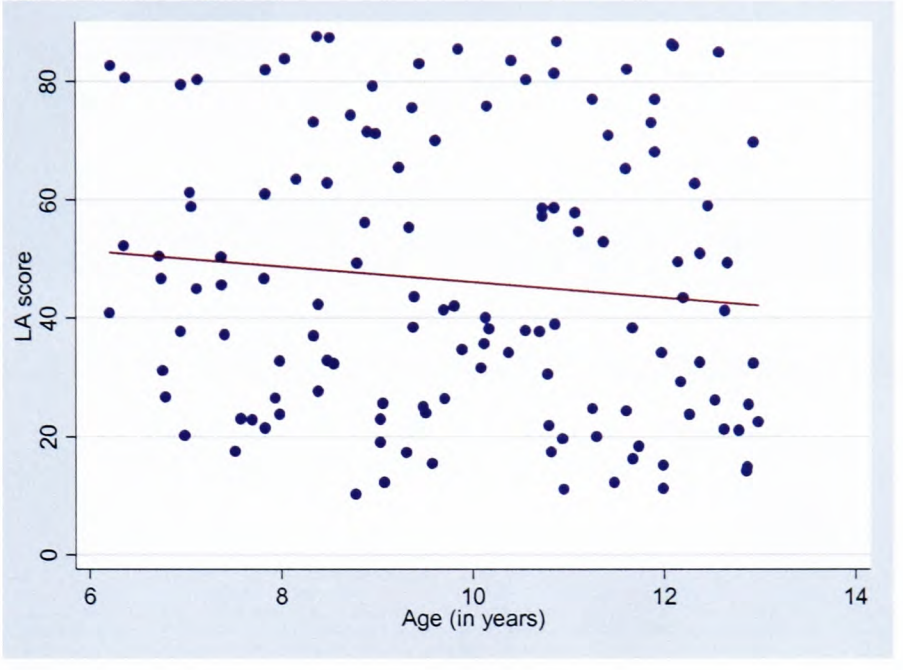
Table 8.15 Crude effects of explanatory variables on LAQ-CP summary score

Variable	Levels	Number of cases	Crude effect	R^2_{adj}	*p*
Age	per year	129	-1	0.00	0.22
Gender	female	57	*		
	male	72	3	0.00	0.40
Type of cerebral palsy	hemiplegia	46	*		
	bilateral	83	26	0.29	<0.01
Hearing loss	not deaf	124	*		
	Hearing loss	5	24	0.03	0.03
Vision loss	not blind	104	*		
	Vision loss	25	37	0.39	<0.01
Intellectual delay	no delay/IQ>70	74	*		
	IQ 50-69	13	18	-	<0.01
	IQ<50	26	40	0.54	<0.01
Seizures	no seizures	93	*		
	Seizures	28	17	0.09	<0.01
Speech problem	no speech problem	78	*		
	Speech problem	43	23	0.22	<0.01
GMFCS Level	GMFCS Level I	25	*		
	GMFCS Level II	43	15	-	<0.01
	GMFCS Level III	15	32	-	<0.01
	GMFCS Level IV	14	45	-	<0.01
	GMFCS Level V	23	57	0.80	<0.01
MACS Level	MACS Level I	14	*		
	MACS Level II	30	3	-	
	MACS Level III	18	20	-	<0.01
	MACS Level IV	13	42	-	<0.01
	MACS Level V	13	53	0.66	<0.01
IMD 2004	Quintile I	67	*		
	Quintile II	23	-5	-	0.35
	Quintile III	17	1	-	0.86
	Quintiles IV/V	20	8	0.01	0.15

*Baseline against which other levels of the same impairment are compared

Increasing age was associated with a small decrease in LAQ-CP summary score, indicating marginally less participation restrictions. The small additional participation restrictions for males compared to females was not statistically significant.

Figure 8.31 Scatter plot showing the effect of age on LAQ-CP summary score



Bilateral cerebral palsy was associated with increased participation restrictions compared to hemiplegia; similarly, hearing and vision loss, seizures and speech problems were also all associated with greater restriction in participation. Increasing severity of intellectual, movement and manual disability were, likewise, all associated with increasing restrictions in participation in univariate analyses (Figures 8.32 to 8.34). Increasing area deprivation showed a slight association with greater participation restrictions but the effect was not statistically significant.

Figure 8.32 Box plot showing variation in LAQ-CP summary score by intellectual delay

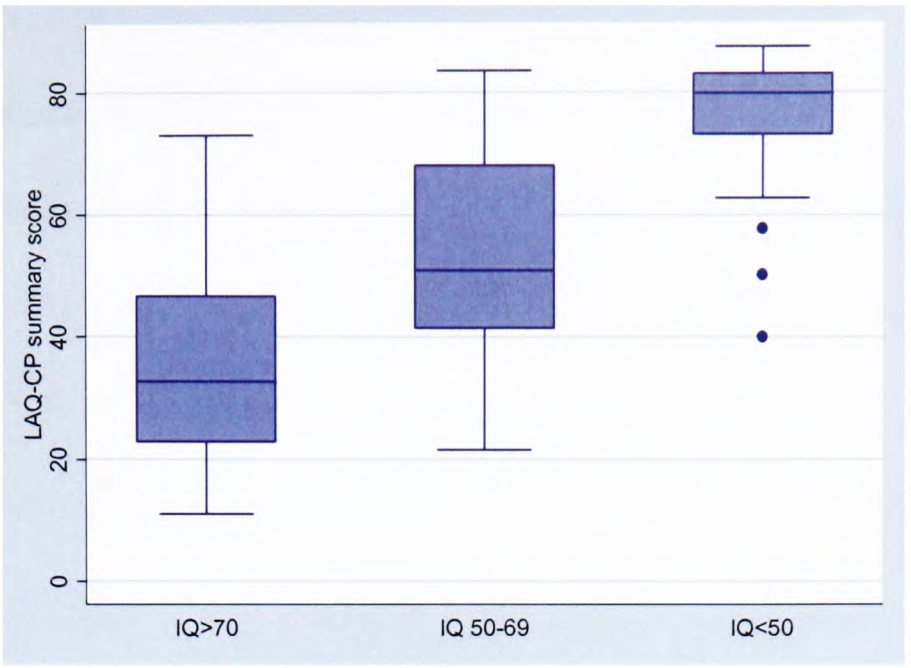


Figure 8.33 Box plot showing variation in LAQ-CP summary score by GMFCS

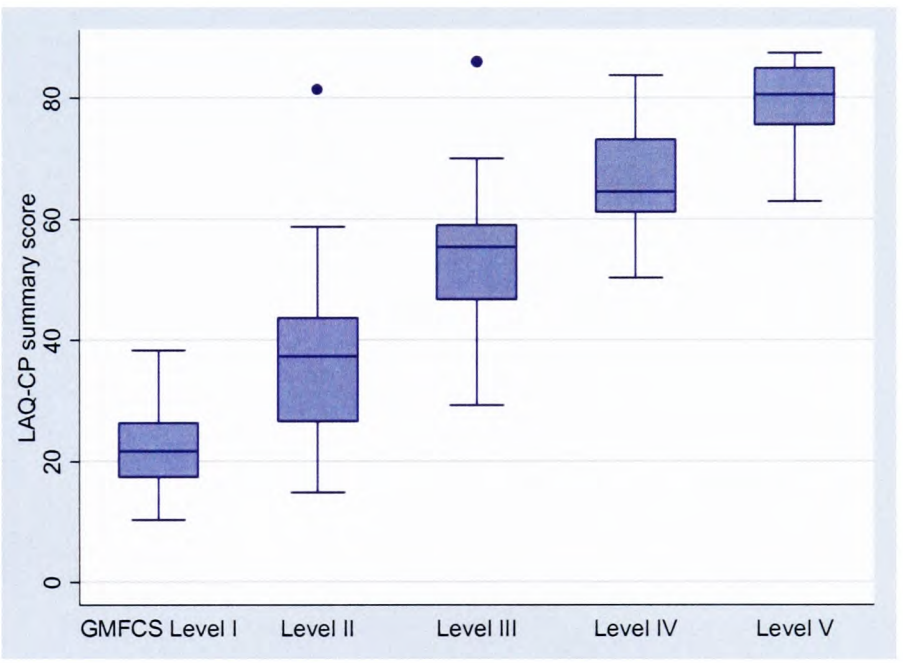
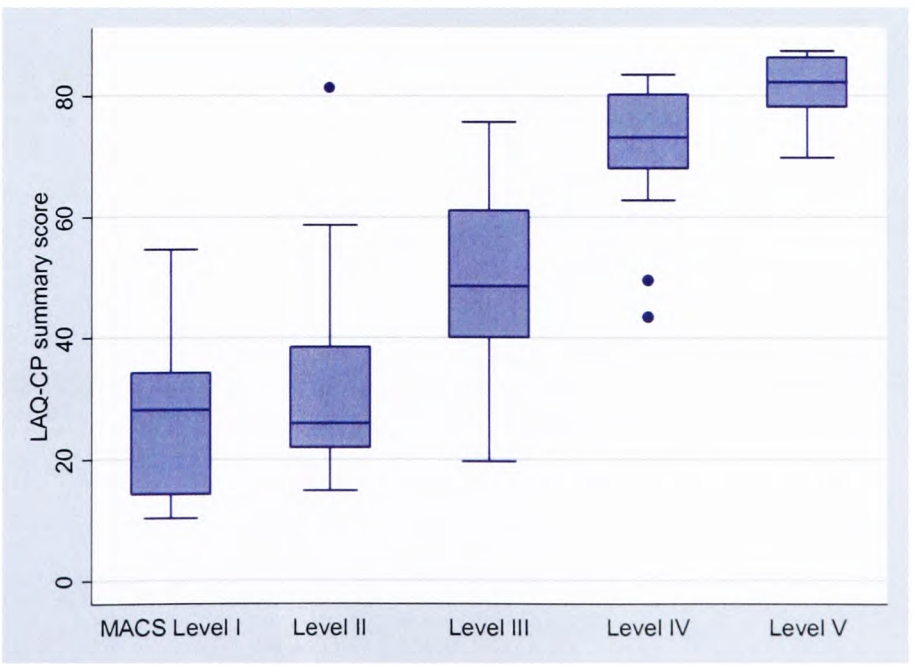


Figure 8.34 Box plot showing variation in LAQ-CP summary score by MACS



After controlling for the effects of other variables only movement and manual ability (GMFCS and MACS) remained significant in the model explaining 85% of the variance in LAQ-CP summary score ($F(8, 77) = 61.9$, $p < 0.001$). The effects are summarised in Table 8.9. Age, type of cerebral palsy, hearing and vision loss, seizures, intellectual ability and area deprivation were not statistically significantly associated with the LAQ-CP summary score when the effect of other variables was held constant. However, including intellectual disability in the model, although not reaching statistically significance, did explain a fraction more of the variance (86%).

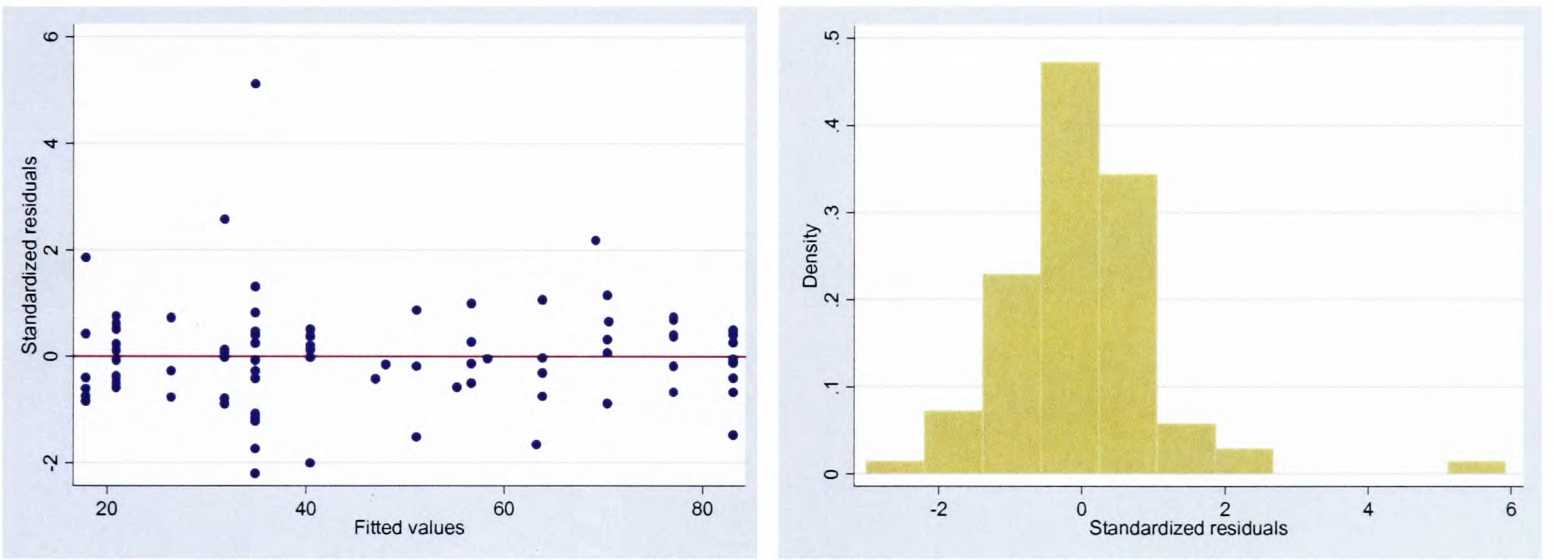
Table 8.16 Statistically significant mutually adjusted explanatory variables for LAQ-CP summary score.

Variable	n	Effect on LAQ-CP summary score		95% confidence intervals		*p* value
		Crude	Adjusted			
GMFCS Level I	25	*	*			
GMFCS Level II	43	15	14	9	19	< 0.001
GMFCS Level III	15	32	30	23	38	< 0.001
GMFCS Level IV	14	45	37	29	46	< 0.001
GMFCS Level V	23	47	44	34	54	< 0.001
MACS Level I	14	*	*			
MACS Level II	30	3	3	-3	9	0.31
MACS Level III	18	20	9	2	16	0.02
MACS Level IV	13	42	15	6	25	0.002
MACS Level V	13	53	21	10	33	< 0.001
Constant		-	*18*	*12*	*24*	< 0.001

*Baseline against which other levels of the same impairment are compared

The histogram in Figure 8.35 shows that the standardised residuals were fairly normally distributed. The plot of standardised and predicted (fitted) residuals shows one outlier who was the same child as for the models explaining the ASK and LAQ physical independence scores. This child recorded a high LAQ-CP summary score of 81, indicating very poor participation, despite being classified at GMFCS Level II and MACS Level II for which the model estimates a score of around 35 points. As mentioned earlier, unusually for GMFCS and MACS Levels II, this child requires 24 hour supervision, frequent lifting and to have everything done for her.

Figure 8.35 Histogram of standardised residuals and scatter plot of standardised with predicted (fitted) residual values from multiple regression of LAQ-CP summary score



The Breusch-Pagan / Cook-Weisberg test for heteroskedasticity was marginally statistically significant ($p < 0.08$) indicating that the variance in the residuals was probably constant, and that the model might be a reasonable fit.

This model suggests that the LAQ-CP summary score was predicted by movement ability (GMFCS) and manual ability (MACS), and a large amount of the variance (85%) was explained by these two factors alone.

8.10 FACTORS AFFECTING TYPE OF SCHOOL ATTENDED

The LAQ-CP scale for schooling score only includes two items: 'distance travelled to' and 'type' of school and, with so few items, the internal consistency cannot be determined in the usual way. A multiple regression was undertaken, as was conducted for the other LAQ-CP domains, in which only intellectual delay of IQ<70 remained statistically significant when the effect of ther variables was held constant but this only explained 35% of the variance. Of more interest, perhaps, is the type of school children attend: whether it is a mainstream school with or without a support unit, or a 'special school' for children with profound physical or learning difficulties. Tables 8.10 to 8.13 show to what extent each of the explanatory variables is associated with attending a particular type of school. Table 8.10 shows that 85% of children with hemiplegia attended a mainstream school whereas only half of the children with bilateral cerebral palsy did so.

Table 8.17 Type of school attended by distribution of cerebral palsy.

	Hemiplegia		**Bilateral**		***p* value**
	n	*%*	n	*%*	
	46	*36*	83	*64*	
Mainstream school	29	*63*	24	*29*	<0.01
Mainstream with support unit	10	*22*	19	*23*	
Special school	5	*11*	37	*45*	
Boarding school	2	*4*	3	*4*	

No children diagnosed with hearing loss and only one child with vision loss attended a mainstream school. There were four children with hearing loss in special schools and one child at a boarding school, and 21 children with vision loss were in special schools and three were at boarding schools, including one school especially for blind children.

Table 8.11 shows that the greater proportion of children in GMFCS Levels I, II and III attended mainstream school, although around a quarter in Levels I and II and half of children in Level III attended schools that had a support unit for children with disabilities. Half of the children in GMFCS Level IV and all but one child in Level V attended special or boarding schools.

Table 8.18 Proportion (%) of children in each GMFCS Level attending each type of school

	GMFCS Level				
	I	II	III	IV	V
	n=32	n=43	n=15	n=14	n=23
Mainstream school	20	24	4	4	-
Mainstream with support unit	9	10	7	2	1
Special school	2	8	3	7	21
Boarding school	1	1	1	1	1

Table 8.12 shows that the greater proportion of children in MACS Level I, II and III attended mainstream schools with around a quarter using schools with support units; whereas children with MACS Level IV and V predominantly used special or boarding schools.

Table 8.19 Proportion (%) of children in each MACS Level attending each type of school

	MACS Level				
	I	II	III	IV	V
	n=14	n=30	n=18	n=13	n=12
Mainstream school	8	19	9	-	-
Mainstream with support unit	5	8	4	2	-
Special school	1	3	5	9	11
Boarding school	-	-	-	2	1

Table 8.13 shows that almost all the children with an IQ of 70 or greater attended mainstream school, whereas almost all children with IQ less than 70 attended some form of special school.

Table 8.20 Proportion (%) of children in each IQ category attending each type of school

	IQ>70	IQ 50-69	IQ<50
	n=74	n=13	n=26
Mainstream school	49	-	-
Mainstream with support unit	22	2	-
Special school	1	11	24
Boarding school	2	-	2

In summary 36% of children did not attend mainstream schools including five children who used special boarding schools. Children with bilateral cerebral palsy, hearing or vision loss, GMFCS Level V, MACS Level IV and V or intellectual delay (IQ<70) were more likely to utilise special schooling. Attempts to use logistic regression modelling to determine the odds of using special schooling by the explanatory factors were undermined by the small or absent numbers of children in some categories

8.11 CHAPTER SUMMARY

In general, all the explanatory variables were statistically significantly associated with all the outcomes in univariate analyses except gender and area deprivation. The crude effects of variables were moderated when they were included in multivariate analyses and therefore the mutually adjusted effect on outcome scores that was highlighted (Table 8.14). The regression models for the ASK and LAQ-CP domains for Physical Independence and Mobility and the LAQ-CP summary score, appeared to be good fits and explained a large amount of the variance. The indices of children's movement and manual disability (GMFCS and MACS) and intellectual delay (IQ) were incorporated into each of these models, whilst seizures and speech problems explained some of the variance, and age had a minor but pervasively statistically significant effect on outcome scores.

The Clinical and Economic Burden domain scores from the LAQ-CP instrument were not well explained by the explanatory variables used in the analysis. However both Clinical and Economic Burden were to some extent associated with severity of movement disability. Clinical Burden score was notably affected by children in GMFCS Level V, and to a lesser extent Levels II to IV, compared to GMFCS Level I. Economic Burden score appeared to be elevated in GMFCS Levels III to V compared to Level I. However, these models did not fit the data well. The model predicting Social Integration score included movement ability (GMFCS Levels II to V compared to Level I) and intellectual delay (IQ < 70 compared to IQ ≥70), was a good fit but explained only 36% of the variance. Descriptive statistics were used to examine factors affecting the type of school children attended. Children with bilateral cerebral palsy, hearing or vision loss, GMFCS Level V, MACS Level IV and V, or intellectual delay (IQ < 70) were more likely to utilise special schooling.

Table 8.21 Statistically significant variable coefficients (standard error) for each of the indices of activities and participation.

Variable	Levels	N	ASK	LAQ-CP summary	Physical Independence	Mobility	Economic burden	Clinical burden	Social Integration
Variance explained (R^2_{adj})			**0.89**	**0.85**	**0.89**	**0.77**	**0.56**	**0.32**	**0.35**
Baseline			77 (9.7)	18 (2.9)	18 (8.9)	18 (2.3)	6 (4.9)	27 (5.0)	24
Age	Per year	129	2 (0.9)		-2 (0.8)			-1 (0.5)	
Gender	Female	57							
	Male	72							
Type of CP	Hemiplegia	46							
	Bilateral	83							
Hearing loss	Not deaf	124							
	Hearing loss	5							
Vision loss	Not blind	104							
	Vision loss	25							
Intellectual delay	No delay/IQ>70	74	*		*				*
	IQ 50-69	13	-10 (5.7)		21 (5.5)				11 (5.2)
	IQ<50	26	-15 (6.1)		6 (5.9)				13 (6.3)
Seizures	No seizures	93	*			*			
	Seizures	28	-9 (3.9)			6 (2.9)			
Speech problem	No Speech Problem	78			*				
	Speech Problem	43			10 (3.7)				
GMFCS Level	GMFCS Level I	25	*	*	*	*	*	*	*
	GMFCS Level II	43	-20 (4.0)	14 (2.7)	16 (3.9)	1 (2.1)	3 (4.6)	8 (2.3)	13 (4.2)
	GMFCS Level III	15	-47 (5.5)	30 (3.8)	33 (5.1)	37 (4.9)	23 (6.4)	11 (3.1)	27 (5.5)
	GMFCS Level IV	14	-55 (6.2)	37 (4.2)	44 (6.4)	48 (4.1)	29 (7.0)	12 (3.2)	20 6.3)
	GMFCS Level V	23	-60 (7.9)	44 (5.2)	54 (7.7)	59 (3.7)	27 (8.7)	19 (2.7)	22 (7.5)
MACS Level	MACS Level I	14	*	*	*		*		
	MACS Level II	30	-6 (4.4)	3 (3.0)	9 (4.3)		2 (5.1)		
	MACS Level III	18	-13 (4.9)	9 (3.5)	23 (4.9)		4 (5.9)		
	MACS Level IV	13	-14 (7.1)	15 (4.8)	20 (6.9)		16 8.0)		
	MACS Level V	13	-16 (8.8)	21 (5.7)	31 (8.5)		20 (9.6)		
IMD 2004	Quintile I	67							
	Quintile II	23							
	Quintile III	17							
	Quintiles IV/V	20							

* Baseline against which other levels of the same impairment are compared; empty cells indicate variable was not statistically significantly associated with outcome

8.12 RESULTS SUMMARY

As described in *Chapter 6*, families of 40% of the children invited to be part of this research participated fully in the study. These children did not differ from all the surviving children in the complete geographically-defined population who did not take part in the full survey by their age, gender or the characteristics of their cerebral palsy or associated impairments. Families residing in areas corresponding to the least deprived national quintile for the Indices of Multiple Deprivation (IMD 2004) were more likely to participate.

The analyses described in *Chapter 7* showed that the reliability of family assessment of children's movement and manual ability, using the GMFCS and MACS, compared to health professionals was excellent. Reliability was higher when health professionals made their assessment by direct observation rather than by reviewing clinical records. The performance of the ASK and LAQ-CP instruments in this study was examined and found to be acceptable.

As described in *Chapter 8*, ASK scores were predicted by age, movement and manual ability, intellectual delay and seizures. The LAQ-CP domain of Physical Independence was predicted by age, movement and manual ability, intellectual delay and speech problems. Mobility was most associated with movement ability, and marginally with manual ability and seizures. Clinical and Economic Burden were to some extent associated with movement ability, however these models did not explain much of the variance. Similarly, Social Integration was associated with movement ability and seizures but the regression model only explained a limited amount of the variance in scores. The LAQ-CP summary score was predicted by movement and manual ability in a more robust model. Deprivation in the families' area of current residence using the IMD did not significantly affect any outcomes.

CHAPTER 9

THAMES VALLEY SURVEY

DISCUSSION

9.1 CHAPTER OVERVIEW

This chapter discusses the methodology and findings of the study undertaken in the Thames Valley with reference to the aims, objectives and research questions set out in *Chapter 3*. To reprise briefly the four aims were (i) to conduct a survey with a complete geographically-defined population of families with children with cerebral palsy; (ii) to determine the reliability of family report for the Gross Motor Function and Manual Ability Classification Systems (GMFCS and MACS); (iii) to measure and describe children's activities and participation; and (iv) to examine the effect of disability on the children and their families.

9.2 SURVEY SAMPLE AND POTENTIAL BIASES

9.2.1 Invitation bias

The survey was accomplished with support from the National Perinatal Epidemiology Unit (NPEU) which hosts *4Child*, hence there were no institutional barriers to using the database. Families' current addresses and GP details were identified through the NHS National Strategic Tracing Service (NSTS). The tracing identified that one child had died and addresses were not available for four children; GPs then declined access to contact the families of a further eleven children. Therefore, despite beginning with a complete geographically-defined population, attrition from the study became inevitable and creates uncertainty as to whether the sample that participated was representative of the complete population. This is a potential challenge to the 'external validity' of the research, in terms of the extent to which the findings can be generalised to populations outside of this study, as opposed to 'internal validity' which concerns only what happens within the study. A further seven invitations were returned as undeliverable leaving only 95% of the surviving children's families in the survey population. This is the first potential source of bias, in that the 5% of children and families who were not invited to take part may be different in some

important way from those who were invited. The GPs who provided a reason for excluding families cited either the children or parents being physically or psychologically unwell, referred to their challenging social circumstances, or in one case indicated that the family was abroad. It is therefore possible that some of the children excluded have more severe cerebral palsy or it could be that the parents were not coping. If this were the case then any findings may underestimate any adverse effects of children's disability.

9.2.2 Response bias

The second potential source of bias was 'response'. The response reported depends upon which numerator and denominator are selected. Bowling recommends using the number of subjects who could have possibly responded as the denominator (Bowling 1997). Therefore, for this study, although the families of 321 children were invited, as seven invitations were returned undelivered, the denominator selected was 314 children. The numerator for response to the initial invitation could include the families of 34 children who actively responded but indicated they did not wish to take part; the numerator could also include families of 51 children who eventually returned a rating only of their child's GMFCS Level. If all the 'responses' are included in the numerator then the response was 212/314 (68%). Of these, 175/314 (56%) provided an eligible classification of the child's movement ability using the GMFCS. However, the families of 139/314 (44%) of the children invited to take part initially gave their consent to take part in the survey and of these, 129/139 (93%) subsequently returned completed questionnaires. The final sample of children whose families were invited and fully participated in the survey, and who are therefore represented in most of the subsequent statistical analyses was 129/314 (41%); or 129/337 (38%) of the complete geographically-defined and surviving study population.

The response that can be expected to a postal survey is dependent on the context of the survey and whether the target population feel the aims warrant their time to complete the questionnaire (Edwards et al. 2002). The Picker Institute Europe has conducted fifteen large national surveys of NHS patients between 1998 and 2004 and reported responses from 42% to 75% (Picker Institute Europe 2005). In their survey that focused particularly on "young patients" (less than 17 years old) the average response was 50% and the response from individual NHS Trusts ranged from 32% to 64% (Ramm et al. 2004). Therefore the response to the Thames Valley study is within the range typically reported by the Picker Institute Europe, albeit towards the lower end.

The LAQ-CP is routinely used in the North of England Collaborative Cerebral Palsy Survey (NECCPS) and typically achieves approximately a 70% response (A. Colver, personal communication), although a response of 92% was reported in a related paper by Hammal et al. (2004) of which 86% of the questionnaires were completed. In most circumstances, however, those families are forewarned to expect the questionnaire by the paediatrician who provides the child's details to the NECCPS when the children are around 4 to 5 years old. This differs from the Thames Valley study where the invitation to take part in the study will have arrived unexpectedly and unsolicited from an unknown source, for children who were between 6 and 12 years old. To address these issues details about the aims of the Thames Valley study and background to the research and researchers were presented in the invitation letter and information sheets with reference to further information available on the study website. However, whether families that did not respond actually read the information or even received these documents cannot be determined. The method of enclosing all the questionnaires at the same time as mailing the invitation, as was done in this study for families living outside the Thames Valley, versus gaining consent from families prior to

mailing the study questionnaires as was done for families residing within the Thames Valley, had no apparent effect on response.

A survey asking about access and use of physiotherapy services conducted with families of children known to the Northern Ireland Cerebral Palsy Register achieved a response of 85% (Parkes et al. 2002). This study only invited children with "abnormal gait" or no walking ability as they were the children who were likely to be receiving therapy. The high response may reflect that provision and frequency of physiotherapy for children with cerebral palsy is often an emotive issue with many families believing that their child would benefit from more input. Parkes and her colleagues reported that 90% of families with children in mainstream schools or schools for children with physical disabilities, and 75% overall, indicated they wanted more physiotherapy. Therefore the purpose of the Northern Ireland survey was perhaps more overtly in keeping with the priorities of families than the present survey in the Thames Valley, and focused more precisely on the families of children likely to be more dependent on that particular service.

In contrast, the Thames Valley survey included all of the eligible children born to a mother resident in a defined geographical area at the time of birth and had an aim that may have been perceived as less relevant and important to families. The study's short title 'Measuring Children's Abilities' was used on all correspondence and the information sheets suggested how children could take part by signing the consent form themselves and then by answering the questionnaires. The topic of the study may not have appealed to all of the children although efforts were made to produce colourful and appealing documents. If the child was unable or did not want to take part then the family may not have taken any further interest even though their own participation in study would have been useful. It is possible that

some families may not have wanted to take part in a study that highlighted the activities their child was unable to perform. Again, these issues were addressed in the information sheets and also emphasised the importance of including all children whatever their abilities.

In a survey with families of preterm infants to validate a family-assessed measure of children's cognitive abilities Johnson et al. (2004) reported a response of 84/144 (58%). However, of these, only 64 children were subsequently included in their statistical analyses reducing the proportion of full participants to 44%; this is closer to the proportion of full participants in the Thames Valley study. The aim of the survey by Johnson et al. was to validate a family-assessed instrument as was one of the aims of the Thames Valley study. This lends weight to the possibility that one reason for the lower response to these surveys compared to those by Parkes and her colleagues (2002) was that families perceived the aims as less directly relevant to them.

Examining the number of children in each family-assessed GMFCS Level shows that 60% of the children were in GMFCS Levels I and II. It is possible that the survey attracted a greater response from families of children who are more able, as was suggested in the comparison of full and non-participants using *4Child's* indices for upper and lower limb function. The survey of non-participants in the Thames Valley requesting only a GMFCS assessment displayed the same pattern of ability amongst responders, as 50% of the children from families that replied to the later mailing were in GMFCS Levels I and II and maintaining the overall proportion represented in those levels at 59%. Overall there were substantially fewer children in GMFCS Levels III and IV. To determine whether this was a response bias, towards the more able children taking part in the survey, or whether there are proportionately fewer children in Levels III and IV, reference was made to three studies that have reported

the profile of GMFCS in geographically-defined populations. Beckung and Hagberg (2000) reported the GMFCS profile for a population of children born in 1991 and 1992, then five to six years old, and living in a western Sweden. The same authors later reported the GMFCS profile for the same area but this time included children born between 1991 and 1994, then five to eight years old (Beckung and Hagberg 2002). More recently, Parkes et al. (2005) have reported the GMFCS profile for the population of children recorded on the Northern Ireland Cerebral Palsy Register born between 1981 and 1997 using an algorithm based on the children's abilities recorded on their standard assessment form (Evans et al. 1989). The proportion of children reported in each GMFCS Level in these studies is shown in Table 9.1 alongside the findings from the Thames valley study for the final study sample (available for 127/129 children) and all children for whom a family-assessed GMFCS classification was available.

Table 9.1 Comparison of proportions (%) reported in each GMFCS Level from population-based studies

Method	**Thames Valley all** ***family-assessed***	**Thames Valley final sample** ***family-assessed***	**Parkes et al. (2005)** ***algorithm***	**Beckung & Hagberg (2002)** ***professionally-assessed***	**Beckung & Hagberg (2000)** ***professionally-assessed***
Total N =	*175*	*127*	*938*	*233*	*116*
Missing (%)	*48*	*62*	*5*	*3*	*6*
% of study participants reported in each GMFCS Level					
Level I	25	25	14	41	38
Level II	32	34	46	20	22
Level III	10	12	11	7	8
Level IV	10	11	7	16	17
Level V	23	18	21	16	15

It can be seen from Table 9.1 that there were strikingly similar proportions found in all of these studies in combined GMFCS Levels I and II, 57%, 59%, 60%, 61% and 60%, and relatively fewer children classified in Levels III and IV. Discrepancies in the proportions reported between Levels I and II could probably be attributed to the different methods of assessment in each study. Beckung and Hagberg (2000 and 2002) used professionals to assess the GMFCS and report more children in GMFCS Level I, whilst in the Thames Valley study families classified children and report more children in GMFCS Level II. As has been noted already, GMFCS Levels I and II appear to be the most difficult to distinguish from each other (Palisano et al. 1997; Morris et al. 2004). Not only does this finding further strengthen the case that the sample in the Thames Valley study is likely to be representative of the complete population, but also suggests that these proportions may in fact represent the profile of severity of cerebral palsy in complete geographically-defined populations. In this interest, the proportion of children in each GMFCS Level is shown again in Table 9.2, this time with 95% confidence intervals. This provides an estimate of the profile of movement disability in the Thames Valley population.

Table 9.2 Proportion (%) of children in each GMFCS Level, with 95% confidence intervals, in the sample of children for whom a family assessment was received.

GMFCS	% of sample N=175	95% confidence intervals	
		Lower	Upper
Level I	25	*16*	*32*
Level II	32	*25*	*39*
Level III	10	*6*	*14*
Level IV	10	*6*	*14*
Level V	23	*15*	*31*

Compared to the high responses to surveys in the north of England and Northern Ireland, the response in the Thames Valley was a little disappointing. However the higher response in those surveys probably reflects the more established relationships that exist between families and researchers. To determine whether the Thames valley sample could be considered representative of the study population, an extensive analysis was performed to compare the known characteristics of the children and families who did and did not take part. Children of families who fully participated did not differ from those who did not take part in terms of their age, gender or the characteristics of their cerebral palsy or any associated impairments. There was a marginal trend for the children of families who took part to have slightly better upper and lower limb function, according to the *4Child* indices, and higher IQ than children of families who did not take part but the differences were not statistically significant. These comparisons reassure us that, at least in terms of their age, gender mix, the characteristics of their cerebral palsy and other impairments, the children of families who fully participated can indeed be considered representative of the complete population and make the findings generalisable.

There was however a greater response from families residing in areas corresponding to the least deprived national quintile using any of three indices of area deprivation available for this study, including the IMD, Townsend and Carstairs' indices. This is in keeping with other surveys that have examined response patterns using area deprivation indices. For instance Roberts et al. (2004) reported reduced response with increasing area deprivation using the Townsend score, and Urwin et al. (1998) reported non-response from areas of higher deprivation measured by the Carstairs' index. The Thames Valley is "arguably the most prosperous sub-region of the South East" but does include pockets of area deprivation falling within the worst 20% in England and found in urban environs of Reading, Slough,

Wycombe, Oxford and Milton Keynes (South East England Development Agency 2004). The response was marginally lower from families residing in IMD national quintiles II to V, however the overall difference between deprivation in the areas that families who did and did not fully participate resided was not statistically significant. The families who fully participated in the survey can be considered representative of the complete study population in terms of area deprivation where families live. The consequent limitation is that, because the Thames Valley is generally a prosperous region, the findings may be biased by the high proportion of families living in less deprived areas relative to England as a whole. Therefore how generalisable the findings are to families of children with cerebral palsy residing in regions with different area deprivation profiles cannot be determined.

The best available evidence for increasing response to postal surveys was followed as much as possible. The correspondence was personalised and printed using coloured inks and outgoing and return envelopes used first class stamps. Incentives were not permitted by the ethics committee. The key issues affecting response to the invitation were probably that families were unfamiliar with *4Child* and that the topic of the survey was not of sufficient interest to families. The response from families who consented to receive the questionnaires was excellent and the small non-response amongst these families may have been, in part, due to the length of the questionnaires in the full survey. The overall response to this study increased by 16% when the single sheet GMFCS questionnaire was mailed on its own. This had been the method used to administer the questionnaire in the Canadian study which achieved a respectable response of 48% within three weeks after a single mailing without any reminders.

With reference to the first aim of this research, conducting a survey with a complete geographically-defined population of families with children with cerebral palsy, this was achieved with partial success. The moderate response to survey must be acknowledged as a potential limitation to the generalisability of the findings. However, the extent of any response bias has been examined, and no differences were found between the characteristics of full and non-participants. The conclusion is therefore that the findings are likely to representative of the complete population in the Thames Valley; however the wider generalisability is subject to the effect of any differences in area deprivation profiles.

9.3 RELIABILITY OF FAMILIES USING THE GMFCS

The reliability of families classifying children's movement ability using the GMFCS was first highlighted in the preparatory work carried out in Canada and described in *Chapter 4*. In that study, the fact that many of the families had previously discussed the GMFCS with health professionals (38%) and all had taken part in a longitudinal study using the GMFCS may have lead to an overestimation of the reliability of family report (Morris et al. 2004). However, in the Thames Valley study, only one in ten families that took part indicated having discussed the system with a health professional and some of the health professionals were also unfamiliar with the GMFCS. Of the 180 classifications provided by families, only five families did not indicate a single description corresponding to a GMFCS Level. This suggests that the questionnaire is easy to understand and that the descriptions of children's movement abilities are meaningful to families.

The response from health professionals nominated by families to provide classification of children's movement abilities using the GMFCS was exceptional, exceeding 90% for all the professional groups. This high response may be attributable to a sense of professional duty

to the families who, as the letter pointed out, were already taking part in the study and had nominated them personally, and/or that they were interested to help generally with research. All the health professionals subsequently received the same newsletters about the study as families which, from informal feedback, were appreciated. Representatives of all the key professional groups were not nominated for every child as not every child was receiving those services. The statistical analyses were therefore conducted as pair-wise comparisons between families and each professional group, and between each of the professional groups. Some children, those who have contact with more health professionals, were therefore represented in several of the analyses whilst other children, such as those who perhaps only see either a physiotherapist or paediatrician, are only included in one analysis. Children were classified in all GMFCS Levels by professionals, however, there were fewer children classified in GMFCS Level I by orthopaedic surgeons, indicating that perhaps the most able children were less likely to be consulting an orthopaedic surgeon. Several classifications were made by members of professions for which there were insufficient classifications to conduct pair-wise analyses; however, inspection of these data did not alter any of the findings or their inferences.

There was not absolute agreement between all families and each of the key professional groups. Values of absolute agreement ranged from 59% to 64%. The overall measures of chance-corrected agreement (κ) ranged from 0.47 to 0.54, which are considered fair to good agreement (Landis and Koch 1977). These indices of agreement were similar to those found between the professional groups in this study, which ranged from 50% to 72%, and chance-corrected agreement from 0.37 to 0.65. The highest agreement was found between families and physiotherapists, and by paediatricians and physiotherapists. All these indices of agreement are slightly lower than those reported by the developers ($\kappa = 0.75$) in their initial

study examining agreement between therapists for children between 2 and 12 years (Palisano et al. 1997). Examining the indices for individual GMFCS Levels there was highest agreement for GMFCS Level V ($\kappa = 0.8$) whilst there was lower and somewhat similar agreement for the other levels (all around $\kappa = 0.5$).

However, what is not reflected in the analyses of agreement using the kappa statistic is that the most frequent disagreement was between GMFCS Levels I and II. This concurs both with the developers' findings observing therapists (Palisano et al. 1997) and the study conducted with families and therapists in Canada (Morris et al. 2004), that Levels I and II were the most difficult to distinguish between. Notably, and again consistent with the earlier Canadian study, nearly half of the children that physiotherapists classified in Level I were classified in Level II, or less able, by their families whereas the reverse situation was rarer (Morris et al. 2004). A similar trend was observed between families and paediatricians. However the opposite pattern occurred between families and orthopaedic surgeons, the surgeons classified several children in Level II that families classified in Level I almost exclusively using information from the child's medical records.

There were only three occasions when families and any professionals disagreed by more than one GMFCS Level however the statistical methods used to determine absolute and chance-corrected agreement take no account of partial agreement. Reliability is conceptually different to agreement for ordinal and continuous measures and indicates the amount of error in the measurement. Reliability is defined as the ratio of the 'variability of the subjects being assessed' to the 'variability of the subjects being assessed' plus 'measurement error' (Streiner and Norman 2003). The correct method for determining reliability with ordinal data is by using either the Intraclass Correlation Coefficient (ICC) or weighted kappa, which

are equivalent when quadratic weights are used for the latter (Fleiss and Cohen 1973). The analyses presented in this dissertation were ICCs, with 95% confidence intervals. Very high reliability, of greater than or equal to 0.9 with narrow confidence intervals, was found between families and professionals, which was equivalent to the reliability between the different professional groups. As reliability coefficients are directly related to the variability in the subject population, as well as the competency of observers, the high reliability found in the first study in Canada was interpreted with some caution (Morris et al. 2004). However, having now replicated these findings in a second study, the claim that family report for the GMFCS is reliable can be asserted more confidently. The caveat remains that, despite the high reliability, families and professionals do not always completely agree, though the disagreement is usually by only one level.

Possible reasons for disagreement were discussed in *Chapter 4* following the first study (Morris et al. 2004). Principally, health professionals and families made their classifications using the GMFCS based on their knowledge of the child's movement abilities and the instructions provided. As the same child is being assessed one source of measurement error could be the abilities of the observers. However, based on the reports by Palisano et al. (2003) and Tieman et al. (2004a&b), it is likely that some children move differently according to where they are, for instance at hospital, school or in their home. This is in keeping with the ICF model that stresses that both personal and environmental factors interact with children's abilities resulting in their activities and participation. So it could be that whilst the child is the same, their movement abilities are perceived and experienced by family or professional observers as different because one group sees the child at home and the other observes the child at school or in the hospital setting. Almost certainly the family will have greater knowledge of the child's abilities generally and across a wider range of

environments than professionals. To further support this line of reasoning, the reliability between families and professionals was higher when professionals made their classification by direct observation of the child rather than by reviewing their clinical records. Therefore, although professionals classifying children using the GMFCS by reviewing their records has been shown to be reliable (Wood and Rosenbaum 2000), the level at which children are classified using that method may not always concur with a rating made after visually observing the same child. Medical records often contain varying amounts of information about children's functional ability and recording the GMFCS Level in future will improve inter-professional communication (Morris and Bartlett 2004). Knowledge of children's movement abilities is ultimately dictated by either asking explicitly about the child's performance in specific environments or by spending time with the child, in either case it is likely that the parents will know their children best.

For research purposes, some people may prefer to continue seeking the professionals' view of children's abilities thinking, unjustifiably, that the professional will be more objective. As discussed in the introductory chapters, objectivity is defined by what is being measured and not who undertakes the measurement, or in this case classification (McDowell and Newell 1996). What must be determined is the amount and nature of any measurement error introduced by different types of assessor and the results presented in this thesis demonstrate that there is no more or less error in family versus professional classification. For scientific rigor, it is not crucial whether families or professionals classify children in research studies but authors of scientific papers should now, for clarity, state explicitly in the methods who undertook the classification. In addition, perhaps monitoring the reliability between families and professionals is important if professional assessment is used, especially as there would be minimal additional costs to do so.

In the clinical context, providing family-centred services requires professionals to enter into partnership with families and recognise parents as the experts regarding their child's needs. This approach is typically associated with better health outcomes for both children and their parents (King et al. 2004). The reliability coefficient for classification of the GMFCS by families (0.9) meets the criterion suggested for using measures with individuals as well as for groups (Fitzpatrick et al 1998). Gathering a classification from a family of their opinion of the child's abilities using the GMFCS may empower them and help them feel that their opinion matters. If this information were gathered prior to a consultation, any disagreement could then be discussed and might prove enlightening for either parent or professional, or indeed for both. The resulting collaboration might provide a basis for informing families about prognosis as described by Rosenbaum (2002), and improve the care of the child by enabling appropriate family-centred treatment or physical management to be planned appropriate to their GMFCS level (Bower 2001). If it is known in advance that a child is less able and more dependent on mobility aids and postural management equipment then perhaps the health professionals providing those services could be available at the same time of a hospital appointment. This might reduce the number of appointments the child attends which is another factor in making services more family-centred (Law et al. 2003).

When presenting these findings to colleagues and clinicians, before showing the results, it was interesting to ask audiences whether they thought families would consider their children more or less able than professionals. Whilst some people suggested that families would overestimate their child's abilities, presuming they would want to think the best of their child, others thought families would rate their children less able, perhaps believing that this would enable them to receive more intervention or attention. The findings show that, apart

from the disagreements between GMFCS Levels I and II discussed already, there was little evidence of any systematic bias one way or the other.

That a family can reliably report the GMFCS for their child simply adds to the number of ways in which these data can be collected, and this method will usually be inexpensive compared to a clinician making an assessment in person. Following publication of the reliability study conducted in Canada (Morris et al. 2004), several researchers have sought to use the family-assessed method for classification of the GMFCS. These include two groups in Australia, one developing a condition-specific quality of life measure (E. Davis, personal communication; Waters et al. 2005), and another examining the impact of transition to high school on children's participation (C. Imms, personal communication). There have also been requests to use the measure from researchers in the USA (D. Wright, personal communication) and Canada (D. Maltais, personal communication).

The reliability studies described in this thesis only included children between 6 and 12 years old and the GMFCS has several other age categories. Therefore it is noteworthy that, using modified descriptions of children's abilities for those ages, colleagues in Northern Ireland are examining the reliability of families classifying children between 4 and 6 years old (C. Kerr, personal communication), and colleagues in Canada found promising reliability for families classifying small number of children between 2 and 4 years old (D. Bartlett, personal communication).

9.4 RELIABILITY OF FAMILIES USING THE MACS

This thesis reports only the second study examining the reliability of the Manual Ability Classification System (MACS). In their latest paper, the developers of the MACS report excellent reliability between therapists for 168 children (ICC=0.97, 95% CI from 0.96 to 0.98) and between families and therapists for 25 children (ICC=0.96, 95% CI from 0.89 to 0.98) (Eliasson et al. In press); note the wider confidence interval for the latter due to the much smaller sample. In the Thames Valley study families and health professionals were asked to classify their child's manual ability using the same standard instructions for the MACS. These surveys achieved a marginally lower response from professionals than for the GMFCS but still high, ranging from 85% from physiotherapists to 93% from paediatricians. The families of only 24 children nominated occupational therapists, a surprising and relatively low number in comparison to the other professions, especially as the focus of occupational therapists are more often involved in assessing and improving children's manual skills. This finding is also of considerable concern as the goals of occupational therapy are generally focussed towards overcoming environmental barriers that limit peoples' participation in life situations.

Only three out of 91 responses from families did not indicate a single level which suggests that families found the instructions reasonably easy to interpret; only one family commented that they did not "find the leaflet helpful or clear enough". However the health professionals made a number of comments on the difficulties they encountered in making classifications using the MACS. One comment made by several professionals was that children had a certain level of manual ability but that their cognitive or learning disabilities were the reason they could not usefully handle objects. Other professionals commented on variability in children's level of functioning depending on the objects being handled; variability from day

to day or between the beginning and end of day due to fatigue; insufficient detail recorded in the medical record to make a classification; and some found classifying children with hemiplegia more difficult. Most of the issues raised were actually dealt with in the user instructions and it was evident that some professionals were uncertain how to classify partly due to their unfamiliarity with a new system. However several therapists were very interested in using the MACS and sought further information regarding its development.

Notably all the observing groups, families and professionals, classified more children in MACS Level II than other levels. Coincidentally a similar pattern was seen in the inter-observer reliability study undertaken by the developers (Eliasson et al. In press). Similar to the findings from the study of families classifying children using the GMFCS, there was not absolute agreement between families and professionals or between professions for all children. The exception involved occupational therapists and physiotherapists, who concurred exactly for the 14 children for whom classifications were made by both kinds of therapist producing an unusual reliability coefficient of 1 (κ and ICC). It is not clear if the MACS really is that reliable, whether this happened by chance as only a small number of children were included, or whether they conferred or were otherwise cognisant of one another's response prior to submitting their classifications. Whilst this may confound the analysis of reliability between these professions it would suggest that they did agree on a level for those children, and that is probably more how the system would be used in practice.

Looking at the disagreements between families and professionals, there was a trend for families to classify children as less able than did professionals but usually only by one level; there were a few instances were observers disagreed by two levels. When they occurred disagreements were most often for the more able children being classified in MACS Levels

I, II and III. The indices of absolute and chance-corrected agreement, and reliability, were slightly lower than were found for the GMFCS but these were still in the range that would infer 'moderate to fair agreement' and good reliability for use with groups (ICC=0.7) (Fitzpatrick et al. 1998). The indices of reliability between families and physiotherapists, and paediatricians and physiotherapists, also border on the acceptable range for use with individuals (ICC $\geq$0.9) (Fitzpatrick et al. 1998), and especially if occupational therapists and physiotherapists are involved together. Several of the analyses include very small numbers, for instance examining chance-corrected agreement in MACS Level III and the reliability between occupational therapists and other groups of observers. In these instances any disagreement has a more profound effect on the agreement or reliability statistic and is thus susceptible to greater measurement error.

The latest version of the MACS instructional leaflet being publicised by the developers has been edited slightly from the original version used in this study. Chiefly three key points from the instructions have been presented on the front page. These are, firstly, that the MACS should be judged on a child's performance not their best capability; and secondly that it is the ability to 'handle objects' that should be classified rather than the function of either hand on its own. This second point was one issue that had confused several observers in the Thames Valley study, and particularly when classifying children with hemiplegia when one hand may have significantly better functioning than the other hand and may also used their teeth to assist in manipulating objects. In the Thames Valley study a child without any hands was excluded from the survey using the MACS because the family may not have interpreted the request in the manner in which it was intended. A child with no hands may still usefully 'manipulate' objects using their forearms, feet or teeth, however this may not consistently be interpreted as 'handling' objects although this is precisely the activity that the

MACS aims to classify. The wording is therefore vital to ensure the system is valid and that the instructions are clear; the wording in the present version may not yet be as precise as it needs to be. Thirdly, as the age range has been expanded from children aged 8 to 12 years to now include those between 4 and 18 years old, a note has been added for observers to incorporate an age-related perspective when making classifications using the MACS. This is, however, left to the observer's intuitive judgment in the present version of the MACS leaflet.

Overall including the MACS in the Thames Valley survey enabled an additional and valuable aspect of children's physical abilities to be measured using family assessment. Another family-assessed method for measuring the manual ability of children with cerebral palsy, called the ABILHAND-Kids, has since been published (Arnould et al. 2004). This is a parent-assessed measure of the manual ability solely intended for children with cerebral palsy which has been developed using Rasch analysis. The final version includes 21 items which have an underlying order of difficulty, although this is not the order in which the items are presented on the questionnaire. The ABILHAND-Kids produces a score on a functional scale, more similar in concept to the GMFM than a classification system like the MACS. The ABILHAND-Kids can be used to discriminate between children's manual abilities and to measure any change over time (Arnould et al. 2004). This presents an opportunity to study any correlation between MACS Levels and scores from the ABILHAND-Kids as was done between the GMFCS and GMFM in the Ontario Motor Growth Study (Rosenbaum et al. 2002). Monitoring developmental changes over time may further elucidate the abilities of children in each MACS Level and thereby improve the precision of the descriptions used to report children's abilities in each level.

The second aim of this research, determining the reliability of family report for the GMFCS and MACS, was successfully achieved. Evidence of the excellent reliability of families classifying their children's movement ability using the GMFCS has been demonstrated in this thesis in two separate surveys. The excellent reliability of families assessing their children's manual ability using the MACS demonstrated in the Thames Valley study supports similar evidence already presented by the developers (Eliasson et al. In press).

9.5 THE UTILITY OF FAMILY-ASSESSED MEASURES

The findings in this study add to an increasing body of evidence that family-assessed instruments offer a reliable, as well as an economical and expedient method for gathering information about children's functional abilities. The fact that families and professionals do not always completely agree emphasises the utility of employing those people who know the child best - their families - to make the assessment. That is, of course, providing the criteria of validity and reliability of the method are assured. A response from families cannot be assured; but could probably be expected to be better than reported in the study in this thesis if the information were requested for a child as part of routine clinical practice or whose family had already consented for them to take part in a research study.

Family assessment of the GMFCS and MACS adds to a battery of instruments that can be used to measure the important outcomes of perinatal and neonatal clinical trials from the families' perspective. For instance, a family-assessed instrument measuring cognitive ability (Johnson et al. 2004) will be used in the follow up of children taking part in the *Programs* trial who were born preterm and at risk of infection (NPEU 2004). Another clinical trial, the *TOBY* study, which is investigating whether total body cooling helps to reduce disability in children who experience perinatal asphyxia, will employ professional assessment including

the GMFCS as an outcome initially when children are eighteen months old (NPEU 2004). Family assessment may be useful for longer term follow up of these children in the future.

Whilst the discrete aspects of children's movement, manual and cognitive abilities can be assessed using separate measures, these issues could also be assessed in a single instrument. The Health Status Classification System for preschoolers (HSCS-PS) is a new 12-attribute multidimensional family-assessed measure for preschool children which includes each of these elements and has the potential to be scaled as a utility instrument (Saigal et al. 2005). The decision about which instrument to use in particular circumstances may not always be straightforward and will largely depend on the purpose of the research. However, there is little doubt that family-assessed measurement of children's abilities using the GMFCS and MACS is efficient, reliable and extremely useful.

The study described in this thesis focussed upon children's activities and participation and proposed that, as this is an objective construct, no distinction needed to be made between child self-report and proxy reports by families. However, as quality of life is fundamentally a subjective phenomenon it preferably requires the response of the children themselves. An investigation using the PedsQL quality of life instrument particularly with children with cerebral palsy has recently been published (Varni et al. 2005). This study showed higher concordance between children and their parents for the PedsQL scales for indices of physical and social functioning but considerably lower concordance for children's emotional functioning. The finding is also consistent with the majority of other studies that have examined child and proxy concordance for quality of life measures (Eiser and Morse 2001). So, whilst activities and participation can either be reported by the child or a person that

knows them well, ratings of children's emotional and psychological wellbeing should, whenever possible, be obtained from the children themselves.

9.6 MEASURING CHILDREN'S ACTIVITIES AND PARTICIPATION

An increasing number of family-assessed health status instruments are available and the selection of instruments for a particular circumstance depends primarily upon the purpose of undertaking the measurement. For the Thames Valley study a structured review was used to identify instruments appropriate for measuring the 'activities and participation' of children with cerebral palsy. The further criteria of validity, reliability, precision, interpretability, acceptability, and feasibility recommended by Fitzpatrick et al. (1998) were employed to make the final selection. The responsiveness of instruments was not assessed in detail as the Thames Valley survey was a cross sectional study. The ASK and LAQ-CP were subsequently selected as the best available instruments and permission to use them for the Thames Valley study was granted by their developers. The instruments were administered by mail and the data managed relatively easily using software provided by the developers of the LAQ-CP and a custom database for the ASK. No problems were encountered regarding the feasibility of using the instruments in the survey.

The performance of the ASK and LAQ-CP measurement scales in the present study was monitored and found to be acceptable in terms of the internal consistency, convergence between similar constructs and divergence between unrelated constructs. Several families of children in GMFCS Level V, those who are least able to move, indicated that they felt the ASK was not appropriate for their children as they were unable to achieve any of the items included. There was a consequent floor effect where most of these children scored zero. This had not occurred in the development of the ASK as these children were excluded

(Young et al. 2000). Whilst those children could not be identified in advance of requesting they complete the questionnaire in this survey, the ASK clearly does not meet the criterion of acceptability for families of children in GMFCS Level V. The ASK scores were easy to interpret, with high scores indicated better physical functioning equating to greater activities and participation. However, slightly confusingly, higher scores for the LAQ-CP represent greater activity limitations and participation restrictions.

Despite the overall satisfactory performance of the instruments, the domains of the ICF Version for Children and Youth are not comprehensively covered by the items and scales of the ASK and LAQ-CP. The most striking omissions are any items included in the major ICF domains for Learning and Applying Knowledge, Communication and little representing Interpersonal Interactions and Relationships. As regards the latter, the LAQ-CP includes one item asking "how many friends the child has seen" as part of the domain for Social Integration. However, elements of the omitted domains are incorporated to some extent in other patient-assessed instruments and some items could perhaps have been incorporated for descriptive purposes to augment the ASK and LAQ-CP. The other domains of the ICF are however well covered by the items in the ASK and LAQ-CP even though the domain scales of the LAQ-CP are constructed according to the domains of the ICIDH, the ICF's predecessor (Mackie et al. 1998a). Therefore, items from the LAQ-CP would have to be reorganised into the categories of the ICF domains which they best represent.

The Mobility domain would not necessarily need to change but the items in the LAQ-CP domain for Physical Independence would be split between Self Care and Domestic life. Items in the scale for Economic Burden and Schooling would fit with Major Life Areas; however, items in the Clinical Burden scale are perhaps more reflective of environmental

factors within Services, Systems and Policies that contribute to, rather than define, children's activities and participation. Items in the ASK could be allocated across most of the domains reflecting the broad approach to children's physical functioning that was taken when the items were being selected for the instrument (Young et al. 2000). Reallocating items from the ASK and LAQ-CP, and finding new items to cover the areas not currently represented in order to develop a more comprehensive instrument for measuring children's activities and participation would require substantial validation and is beyond the scope of the research described in this thesis. However, developing and testing such an instrument would be extremely useful and provide an alternative to structured interviews such as those advocated by Kronk et al. (2005).

Children in this survey experienced a wide range of activity limitations and participation restrictions. The survey included children with all levels of manual and movement ability and their scores on the ASK and most LAQ-CP scales of the ranged from the minimum to the maximum. The two LAQ-CP scales where the maximum score was not observed were Clinical and Economic Burden; however this might reflect the level of burden described at the extreme ends of those scales. The extreme circumstances for two of the Clinical Burden items indicated by none of the respondents were spending six or more months in hospital or having four or more operations in one year. Trends for the health care of children may have changed since the measure was originally developed and perhaps children with cerebral palsy do not spend as long in hospital or have as many operations. In the Economic Burden scale, few respondents used the response options indicating maximum burden to the questions asking about any adaptations to the house over "the last year". It is perhaps unlikely that ten or more adaptations to the house would have been necessary in one year. The iface validity of these items is also undermined by the fact that the family may have

purchased equipment, moved home, or adapted their house in previous years. The rescaling of these items should therefore be considered. Scores from the ASK ranged from zero to the range expected for children without any impairments, which is 93 (SD 6.5) (Plint et al. 2003), indicating that children with cerebral palsy do not necessarily experience activity limitations exceeding those of unaffected children.

In summary, in terms of the third aim of this research, to measure and describe children's activities and participation, the survey was fairly successful. The survey enabled a wealth of information to be gathered describing to what extent children performed a range of relevant activities and participated in a variety of life situations. However, not all the domains in the ICF were assessed due to limitations inherent with the family-assessed instruments presently available and selected for use in the survey.

9.7 THE EFFECT OF DISABILITY OF CHILDREN AND FAMILIES

The fourth aim of this research was to examine the effect of disability on children and their family's life using the ICF framework. The findings of the Thames Valley study concur with similar studies that used professionally-assessed instruments to explore associations between impairments, activities and participation (Beckung and Hagberg 2000 and 2002). In general, whether professionally or family-assessed instruments are used, indices of activities and participation for children with cerebral palsy appear to be most associated with, in order of the size of the effect, movement, intellectual and manual ability. To a lesser extent, the presence of seizures and speech problems were also associated with greater activity limitations and participation restrictions. As children get older, except for children GMFCS Level V, there appears to be a small, but nevertheless pervasive, decrease in their activity limitations.

Scores in the three LAQ-CP domains for Clinical and Economic Burden and Social Integration were generally not well explained by any of the available explanatory variables. This might be expected as, using the ICF model, these constructs are likely to be more strongly influenced by an array of other personal and environmental factors. In fact, these domains, as they are currently constructed, do not equate to the activities and participation of children themselves, but are nonetheless extremely important issues for the family with whom the child lives.

Clinical Burden, which is the frequency of hospital appointments and interventions, logically should have some correlation with severity of a child's medical condition, as was observed in the model by the GMFCS. One might expect that children with a greater number of impairments would experience greater clinical burden but this was not the case as the items in the scale do not appear to be sensitive to these explanatory factors in the regression. However, aside from a child's impairments, clinical burden may also be a function of the environment, such as the preferences of health professionals favouring particular treatments (Morris et al. 2002) or the overall efficiency with which local services are organised and delivered (Law et al. 2003).

Economic burden, representing whether the family had had to purchase and maintain special equipment or adapt their house because of their child's disability, was observed to increase slightly for families of children in GMFCS Level III to V compared to those in Level I. Families of children in GMFCS Levels III to V are likely to require assistive technologies or special equipment to help with their lifting and handling. The wide range of equipment used by families of children with disabilities was reported by Beresford et al. (2002). The report of that survey described how the majority families have unmet needs for equipment, and

especially those in ethnic minorities; also that many families have to purchase equipment themselves (Beresford et al. 2002). Interpreting the effect of disability on economic burden as it was measured in the Thames Valley may be misleading due to the general affluence of the area relative to the rest of the UK.

Social Integration, representing the availability of help and support together with whether parents or siblings experienced extra stress because of having a disabled child, showed some association with the presence of intellectual and movement disability but these factors explained little of the variance in the data. This suggests that the severity of a child's disability is not in itself a good marker for the family's ability to cope. More elegant investigations using structured equation modelling have examined the health and well-being of caregivers of children with cerebral palsy (Brehaut et al. 2004; Raina et al. 2005). The study by Brehaut and his colleagues indicated that caregivers of children with cerebral palsy were more likely to have physical and psychological health problems than other caregivers in Canada. Raina et al. suggest that the health and wellbeing of caregivers is strongly influenced by children's behaviour, care-giving demands and family functioning (Raina et al. 2005). Raina and his colleagues therefore emphasise the importance of supporting families and family functioning alongside delivering the "technical aspects" of health care.

The findings from this study in the Thames Valley differ from the factors affecting LAQ-CP scores reported by Hammal and colleagues from their regression analyses for families living in the North of England (Hammal et al. 2004). However, the explanatory variables used in that study were slightly different, being based on the professional assessments from the NECCPS standard assessment form (Evans et al. 1989), and also much larger sample of nearly 450 children were included. The larger sample size is more likely to have ensured

that there were enough children in the groups being compared, hence providing adequate statistical power to more reliably detect differences in the ASK and LAQ-CP scores. The smaller number of children in many groups in the Thames Valley study rendered many of these analyses underpowered other than to detect large differences which manifest in the wide confidence intervals for some of the explanatory effects. Together with those issues, the fact that the Thames Valley study was cross sectional rather than longitudinal, means that the explanatory models proposed by the present study should be used to generate hypotheses for further research.

Hammal et al. (2004) aimed to demonstrate the effect of environmental factors and therefore they created a composite index representing the districts in which families lived. The socioeconomic indices used in the Thames Valley study were of limited use in this respect because of the relatively prosperous nature of the region and the low variability of area deprivation. A major limitation in determining the effect of disability on children's activities and participation in the Thames valley study was the dearth of information on the children's or families' personal and environmental factors. In contrast to these threats to the internal validity of the Thames Valley study, the indices of children's movement and manual abilities (GMFCS and MACS) have been shown to be valid and reliable; the indices used by Hammal and colleagues were, by their own admission, not validated (Hammal et al. 2004).

The influence of personal and environmental factors on children participation in formal and informal recreation and leisure activities has been studied in Canada using the Children's Assessment of Participation and Enjoyment (CAPE) alongside other instruments (Law et al. 2005). In that study children's functional abilities, child preferences for activities and the families' orientation for activities were the significant predictors of participation intensity (Law et al. 2005). Interestingly, the child's particular diagnosis was not a significant

predictor of participation once functional limitations and age were taken into account (Law et al. 2004). Family cohesion, income, and the presence of accessible, accommodating, socially supportive and non-discriminatory environments, were indirectly associated with participation through their effect on the child and family preferences and family functioning (Law et al. 2005).

The following sections consider to what extent disability affects children's activities and participation as defined in the ICF Version for Children and Youth, taking each ICF domain in turn and with reference to evidence elicited from the survey,

9.7.1 Learning and Applying Knowledge

There is little evidence from the study to advance our understanding of whether disability affects children's activities and participation to learn and apply knowledge. Many children completed the ASK questionnaire themselves and other children helped their families in some way to complete that questionnaire. It would be reasonable to assume that the presence of any intellectual disability, rather than any physical disability, is likely to restrict children's participation in learning to read, write or do arithmetic.

9.7.2 General Tasks and Demands

This domain is very broad and is summarised in the ICF by the major levels for 'undertaking single and multiple tasks', 'carrying out daily routines', 'managing one's own behaviour' and 'handling stress and other psychological demands'. The execution of single and multiple tasks and daily routines, including organising oneself to as well to do the tasks as actually doing them, are implicit in many of the items in the ASK and LAQ-CP. The organising element of undertaking tasks is likely to be affected by any intellectual disability. However,

the types of task attempted and the daily routine will most likely be modified in the presence of any physical disability and therefore not greatly affected; perhaps only in the range of tasks undertaken.

It is notable that families of just under a fifth of the children in the survey were seeing someone about their behaviour according to an item in the LAQ-CP. Whilst there were no items specifically asking about stress on the child with cerebral palsy, there were two items in the LAQ-CP that asked about stress on the parents and any siblings. Around three-quarters of the respondents indicated that having a child with cerebral palsy had placed extra stress on the family. The responses offered to that item are 'none, slight and severe' which were criticised by one respondent, and may have led to some central tendency or 'end-of-scale aversion bias' (Streiner and Norman 2003), as families may have been disinclined to indicate experiencing 'severe' or no stress.

9.7.3 Communication

Unfortunately no data were gathered from families in the survey regarding how or whether children communicated. Children can use a range of assistive technologies to produce and receive messages and maintain conversations. Physical disabilities can be overcome to enable communication and only intellectual disability preventing the formulation of conversation might not be modified by any environmental adaptations. Interestingly, there was information on whether children had a problem with speech from *4Child* although this was not well defined and therefore may be recorded inconsistently. These data suggested having a 'speech problem' explained some of children's Physical Independence scores.

9.7.4 Mobility

Logically, the ability to sit, stand and walk, as evidenced by children's GMFCS Level, affects substantially children's outcomes in the Mobility domain of the LAQ-CP and ICF. Although Mobility involves elements of carrying and manipulating objects, children's manual ability classified using the MACS did not remain statistically significantly associated with the Mobility score in the LAQ-CP. The association is strong enough that the GMFCS could probably be used as a single summary item as a proxy for this domain although the presence of seizures also mediated Mobility scores adversely.

9.7.5 Self Care and Domestic Life

The two separate ICF domains for Self Care and Domestic Life are well covered by items in the ASK and Physical Independence scale of the LAQ-CP; the scores on these scales were substantially explained by children's movement, manual and intellectual abilities. Therefore whether children wash, dress, eat and drink themselves, or carry out their usual household tasks is almost certainly influenced by the presence of any physical or intellectual disability. Environmental modifications may make it easier for children to be more independent. For instance, very few children, even the most able in GMFCS and MACS Level I, managed buttons or buckles unaided however one family noted they had replaced all these fastenings with Velcro.

9.7.6 Interpersonal Interactions and Relationships

The survey did not elucidate this issue because of the limited coverage of these areas in the ASK and LAQ-CP. In principle, a child's disability should not affect their potential to experience interpersonal interactions and relationships with a variety of people. However, actively contributing to the development of social interactions will be affected by any

intellectual disability. Children with purely physical disabilities may need to adapt the way they give and receive 'physical contact' and 'social cues' but should not be impeded from developing relationships. Most children's predominant relationships will be amongst their family members. The survey showed that three-quarters of children were residing with both of their birth parents and all but a couple with at least one birth parent, which is unlikely to be dissimilar to the general population. However, a third of the children in this study had not spent any time with friends outside of school and family environments probably indicating low participation in 'informal social relationships' relative to the general population. As several respondents to this survey made clear, even children with the most profound disabilities are no less loved by their families. These children may be, in some cases, more passive participants in their relationships with family, friends and school staff, but they are still participating in those life situations.

9.7.7 Major Life Areas – education and economic

The two major life areas included in the ICF Version for Children and Youth are education and economic life situations. Education involves informal work and play at home and formal education at school. All the children in the survey were participating in formal education at school. The type of schooling depended largely on their intellectual ability rather than any physical disability, although there were no children in GMFCS Level V attending mainstream school and one child in GMFCS Level II was attending a school for blind children. Children in all levels of the GMFCS attended either mainstream schools with special support units or special schools. Therefore physical disability does not in itself determine educational participation although it may modify how that education is delivered. Information about children's participation in economically related life situations was not gathered in the survey but details of any economic burden on the family were measured in

the LAQ-CP. Inspection of these data showed that very little of the variance in Economic Burden score was explained by the severity of a child's physical or intellectual disability.

9.7.8 Community, Social and Civic Life

This domain of the ICF includes community and school life, recreation and leisure, and religion and spirituality. The ASK includes questions about whether children played sport with competitive clubs which few of the children in the survey did, however, for one child this was because the disabled athletics club she wanted to join was a long distance away. The ICF recreational level also includes clubs for arts, culture, crafts and hobbies and these were not assessed in the survey instruments. Having an intellectual disability probably does affect children's ability to participate in many aspects community, social and civic life but physical disability should not be an impediment to participation. That is providing there is an appropriate range of activities available and, if necessary, appropriate assistive aids.

9.7.9 Summary of effect of disability on activities and participation

Undoubtedly, disability does affect children's activities and participation but not necessarily to prevent them from participating at all, and some investigators have therefore been moved to examine 'participation intensity' (Law et al. 2005). From an equity perspective, it is desirable to know whether the extent of children's participation in activities is consistent with their potential, given the predicament imposed by their disability. Importantly, and consistent with the ICF model, determining whether the provision of any environmental aids or the removal of any barriers combined with consideration of any personal preferences can enhance children's experience of life situations.

Despite the broad issues covered within the ASK and LAQ-CP instruments, some elements of the domains in activities and participation as defined in the ICF were not assessed in the Thames Valley study. Items such as 'learning and remembering' and 'thinking and solving problems' from the HSCS-PS (Saigal et al. 2005) could be scaled appropriately for school age children and potentially used as items in future studies to assess these aspects of children's activities and participation. Issues relating to recreation and leisure activities can also be assessed more formally using the Children's Assessment of Participation and Enjoyment (CAPE) (King et al. 2004). The LIFE-H (long form) has an extensive section for communication which can be used to assess children's performance in a range of verbal, signing and written activities (Fougeyrollas et al. 2004). The LIFE-H also assesses Interpersonal Relationships in terms of 'loving relationships' with parents, siblings and other relations, and also 'creating or maintaining friendships', and Education and Financial Responsibilities (Fougeyrollas et al. 2004). The reliability of family assessment for these items would however have to be established as the LIFE-H is currently relatively long and complicated and solely used as a professionally or interviewer assessed instrument (L. Noreau, personal communication). Children educational participation might specifically be assessed separately using the School Function Assessment (Coster et al. 1998) although the inappropriateness of certain sections of this instrument for some children with cerebral palsy has been highlighted (Wright et al. 2005).

9.8 CONCURRENT RELATED RESEARCH

The Thames Valley study has several limitations that have been taken into account in other concurrent research studies. For instance no specific information on any environmental factors affecting children's participation was gathered. However, the Study of Participation of Children with Cerebral Palsy Living in Europe is a comprehensive and multinational study aiming to provide information about optimal environments for children with cerebral palsy (SPARCLE 2005). A systematic review of relevant environmental factors was conducted in the preliminary phase of that study (Mihyalov et al. 2004), and both personal and environmental factors are being assessed with families using structured face-to-face interviews. Participation is being measured using the LIFE-H, and quality of life using the Child Health Questionnaire and KIDSCREEN, a recently developed generic instrument suitable for use across European countries (Ravens-Sieberer et al 2001). Information on children's impairments will be available from the cerebral palsy databases and supplemented with information from interviews including the GMFCS. The SPARCLE study has, like the Thames Valley study, utilised the conceptual framework advocated by the ICF and the results are expected to be disseminated late in 2005 (A. Colver, personal communication).

Investigators at *CanChild* have recruited around 300 children who took part in the Ontario Motor Growth study and will follow them through their teenage years in the Adolescent Study of Quality of life, Mobility and Exercise (ASQME) Study. The ASK instrument is one of the instruments being used and GMFM will be used to map any changes in young people's motor function within their GMFCS Level. This longitudinal study is expected to run until 2007 (P. Rosenbaum, personal communication) and will provide much needed information on what happens to children with cerebral palsy as they grow up into adulthood.

Similar longitudinal research is being undertaken with cohorts of children with cerebral palsy in Holland in the PERRIN study (J.W. Gorter, personal correspondence).

These studies indicate the increasing importance of the activities and participation of people with chronic disabilities in line with the ICF model. The research agenda is certainly shifting towards this aim and clinical practice will surely follow in time provided relevant policy makers make resources available.

9.9 CHAPTER SUMMARY

This chapter has discussed the findings of the Thames Valley study and described to what extent the aims and objectives set out in *Chapter 3* were achieved. The internal validity of the study was generally high as the methodology included rigorously checking the reliability of family-assessed instruments and the goodness of fit of the regression models. However, the small number of children in the comparative groups used for the regression analyses is a major limitation to the statistical power to reliably detect differences in this study. The lack of any measures of personal and environmental factors is also a weakness when analysing factors affecting activities and participation using the model ICF. The high reliability of family assessment for classifying children's movement and manual abilities using the GMFCS and MACS means there is evidence to strongly recommend using these family-assessed instruments for both research and in routine clinical practice. The sample of children and families that took part in the study was shown to be representative of the characteristics of the complete geographically population from which they were recruited. However, the external validity of the results should be interpreted with the caveat that the Thames Valley is a relatively prosperous region and caution should therefore be taken when generalising these findings to areas with different deprivation profiles.

Whilst the aims and objectives were on the whole achieved successfully, the findings of the study with respect to describing children's activities and participation as defined in the ICF were limited by the instruments used in the survey. Activities and participation of children with cerebral palsy are affected across the range of the domains specified in the ICF, however only part of this restriction appears to be directly associated with physical movement or manual disabilities, and perhaps rather more consistently by intellectual disability. This is because it is usually more straightforward to provide environmental adaptations to enable children with physical disabilities to participate than it is to overcome limitations due to intellectual disability.

The extent to which the research questions in this study were addressed and answered is described in the conclusions and recommendations in the following and final chapter.

CHAPTER 10

CONCLUSIONS AND RECOMMENDATIONS

10.1 FAMILY ASSESSMENT OF CHILDREN'S ABILITIES

The research described in this thesis used family assessment to classify children's movement and manual abilities using the GMFCS and MACS, which are classification systems shown to be valid and reliable when assessed by health professionals. One of the key research questions in this study was to determine whether families can provide reliable assessments for their children compared to health professionals. For the GMFCS, two surveys with families were conducted: one in Canada, and one in the Thames Valley in the UK. For the Canadian study classifications had been made by health professionals as part of the Ontario Motor Growth study and therefore only the families were surveyed by mail. In the UK both families and the health professionals they nominated were surveyed by mail. In both studies the reliability between families and health professionals was excellent (ICC = 0.9); test-retest reliability in the UK was also excellent (ICC > 0.9). There was not always perfect agreement between families and health professionals, or between health professionals, but disagreements of more than one GMFCS Level were rare.

This was the second study to examine the reliability of family assessment for the MACS, as the developers have previously reported good reliability for 25 children in their development of the system (Eliasson et al. In press). The analyses described in this thesis showed the reliability of family assessment to be very high and equivalent to the reliability measured between health professionals (ICC = 0.9). Similar to findings with the GMFCS, there is not always absolute agreement about a child's MACS Level but disagreement by more than one level is rare. In summary, the answer to the research question asking whether families can provide reliable assessments for their children compared to health professionals using the GMFCS and MACS is almost certainly yes, and probably with greater knowledge of how children perform across a wider range of environmental settings.

10.2 EFFECT OF DISABILITY ON CHILDREN AND FAMILIES

The research described in this thesis sought to elucidate our understanding of how cerebral palsy affects the component of health called 'activities and participation' in the ICF Version for Children and Youth (WHO 2004). Activities and participation were affirmed to be an objective construct that can be measured either by self-report or by someone who knows a child well. This distinguishes activities and participation conceptually from quality of life which is a subjective phenomenon that ideally captures the views of respondents. The ASK and LAQ-CP were identified as the best available indices of activities and participation that could be administered in a postal survey in a structured review of family-assessed instruments (Morris et al. 2005). These instruments were subsequently shown to have performed in the Thames Valley survey with good internal consistency and high construct validity, although there was limited face validity to specific scales used with a few items. Families' classifications of their children's movement and manual abilities (GMFCS and MACS), together with details of the children's cerebral palsy and other associated impairments recorded on the *4Child* database, as explanatory factors to explore their impact on children's activities and participation.

The ASK discriminated between children with all levels of movement ability from scores of zero for several children in GMFCS Level V, indicating they were completely dependent, to scores within the expected range for unimpaired children. In terms of its acceptability as an family-assessed instrument, the ASK was inappropriate for use with children in GMFCS Level V. However, the ASK is not only designed to discriminate between children with different physical functioning, as it did in this study, but also to detect changes in individual functioning over time. This aspect of instrument performance could not be assessed in the present cross-sectional study, however, the responsiveness of the ASK score to clinically

important changes for children within GMFCS Levels I to IV warrants further evaluation. In the multivariate analysis, decreases in ASK score, representing adverse effects on physical functioning, were well explained by movement, intellectual and manual disabilities; seizures also had an adverse effect but, excluding children in GMFCS Level V, there was a small but pervasive increase in ASK score, indicating decreasing dependence as children got older.

The LAQ-CP is a multidimensional instrument and the domains for Physical Independence and Mobility showed strong convergence with ASK scores ($r \geq$-0.9). Scores in these domains were all well explained by movement, intellectual and manual disabilities, with the presence of seizures and speech problems also producing an adverse effect. Interestingly, there was a trend for an overall decrease in children's dependence for older children. The Clinical and Economic Burden and Social Integration scales were not well explained by the severity of disability or any of the explanatory factors available for these analyses. This may in part be due to the scales used for some items, but it is conceptually consistent with the ICF model that personal and environmental factors probably play more of a role in mediating these outcomes. The overall effect on family life of having a child with any level of disability should therefore not be underestimated. Minimisation of these effects of disability on the family should be integrated into health and social care, and principally by ensuring services are family-centred. All children in the Thames Valley study were found to be participating in education to some degree and the type of school children attended was largely dependent on their intellectual ability. The LAQ-CP summary score is heavily weighted towards the domain scores for Physical Independence and Mobility. The LAQ-CP summary score was strongly associated with movement and manual disability and, as these factors explained almost all of the variance in the data, the LAQ-CP summary score appears to represent a reasonable measure of the severity of children's disability.

10.3 RECOMMENDATIONS FOR FURTHER RESEARCH

10.3.1 Developing a family-assessed instrument to measure activities and participation

A family-assessed instrument is required to measure children's activities and participation inexpensively in postal surveys, utilising the families' knowledge of how a child performs across a broad range of the environments, and at least should meet the following criteria. The instrument should be generic so that it would be useful for children with or without any kind of disability, particularly as the issues are not apparently dictated by diagnostic categories (Law et al. 2004). The instrument should be as brief as possible whilst also comprehensively covering the relevant domains. The validity and reliability of the instrument would have to be demonstrated.

10.3.2 Researchers engaging more with families

One of the disappointing aspects of the Thames Valley study was the moderate response from families to the initial invitation to take part in the survey. The contention in this thesis was that families may not have recognised the organisation from which the survey or perceived the aims of the study to be relevant to them and their priorities. Engaging more with families in the future would foster feelings of collaboration between researchers and families. Asking families what research they would like to see done might be a good first step towards this goal and is therefore a strong recommendation from this thesis. The question could readily be posed to families when the findings of the Thames Valley study are presented in the final newsletter, subject to approval from the *4Child* Advisory Group. The *4Child* Advisory Group might also consider recommending running formal meetings with families, as has been done with some considerable success by the North of England Cerebral Palsy Survey, to disseminate information derived from maintaining databases.

10.3.3 Responsiveness of the ASK for children with cerebral palsy

Despite being recommended as one of four family-assessed instruments for use in paediatric orthopaedics (Furlong et al. 2005), the ASK has yet to be cited in any evaluative studies of interventions for children with cerebral palsy. This may be due the lag between publication of instruments, their uptake, and use and subsequent publication of those studies. However evidence of responsiveness of the ASK was not assessed separately for children with cerebral palsy. The responsiveness of the ASK should therefore be assessed for children with cerebral palsy within each of the GMFCS Levels I to IV. This might demonstrate the utility of the ASK to clinicians and researchers who as yet, so far as can be determined from published material, been reluctant to use the instrument with children with cerebral palsy.

10.3.4 Gather and report family-assessed ratings for the GMFCS and MACS

The GMFCS has become the standard way to classify and describe the movement abilities of children with cerebral palsy, and the MACS may, in time, become similarly established for classifying manual ability. However, to date, researchers have always presented professional classifications for the GMFCS and this may not always agree absolutely with families as the two studies described in this thesis have shown. Therefore, for clarity, researchers should in future specifically state which method was used to classify children's abilities using either the GMFCS or MACS. Researchers should also consider gathering and using family assessments for these variables for their analyses.

10.4 RECOMMENDATIONS FOR CLINICIANS

Clinicians should consider gathering a classification from a family of their child's abilities using the GMFCS and MACS prior to, or during, consultations. The results might provide a

basis for informing families about prognosis, facilitate family-centred care, and help physical management regimens to be planned appropriate to the child's GMFCS and MACS Levels. Clinicians should be aware that their assessment of children's abilities may be strongly influenced by the environments in which they typically observe children functioning. Children's performance of activities at school or in clinical settings may not represent how they perform at home or outside in the community due to personal and environmental factors. This should further encourage clinicians such as paediatricians and therapists to gather family assessments of children's abilities using the GMFCS and MACS.

Clinicians must continue to be made aware of the ICF and the importance of enhancing children's activities and participation as well as the typical medical outcomes that focus on impairments of body structures such as spasticity, range of motion, and muscle contractures. Clinicians should then seek to evaluate the effects of their interventions and management regimens on children's activities and participation.

10.5 RECOMMENDATIONS FOR COMMISSIONERS AND POLICY MAKERS

Those responsible for commissioning health and social services at all levels of national and local government, and the strategic health authorities, must reminded of the predicament and specific difficulties encountered by families of children with cerebral palsy. Children's activities and participation are affected by their disabilities but, consistent with the ICF model, are also mediated by personal and environmental factors. With the emphasis on enhancing children's participation becoming increasingly recognised as a priority, a review of access to occupational therapy services is warranted considering how few children in this survey indicated any contact with an occupational therapist.

Public policies that support the wellbeing of families and parents, the child's immediate environment, such as providing appropriate equipment and respite services when necessary, can directly improve children's health as defined in the ICF. Conversely, withdrawing or limiting access to these and similar services that families feel are important may have a consequent deleterious effect on the health of children and their families.

10.6 CONCLUSIONS SUMMARY

The research described in this thesis has presented considerable evidence to support the reliability of family report for classifying children's movement and manual abilities using the GMFCS and MACS. Family-assessed instruments appropriate for measuring activities and participation for children with cerebral palsy were identified in a structured review. A subsequent survey conducted with families of children with cerebral palsy concurred with other studies that have used professional assessment by indicating that movement, manual and intellectual ability are the strongest predictors of children's participation. However, current family-assessed instruments are not well refined for measuring children's activities and participation as defined in the ICF Version for Children and Youth. The development of a brief generic instrument that can be used to assess these important components of health for all children should be undertaken in future research.

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APPENDIX A

SURVEY CORRESPONDENCE,

QUESTIONNAIRES

& NEWSLETTERS



npeu
National Perinatal Epidemiology Unit

ORECI
Oxford Register of Early Childhood Impairments

23 September 2005

Dr «GPInitials» «GPSurname»
«GPAdd1»
«GPAdd2»
«GPAdd3»
«GPAddPC»

Dear Dr «GPSurname»

Re: «ReturnedForename» «ReturnedSurname» DoB: «DOB»
«ReturnedAddressLine1», «ReturnedAddressLine2» «ReturnedPostcode»

We are undertaking a postal survey of the families of children with cerebral palsy who were born to mothers resident in the Thames Valley between 1991 to 1997. The aim of the study is to measure the disability of children with cerebral palsy in a complete population. We will send the family an invitation letter, an information sheet about the survey (copies enclosed) and the questionnaires.

Your patient named above is recorded on the Oxford Register of Early Childhood Impairment as having a diagnosis of cerebral palsy and born to a mother who was resident in Oxfordshire or West Berkshire. We would therefore like to include them in our survey.

If there is any reason why we should not contact this family, please let us know or return this whole letter before **22 October 2004**.

Yours sincerely

Chris Morris

Encl. Family invitation letter and information sheet.

I am the family GP for the above named child and consider that it would <u>not</u> be appropriate to contact them to ask if they want to take part in this survey

GP name _____

Signature _____

Reason (optional) _____

Tel: 01865 226706
Fax: 01865 227002
christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Old Road Campus, University of Oxford, OX3 7LF



npeu
National Perinatal
Epidemiology Unit

21 June 2004

Parent or carer of «ReturnedForename» «ReturnedSurname»
«ReturnedAddressLine1»
«ReturnedAddressLine2»
«ReturnedAddressLine3»
«ReturnedPostcode»

ORECI
Oxford Register of Early Childhood Impairments

Dear «ReturnedForename»'s parent or carer

Measuring Children's Abilities

We are a team of 'child disability' researchers at the University of Oxford working with colleagues in Canada. We invite you to help us measure the abilities of a population of children with impairments that affect movement. We are pleased to provide you with the enclosed information about our survey. Do take some time to read and think about the information we have provided or discuss it with your family or friends. If you have any questions please contact the lead researcher using the details at the bottom of the page.

What is the study about?
Movement problems vary from barely noticeable to extremely severe. Difficulty in controlling movement can affect our ability to accomplish tasks and take part in life situations. We want to measure what activities children are able to do and the impact that any disability has on your family. Please read more about why this information is important in the blue information sheet.

What would I have to do?
We are asking you, and «ReturnedForename» if it is appropriate and she wants to, to take part in a postal survey. The questionnaires are in the separate packet enclosed so you can see more about the sort of questions we are asking.

Why have I been chosen?
We have invited families of children with movement difficulties born to mothers who were resident in Oxfordshire, Berkshire or Buckinghamshire between 1991 and 1997.

Do I have to take part?
You do not have to take part; however we would be grateful if you would read the enclosed information before deciding. If you would like to take part please complete the questionnaires and return them to us in the stamped addressed envelope provided.

Kind regards

Chris Morris
Lead Researcher

Tel: 01865 226706
Fax: 01865 227002
christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Institute of Health Sciences, University of Oxford, OX3 7LF

B

National Perinatal Epidemiology Unit

Oxford Register of Early Childhood Impairments



Measuring Children's Abilities



Information Sheet

What will I be asked if I take part?

We would like to send you a questionnaire asking about your child's abilities. There are three parts: the first is a single question about the general physical ability of your child with five possible answers. The second part is a questionnaire designed for children to answer themselves if they wish to and are able (although parents or carers who know the child well can also do it), it asks whether your child thinks they could do certain activities. The final part is a way to measure the impact of these abilities on your family. We are also asking your permission to approach your child's health professionals (paediatrician and physiotherapist) about your child's abilities and to examine your child's medical records.

Why is this information important?

One of the difficult things about impairments of movement is that the effects vary so much from one person to another. Some people seem hardly affected, some might walk on their toes, and others are unable to sit up without support. This wide range makes it difficult for doctors to predict how a young child will be affected later in life and is also one the obstacles when clinicians want to test treatments to overcome disability.

To overcome these problems scientists in Canada have designed a new system for measuring children's abilities, but this currently requires a health professional to make an assessment. We think that parents or other family carers could reliably provide this information just as well. In order to see if this is true we are carrying out this research by collecting information from both families and health professionals.

We want to measure the ability of our population of children born to mothers who were resident in Oxfordshire and West Berkshire between 1991 and 1997. This will help us to ensure that those who make decisions about health and social services are aware of the needs of the population and to enable us to measure if new treatments and services really do make a difference to children and families. It is very important that politicians and health service managers have accurate information in order to decide how to respond to public health needs.

Please turn over to find out more

What will happen to the results?

We will report our results to the Department of Health and the Thames Valley Health Authority and make recommendations for health service policy based on the findings. A report of the findings will be made available to everyone who takes part in the research and disability support groups such as the UK charity Scope. We will also publish the results to our colleagues in professional journals and meetings to encourage further research and improve the care of children in the future.

How will information be kept confidential?

Any information you give us will be treated in confidence and nobody would be identified in any reports or publications. There is an identifying number on each form, which is unique to your child in this study. This is how we will be able to compare family and professional reports for each child and will not be disclosed outside the research team at any stage.

Who has reviewed this study?

This study has been developed through the collaboration of some of the leading world experts in child disability research at the Universities of Oxford and Newcastle in the UK and the *CanChild* Centre for Childhood Disability Research in Canada. The documents provided to you have been reviewed by the UK charity Scope and the Oxfordshire Applied and Qualitative Research Ethics Committee (AQREC) on behalf of the Thames Valley Health Authority.

Who can I contact for further information?

If you would like further information about this study or discuss any issues it may raise then please contact the lead researcher using the contact details below.

Chris Morris
National Perinatal Epidemiology Unit
Institute of Health Sciences
University of Oxford
OX3 7LF

Tel: 01865 226706
Fax: 01865 227002
christopher.morris@perinat.ox.ac.uk

www.npeu.ox.ac.uk
www.fhs.mcmaster.ca/canchild

What do I do now?

If you would like to take part please go to the yellow consent form and tick the appropriate boxes and return it to us in the stamped addressed envelope provided.

National Perinatal Epidemiology Unit

Oxford Register of Early Childhood Impairments

Measuring Children's Abilities





Information Sheet
FOR CHILDREN
Aged 6-8 years

What is this about?

We would like to find out how often you do some common activities. We want to measure this so we can help children with disabilities to be able to do more in the future.



What would I have to do?



We have sent your family some questions by post. There is one section that you could help to do, perhaps if someone explains the questions to you, or if you prefer just let the grown-up do it.

Why have I been chosen?

We have invited families of children who have problems with moving and who were born in the Thames Valley area from 1991 to 1997.



Want to ask any questions?

Why not email us at: christopher.morris@perinat.ox.ac.uk

or ask a grown up if you can phone us at the number below.

Tel: 01865 226706
Fax: 01865 226706
christopher.morris@perinat.ox.ac.uk

AQREC A02.073.childinfo June 2004

Chris Morris, NPEU, Institute of Health Sciences, University of Oxford, OX3 7LF

E

National Perinatal Epidemiology Unit

Oxford Register of Early Childhood Impairments

Measuring Children's Abilities





Information Sheet
FOR CHILDREN
Aged 8-12 years

What is this about?



We would like to find out how often you do some common activities. We want to measure this so we can help children with disabilities to be able to do more in the future.

What would I have to do?



We would send your family some questions by post. There is one section that you could do yourself or if you don't want to you could help or just let the grown-up do it. If you would prefer to do it yourself just ask your parent to show you where to sign the form.

Why have I been chosen?

We have invited families of children who have problems with moving who were born in Oxfordshire or West Berkshire from 1991 to 1997.



Want to ask any questions?

Why not email us at: christopher.morris@perinat.ox.ac.uk or ask a grown up if you can phone us at the number below.

Tel: 01865 226706
Fax: 01865 226706
christopher.morris@perinat.ox.ac.uk

AQREC A02.073.childinfo8/12.v2

Chris Morris, NPEU, Institute of Health Sciences, University of Oxford, OX3 7LF

Id No.

National Perinatal Epidemiology Unit

Oxford Register of Early Childhood Impairments

Measuring Children's Abilities

Consent form

If you would like to take part please tick the appropriate boxes and return this form to us in the stamped addressed envelope provided.

☐ I confirm that I have read and understand the information provided

☐ **YES** I would like to participate in this research

If you tick this box and sign below we will send you the survey questionnaires

☐ **YES** I agree to my child's health professionals being approached and my child's medical records being examined about their physical ability.

Your Name _____

Your signature _____

(required if you want to take part)

Your child's name «ReturnedForename» «ReturnedSurname»

Your child's signature _____

(only required if your child wants to help)

☐ **MAYBE** but I would like more information before I decide

If you tick this box then either provide your phone number or contact us using the details below, we could call you back so you are charged little for the call. Please ring me on tel no: _____

☐ **NO** I do not want to participate in this research

If you tick this box no signature is necessary and we will not contact you further about this study

PLEASE RETURN THIS FORM IN THE ENVELOPE PROVIDED

Thank you very much for your time

Tel: 01865 226706
Fax: 01865 226706
christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Institute of Health Sciences, University of Oxford, OX3 7LF

G



npeu
National Perinatal Epidemiology Unit

ORECI
Oxford Register of Early Childhood Impairments

Parent or carer of «ReturnedForename» «ReturnedSurname»
«ReturnedAddressLine1»
«ReturnedAddressLine2»
«ReturnedAddressLine3»
«ReturnedPostcode»

Dear «ReturnedForename»'s parent or carer

We hope that you will want to take part in this postal survey and are pleased to provide you with the questionnaires, there are four separate parts:

The single yellow sheet **General Physical Ability** requires you to mark the one of the five descriptions that most closely represents «ReturnedForename»'s movement abilities.

There is a sheet for you to provide the names of some of «ReturnedForename»'s **health professionals** and where they are (such as the name of the hospital or school), if you consented for us to contact them.

The **Activities Scale for Kids** was designed for children to answer themselves, although it may be best to read it together initially to ensure they understand the instructions. However if «ReturnedForename» is unable or does not wish to take part we would be grateful if you could answer these questions on her behalf.

The **Lifestyle Assessment Questionnaire** should be filled in by the parent or carer who is most involved with «ReturnedForename».

We hope all the instructions are clear however if you would like to ask anything about the questionnaires please contact me using details at the bottom of the page.

Once you have completed the questionnaires please put them in the stamped addressed envelope provided and send it back to us. We will send you a report of the findings once we have analysed all the results.

Kind regards

Chris Morris
Lead Researcher

Tel: 01865 226706
Fax: 01865 227002
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Chris Morris, NPEU, Old Road Campus, University of Oxford, OX3 7LF

H



General Physical Ability Questionnaire

Id No.
«ORECINO»

Please read the following and mark **only one box** beside the description that best represents your child's movement abilities.

My child, **«ReturnedForename»**...

☐ **Has difficulty sitting on their own and controlling their head and body posture in most positions**
and has difficulty achieving any voluntary control of movement
and needs specially supportive chair to sit comfortably
and has to be lifted or hoisted by another person to move.

☐ **Can sit on their own but does not stand or walk without significant support**
and therefore relies mostly on wheelchair at home, school and in the community
and often needs extra body / trunk support to improve arm and hand function
and may achieve self-mobility using a powered wheelchair.

☐ **Can stand on their own and only walks using a walking aid** (such as a Kaye walker, rollator, or any kind of crutches, walking sticks or canes etc)
and finds it difficult to climb stairs, or walk on uneven surfaces
and may use a wheelchair when travelling for long distances or in crowds.

☐ **Can walk on their own without using walking aids, but needs to hold the handrail when going up or down stairs**
and often finds it difficult to walk on uneven surfaces, slopes or in crowds.

☐ **Can walk on their own without using walking aids, and can go up or down stairs without needing to hold the handrail**
and walks wherever they want to go (including uneven surfaces, slopes or in crowds)
and can run and jump although their speed, balance, and coordination may be slightly limited

---

Please also indicate whether any of your health professionals has ever discussed the *Gross Motor Function Classification System* with your family?

YES ☐ NO ☐

THANK YOU

Developed in conjunction with
CanChild Centre for Childhood Disability Research

I

National Perinatal Epidemiology Unit

Oxford Register of Early Childhood Impairments

ID No
«ORECINO»

Measuring Children's Abilities

Please list the names and contact details for some of «ReturnedForename»'s health professionals, for example any physiotherapists, paediatrician, orthopaedic surgeon etc. We only need the name of the professionals and the name of their hospital or base (for example the name of school).

1

2

3

4

Tel: 01865 226706
Fax: 01865 227002
christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Institute of Health Sciences, University of Oxford, OX3 7LF

ID No. _____

Activities Scale for Kids

PERFORMANCE VERSION

38 ITEMS

(ASKp 38)

NOV. 2002

© Nancy L. Young
The Hospital for Sick Children

INSTRUCTIONS

This is a new booklet that asks you how often you did activities on your own last week. For each question think about what you really did last week. Try to remember the things you did at home, at school, and when you were with your friends. Record your answer by putting an X in the most correct box. If there isn't a good answer for you, write your own answer beside the question.

It is important that you answer all questions.

What do the answers mean?

	Answers	*Meaning*
☐	all of the time	I did it every time I needed to
☐	most of the time	I did it almost all of the time I needed to, but once in a while I didn't do it
☐	sometimes	I did it about half of the time I needed to, but about half of the time I didn't do it
☐	once in a while	I did it at least once last week when I needed to, but most of the time didn't do it
☐	none of the time	I didn't do it at all when I needed to

Example 1

Terry's hands were very sore 3 days last week, so his mum helped him get dressed. He dressed himself without help the other 4 days.
One of Terry's answers looked like this . . .
I fastened my clothes by myself (fastened means doing up buttons & zips)

- ☐ all of the time
- ☐ most of the time
- ☒ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not need to because none of my clothes have fasteners

Example 2

Sally's health was very good last week. She wanted a snack on 5 nights.
She got a snack by herself on 4 nights.
One of Sally's answers looked like this . . .
I made a snack by myself . . . (or prepared breakfast or lunch)

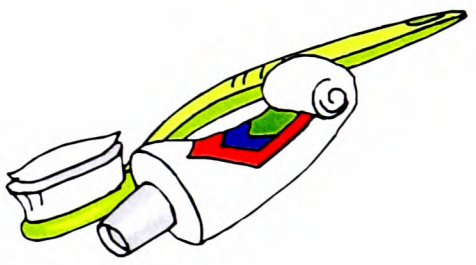
- ☐ all of the time
- ☒ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I am not allowed to have snacks or make my own meals

Note: *If you never had a chance to do an activity last week, do not pick a box. Write your reason beside the question. For example: maybe you never came across a wet pavement, or maybe someone drove you to school, just to be safe.*

1. I put toothpaste on my toothbrush then brushed my teeth by myself . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



2 I used the toilet at home by myself . . . (includes getting on and off the toilet)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



3 I washed my whole body by myself . . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



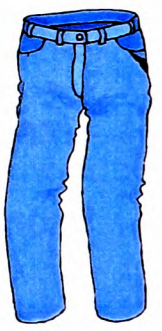
4 I put my shirt on by myself . . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



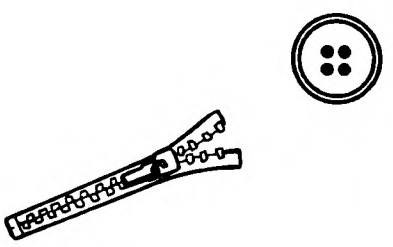
5 I put my trousers on by myself . . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



6 I fastened my clothes by myself. . . (fastened means doing up buttons and zips)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I did not need to, none of my clothes have fasteners

7 I put my shoes on and did them up by myself .

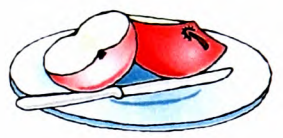


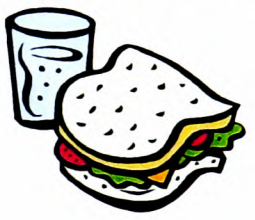
- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not need to wear shoes last week.

Please explain:

8 I made a snack by myself (or prepared breakfast or lunch) . . .





- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I was not allowed to snack or make my own meals

9 I did my usual job or chores . . . (Examples: doing the dishes, cleaning my room, babysitting)



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I don't have any jobs or chores

10 I took care of my medical needs . . . (Examples: put on splints or took medication)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I did not have special medical needs last week

11 I did my handwriting by myself . . . (Examples: to do my school work)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



12 I did the same sports that I usually enjoy by myself . . . (Examples: cycling, swimming, gymnastics, dance)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



13 I walked without any support . . . (No crutches or walking sticks)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



To get around INSIDE I usually used . . .



- ☐ no special support
- ☐ an artificial leg(s)
- ☐ one walking stick
- ☐ two walking sticks
- ☐ crutches
- ☐ a walker
- ☐ a wheelchair
- ☐ a scooter
- ☐ brace, splint or orthosis
- ☐ my hands and knees



(please choose as many of these as you use)

other, please describe

14 I got around inside my home without anyone to help me. . (Example: walked or rolled to the bathroom, to the dinner table, or to the kitchen for a drink)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



15 I walked (or rolled) in crowded areas . . . (Example: school hallways between classes or at a busy shopping centre)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I tried to stay away from crowds



To get around OUTSIDE I usually used . . .
(please choose as many of these as you use)



- ☐ no special support
- ☐ an artificial leg(s)
- ☐ one walking stick
- ☐ two walking sticks
- ☐ crutches
- ☐ a walker
- ☐ a wheelchair
- ☐ a scooter
- ☐ brace, splint or orthosis
- ☐ my hands and knees



other, please describe

Answer the questions on this page and the next page by telling how you did with the support you just marked on the list.

Example: If you said "I usually used crutches", then answer the questions that follow by telling how you did with crutches last week.

16 I got around outside without anyone to help me . . . (Example: walked or rolled to a friend's house, to school, or to the park)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I never had the chance to go outside last week.

Explain:

17 I walked (or rolled) up a gentle hill or slope by myself. . .



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I always tried to stay away from hills

18 I walked (or rolled) on rough or slippery surfaces . . . (Examples: gravel driveways, or wet pavements)



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not need to, I did not come across a rough or slippery surface

19 When I ran (or rolled) around outside, I kept up with my friends...



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not have a chance to be outside with friends

Explain:

WOW!

YOU ARE DOING VERY WELL.

TAKE A SHORT REST IF YOU LIKE.

BUT PLEASE TRY TO FINISH THIS TODAY.

How much help have you had so far?

- ☐ I have done the questions all by myself.
- ☐ Someone has read the questions to me.
- ☐ Someone has helped me with some of the answers.
- ☐ Someone has helped me with most of the answers.
- ☐ other



Please describe:

Reminder: *If you never had a chance to do an activity last week,* ***do not*** *pick a box. Write your reason beside the question. For example: maybe you never came across a wet sidewalk, or maybe someone drove you to school, just to be safe.*

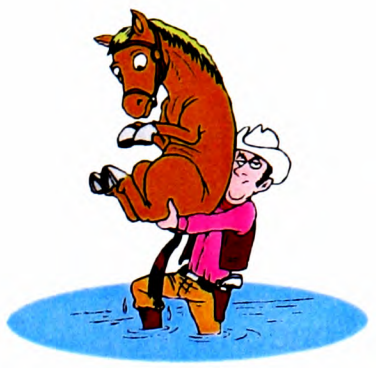
20 I carried a drink or food to the table by myself without spilling . . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I am not allowed to do this

21 I carried things in 2 hands by myself . . . (Example: big or heavy things like stuffed animals, family pet)



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I had no reason to carry anything in 2 hands last week

22 I stood still for 10 minutes without resting . . . (Examples: waiting in a queue at a shop or bus stop)



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

23 I stretched to reach a high shelf (or to see over the person in front of me) . . .



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not need to stretch for anything

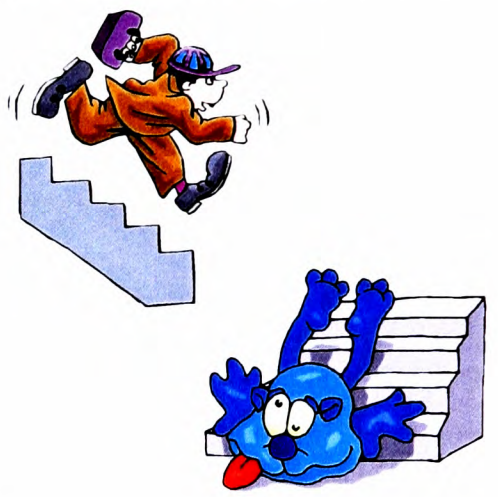
24 I got through heavy doors by myself . . . (Example: the front door at home or at school)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



25 I walked up and down a flight of stairs (even when other people were using them) . . . (Note: one flight is about 14 stairs)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I did not need to, because I did not come across a flight of stairs

26 I got in and out of a car by myself . . . (opened the door, got in, closed the door, and got out again)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I didn't need to go anywhere by car last week

27 I got in and out of a chair (or wheelchair) by myself . . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



28 I sat on the floor . . . (Examples: at a school assembly or watching TV)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I did not have any reason to sit on the floor

29 I got in and out of my bed by myself . . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



30 I got down onto the ground from standing, and got back up again by myself . . .

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I did not need to get onto the floor or ground.

Explain:

31 I did climbing activities . . . (Examples: climbing trees, rocks, or climbing over a fence)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I did not do climbing activities.

32 I played team sports with others in my class . . (Examples: football, hockey, netball or basketball)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time





Or ◯ I did not play team sports in class.

33 I played some sports by myself or with a few friends . . . (Examples: kicking or throwing a ball around)



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not play sports.



34 I played sport with local competitive clubs . . . (Examples: with a local football, hockey, netball or basketball team)



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not play sports with local competitive clubs.

35 I kept my balance while playing rough games . . (Examples: tag, wrestling, karate, judo)



- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time

Or ◯ I did not play rough games.

36 I did activities I usually enjoy for a long time without getting tired out (Examples: swimming, jogging, tennis, badminton, rowing)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time





Or ◯ I did not do these activities .

37. I ran in a race . . . (Examples: 50 metre dash)

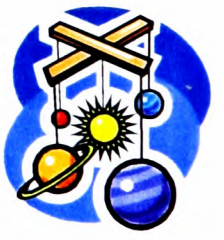
- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time



Or ◯ I did not run in races.

38 I worked carefully with my hands . . . (Examples: building with Lego, making models, sewing, making bead necklaces)

- ☐ all of the time
- ☐ most of the time
- ☐ sometimes
- ☐ once in a while
- ☐ none of the time





Or ◯ I did not work with my hands.

YOU ARE ALL DONE
THANK YOU!

Please feel free to write any comments or suggestions you or your family would like to share with us in the space below.

ID No. _____

Lifestyle Assessment Questionnaire

About this questionnaire and how you can help us:

We are trying to get a better understanding of families who are caring for a child with movement difficulties. You could help us by letting us know your views and completing this questionnaire. These questions are all about the problems in everyday life which you and your child experience.





If you complete this questionnaire, your answers will be treated in total confidence. Although we will use the information you give us, neither you, your child or your family will be identified in any way.

How will we use the information you give us?

Information from you and other parents will help us decide whether we can provide services differently in a way that is better for children and families.

Who should fill in the questionnaire?

The questionnaire should be filled in by the parent or carer who is most involved with the day to day care of your child. Please answer the questions realistically based on what you know your child can do and does every day and not on what you think he or she should be able to do.

We have tried to make filling this questionnaire as simple as possible. For most questions, all you will do is tick the appropriate box from the options given. It will tell you in the question what to do.

There are also spaces for your comments, and we are very interested in what you have to say.

Mackie, Jessen & Jarvis
LAQ - CP 1998

1. How many times has your child been seen **over the last year** by a doctor other than the school doctor or your family doctor? *(please tick one box only)*



0 ☐ 1 ☐ 2-5 ☐ 6-12 ☐ 13+ ☐

2. Has your child had to stay in hospital for any length of time **over the last year?** Please indicate in weeks the amount of time spent in hospital. *(Please tick one box only)*

0 ☐ less than1 ☐ 1-3 ☐ 4-26 ☐ 27+ ☐

3. How many operations have been carried out on your child **over the last year?** *(Please tick one box only)*

0 ☐ 1 ☐ 2 ☐ 3 ☐ 4+ ☐

4. Has your child had a leg, arm or other part of his/her body in plaster **over the last year?** Please indicate in **weeks** the total time your child has spent in plaster? *(Please tick one box only)*

0 ☐ 6 or less ☐ 7-11 ☐ 12-17 ☐ 18+ ☐

5. Has your child had to wear some form of body or leg support **over the last year?** Please indicate in weeks the total time the support was worn. *(Please tick one box only)*

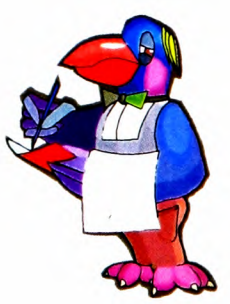
0 ☐ 1-16 ☐ 17-32 ☐ 33-51 ☐ 52 ☐

6. How many tablets, pills or doses of medicines did your child take **yesterday?** *(Please tick one box only)*

0 ☐ 1-4 ☐ 5-8 ☐ 9-12 ☐ 13+ ☐

7. Is your child currently receiving a special diet for any reason? *(Please tick one box only)*

Yes ☐ No ☐



8. How many times has your child suffered from any fits or blackouts **over the last year?** ***(Please tick one box only)***



No fits at all ☐

Occasional fit during the day averaging one per month ☐

Some fits most weeks, day or night ☐

Many fits on most days and nights ☐

Constant fits in frequent succession ☐

9. Has your child been seen by a specialist about difficulties with his/her behaviour **over the last year?** ***(please tick one box only)***

Yes ☐ No ☐

10. How often has your child been seen by any sort of therapist **over the last year?** ***(please tick one box only)***

0 ☐ 1 ☐ 2-12 ☐ 13-52 ☐ 53+ ☐

11. Which of the following services/allowances is your child currently receiving? ***(please tick all boxes that apply)***

Visits from Health Visitor ☐

Visits from Home Help ☐

Visits from Social Worker ☐

Visits from Community Nurse ☐

Voluntary Services ☐

12. How many items of special equipment are there in the home which are currently, or have been essential for your child? ***(please tick one box only)***

0 ☐ 1-3 ☐ 4-7 ☐ 8-10 ☐ 11+ ☐

13. What has been the financial costs to the family for your child **over the last year** in purchasing and maintaining such special equipment? ***(please tick one box only)***

£0 ☐ £1-100 ☐ £101-200 ☐ £201-300 ☐ £301+ ☐



14. What has been the extra financial cost to the family **over the last year** other than in purchasing special equipment, which was not covered by grants and allowances? *(please tick one box only)*

£0 ☐ £1-100 ☐ £101-200 ☐ £201-300 ☐ £301+ ☐

15. Has your child's present home been adapted in any way **over the last year** because of your child? *(please tick one box only)*



Yes ☐ No ☐

If yes, how many adaptations have been made? *(please tick one box only)*

1-3 ☐ 4-7 ☐ 8-10 ☐ 11+ ☐

16. Please indicate how many (further) adaptations are planned or are considered necessary? *(please tick one box only)*

0 ☐ 1-3 ☐ 4-7 ☐ 8-10 ☐ 11+ ☐

17. For **each** of the following activities, please tick **one** of the boxes to indicate how much help you would normally give to your child to complete that activity

	no help given	some help/ supervision given	has to be done for him/her
Washing hands	☐	☐	☐
Eating a bowl of cereal	☐	☐	☐
Putting on a vest/ T-shirt	☐	☐	☐
Doing up buttons or buckles	☐	☐	☐
Getting out of bed	☐	☐	☐
Getting out of the bath	☐	☐	☐
Going to the toilet	☐	☐	☐
Climbing stairs	☐	☐	☐
Getting in and out of a car	☐	☐	☐
Opening doors	☐	☐	☐
Picking up an object from the floor	☐	☐	☐
Carrying a drink the length of a room	☐	☐	☐

18. How many times did you need to lift your child on the last occasion you spent a **full day** with him/her? *(Please estimate number of lifts)*

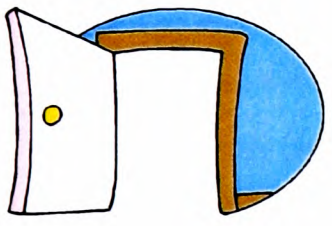
Child's current weight (approximate): ☐☐☐ kg or ☐ stones ☐ lbs

19. How often has your child required assistance during the night **over the last week?** *(please tick one box only)*

0 ☐ 1-3 ☐ 4-7 ☐ 8-10 ☐ 11+ ☐

20. Please list any other areas where your child requires assistance in the course of a normal day?

21.



a) How many rooms (excluding halls and passages) are there in your child's place of residence? ☐

b) Over the **past week**, how many of these did your child go into? ☐

c) How many of these did your child enter unassisted? ☐

22. Does your child normally need help in getting in and out of the house? *(please tick one box only)*

Yes ☐ No ☐

23. What is the furthest distance your child has gone outside without assistance **over the past week?** *(please tick one box only)*

0 ☐ 1-100 yards ☐ 101-440 yards ☐ 1/4 -1/2 mile ☐ 1/2 + miles ☐

24. How often has your child been out of the house by himself/herself **over the pastweek?** *(please tick one box only)*

0 ☐ 1-7 ☐ 8-13 ☐ 14-20 ☐ 21+ ☐

25. Excluding trips to and from school, how many times has your child been on a longer outing **over the past week**, which required some form of transport? *(please tick one box only)*

0 ☐ 1-3 ☐ 4-7 ☐ 8-10 ☐ 11+ ☐

26. What type of school is your child currently attending? *(please tick one box only)*



None ☐

Pre-school (e.g. nursery, playgroup etc.) ☐

Special pre-school ☐

Mainstream without special support unit attached ☐

Mainstream with special support unit attached ☐

Special school: physical disability ☐

Special school: learning difficulties ☐

Home teaching (including portage) ☐

Other (Please specify) ☐

27. How often does you child attend school? *(please tick one box only)*

Part-time ☐ Daily ☐

Weekly boarding ☐ Full-time boarding ☐

28. Approximately how long does it take for your child to travel from home to school? *(please tick one box only)*



0-15 minutes ☐

16-30 minutes ☐

31-45 minutes ☐

46 minutes - 1 hour ☐

1 hour + ☐

29. How many friends has your child seen outside of school hours **over the past week?** *(please tick one box only)*



0 ☐ 1-3 ☐ 4-7 ☐ 8-10 ☐ 11+ ☐

30. Do you have any family or friends locally to whom you can turn for help if necessary? *(please tick one box only)*

Yes ☐ No ☐

31. Do you think that the people in your local area are generally supportive and understanding where your child is concerned? *(please tick one box only)*

Yes ☐ No ☐ Sometimes ☐

32. Do you think that your child restricts your social life in any way? *(please tick one box only)*

Yes ☐ No ☐ Sometimes ☐

33. Do you have any difficulties in organising family holidays because of your child? *(please tick one box only)*



Yes ☐ No ☐

34. Who does your child live with? *(please tick one box only)*

Lives with both birth parents ☐

Lives with one birth parent ☐

Lives with neither birth parent (e.g. grandparents/foster parents, adoptive parents) ☐

35. Please describe if any member of the family has had to change their employment situation to make caring for your child easier.

36. Do you think that your child has placed any extra stress on you as parents/ carers? *(please tick one box only)*



None ☐

Slight ☐

Severe ☐

37. Do you think that your child has placed any extra stress on any other children within the family? *(please tick one box only)*

No other children ☐ None ☐ Slight ☐ Severe ☐

Thank you for your time and co-operation in completing this questionnaire. Please feel free to add comments.



National Perinatal Epidemiology Unit

ORECI
Oxford Register of Early Childhood Impairments

3 September 2004

«ParentTitle»«ParentForenames» «ParentSurname»
«ReturnedAddressLine1»
«ReturnedAddressLine2»
«ReturnedAddressLine3»
«ReturnedPostcode»

Dear «ParentForenames»

Measuring Children's Abilities

You have kindly participated in this study already by providing a rating of «ReturnedForename»'s general physical ability using the enclosed questionnaire. We have surveyed the health professionals whose names you gave us. So far we are able to show that families ratings are reliable when compared to professional ratings.

It is some months since we received your response and we would like to repeat this element of the survey to see if «ReturnedForename»'s ability has changed. We have enclosed the same form again and are asking you to consider which description best represents your child's movement abilities.

We hope that this will not be too much of a burden and thank you once again for taking the time to help with this important research.

Kind regards

Chris Morris
Lead Researcher

AQREC A02.073.sur2inv

Tel: 01865 226706
Fax: 01865 227002
christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Institute of Health Sciences, University of Oxford, OX3 7LF

A



Id No.
«ORECINO»

Manual Ability Questionnaire

I have rated «ReturnedForename»'s manual ability as **MACS Level:**
(please circle only one level)

I **II** **III** **IV** **V**

Do you consent for us to contact «ReturnedForename»'s therapists or paediatrician about this?

□ **Yes** □ **No**

Please also note any changes to occupational or physiotherapists?

Tick this box if you do not wish to take part □

Please return in the prepaid envelope provided
THANK YOU



National Perinatal Epidemiology Unit

ORECI
Oxford Register of Early Childhood Impairments

23 September 2005

«ParentTitle»Parent or carer for «ParentTitle»«ReturnedForename»
«ReturnedSurname»
«ReturnedAddressLine1»
«ReturnedAddressLine2»
«ReturnedPostcode»

Dear «ParentForenames»

Measuring Children's Abilities

We are grateful for your participation in this study. So far we have shown the reliability of family-assessment for a classification system of movement ability. However we recognise that the ability to handle objects is also important for many everyday activities. The MACS is the *Manual Ability Classification System* and was developed recently by researchers in Sweden.

What are we being asked to do?
Enclosed is a leaflet about the MACS and a form for you to indicate which description best represents «Forenames»'s **manual** ability. We are also asking for your consent to contact «Forenames»'s occupational or physical therapists for their assessment.

Why is this necessary?
This will enable us to determine reliability of family assessment for the MACS. The information will also help us understand more about the answers you have provided already in the previous questionnaires.

We hope that this of interest to you and your family and thank you once again for taking the time to help with this important research.

Kind regards

Chris Morris
Lead Researcher

Tel: 01865 226706
Fax: 01865 227002
christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Dept of Public Health, Old Road Campus, University of Oxford, OX3 7LF

Introduction and User Information

The purpose of the ***Manual Ability Classification System*** (MACS) is to provide a systematic method to classify how children with cerebral palsy use their hands **when handling objects in daily activities**. MACS is based upon self-initiated manual ability, with a particular emphasis on handling objects in an individual's personal space (the space immediately close to one's body, as distinct from objects that are not within reach).

The focus of MACS is on determining which level best represents the **child's usual performance** in home, school and community settings. Accordingly, determining the level must be done by asking someone who knows the child well and not by conducting a specific assessment. MACS is not designed to classify best capacity and does not mean to distinguish different capacities between hands. MACS does not intend to explain the underlying reasons for limitations of performance or to classify types of cerebral palsy.

Distinctions between the levels are based on the child's **ability to handle objects** and **need for assistance or adaptations** to perform manual tasks in everyday life. By tasks in everyday life we mean those activities that are relevant and age appropriate for the child including, for example, eating, dressing, playing, writing etc, as distinct from advanced, specially skilled activities such as playing a musical instrument.

As a general principle, if a child's manual ability fits within a particular level the child will probably be classified either at or above that level. Children who do not perform the functions of a particular level will almost certainly be classified below that level. Level I includes children with cerebral palsy with at most minor limitations compared to typically developed children, and where the limitations, if any, barely influence their performances of daily life tasks. The distinctions between each pair of levels are also provided to assist in determining the level that most closely resembles a child's manual abilities.

The scale is ordinal, with no intent that the distances between levels should be considered equal, or that children with cerebral palsy are equally distributed across the 5 levels.

Expert group: Marianne Arner, Eva Beckung, Ann-Christin Eliasson Lena Krumlinde-Sundholm, Peter Rosenbaum, Birgit Rösblad

E-mail:

© Marianne Arner, Eva Beckung, Ann-Christin Eliasson Lena Krumlinde-Sundholm, Peter Rosenbaum , Birgit Rösblad 2004

MACS

Manual Ability Classification System for Children with Cerebral Palsy 8-12 years version II, Sept 2004

© Marianne Arner, Eva Beckung, Ann-Christin Eliasson Lena Krumlinde-Sundholm, Peter Rosenbaum , Birgit Rösblad 2004

Level

I. **Handles objects easily and successfully.** At most, limitations in the ease of performing manual tasks requiring speed and accuracy. However, any limitations in manual abilities do not restrict independence in daily activities.

II. **Handles most objects but with somewhat reduced quality and/or speed of achievement.** Certain activities may be avoided or be achieved with some difficulty; alternative ways of performance might be used but manual abilities do not usually restrict independence in daily activities.

III. **Handles objects with difficulty; needs help to prepare and/or modify activities.** The performance is slow and achieved with limited success regarding quality and quantity. Activities which have been set up or adapted are performed independently.

IV. **Handles a limited selection of easily managed objects in adapted situations.** Performs part of activities with effort and with limited success. Requires continuous support and assistance and/or adapted equipment, for even partial achievement of the activity.

V. **Does not handle objects and has severely limited ability to perform even simple actions.** Requires total assistance.

Distinctions between Levels I and II

Children in Level I may have limitations in handling very small, heavy or fragile objects which demand detailed fine motor control, or efficient coordination between hands. Limitations may also involve performance in new and unfamiliar situations. Children in Level II perform almost the same activities as children in Level I but the quality of performance is decreased, or the performance is slower. Functional differences between hands can limit effectiveness of performance. Children in Level II commonly try to simplify handling of objects, for example by using a surface for support instead of handling objects with both hands.

Distinctions between Levels II and III

Children in Level II handle most objects, although slowly or with reduced quality of performance. Children in Level III commonly need help to prepare the activity and/or adjust the environment since their ability to reach or handle objects is limited. They cannot perform certain activities and their independence is related to the supportiveness of the environmental context.

Distinctions between Levels III and IV

Children in Level III can perform selected activities if the situation is prearranged and if they get supervision and plenty of time. Children in Level IV need continuous help during the activity and can at best participate meaningfully in parts of an activity.

Distinctions between Levels IV and V

Children in Level IV perform part of an activity, however, they need help continuously. Children in Level V might at best participate with a simple movement in special situations, e.g. by pushing a simple button.

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Id No.
«ORECINO»

Manual Ability Questionnaire

I have rated «ReturnedForename»'s manual ability as **MACS Level:**
(please circle only one level)

I **II** **III** **IV** **V**

Do you consent for us to contact «ReturnedForename»'s therapists or paediatrician about this?

□ **Yes** □ **No**

Please also note any changes to occupational or physiotherapists?

Tick this box if you do not wish to take part □

Please return in the prepaid envelope provided
THANK YOU



npeu
National Perinatal Epidemiology Unit

23 September 2005

ORECI
Oxford Register of Early Childhood Impairments

Health professional
name & address

Dear

Re: «ReturnedForename» «ReturnedSurname» DoB «DOB» Study Id No **«ReturnedAddressLine1», «ReturnedAddressLine2», «ReturnedPostcode»**

We are conducting a research study to determine the reliability of family report for the Gross Motor Function Classification System (GMFCS) for children with cerebral palsy. The family of the child named above has provided your name as one of their health professionals and consented for us to contact you.

We would be grateful if you could indicate the GMFCS Level that best represents «ReturnedForename»'s movement ability. If you are not familiar with the system we have provided instructions herewith. Please also indicate whether you made this rating by observing the child in action or by reviewing their clinical record, or whether you used both methods. If you have any questions about this please contact Chris Morris using the details at the bottom of the page.

I have rated this child as GMFCS Level (please circle only one level)

I **II** **III** **IV** **V**

I made this rating using (please tick one)

- ☐ Direct observation
- ☐ Reviewing their clinical records
- ☐ Mixture of both

If you had trouble choosing between levels or any other comments please explain below:

Thank you very much for your time

Tel: 01865 226706
Fax: 01865 227002

christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Old Road Campus, University of Oxford, OX3 7LF

Gross Motor Function Classification System for Cerebral Palsy

Robert Palisano, Peter Rosenbaum, Stephen Walter, Dianne Russell, Ellen Wood, Barbara Galuppi

Introduction & User Instructions

The Gross Motor Function Classification System for cerebral palsy is based on self-initiated movement with particular emphasis on sitting (truncal control) and walking. When defining a 5 level Classification System, our primary criterion was that the distinctions in motor function between levels must be clinically meaningful. Distinctions between levels of motor function are based on functional limitations, the need for assistive technology, including mobility devices (such as walkers, crutches, and canes) and wheeled mobility, and to much lesser extent quality of movement. Level I includes children with neuromotor impairments whose functional limitations are less than what is typically associated with cerebral palsy, and children who have traditionally been diagnosed as having "minimal brain dysfunction" or "cerebral palsy of minimal severity". The distinctions between Levels I and II therefore are not as pronounced as the distinctions between the other Levels, particularly for infants less than 2 years of age.

The focus is on determining which level best represents the child's present abilities and limitations in motor function. Emphasis is on the child's usual performance in home, school, and community settings. It is therefore important to classify on ordinary performance (not best capacity), and not to include judgments about prognosis. Remember the purpose is to classify a child's present gross motor function, not to judge quality of movement or potential for improvement.

The descriptions of the 5 levels are broad and are not intended to describe all aspects of the function of individual children. For example, an infant with hemiplegia who is unable to crawl on hands and knees, but otherwise fits the description of Level I, would be classified in Level I. The scale is ordinal, with no intent that the distances between levels be considered equal or that children with cerebral palsy are equally distributed among the 5 levels. A summary of the distinctions between each pair of levels is provided to assist in determining the level that most closely resembles a child's current gross motor function.

The title for each level represents the highest level of mobility that a child is expected to achieve between 6-12 years of age. We recognize that classification of motor function is dependent on age, especially during infancy and early childhood. For each level, therefore, separate descriptions are provided for children in several age bands. The functional abilities and limitations for each age interval are intended to serve as guidelines, are not comprehensive, and are not norms. Children below age 2 should be considered at their corrected age if they were premature.

An effort has been made to emphasize children's function rather than their limitations. Thus as a general principle, the gross motor function of children who are able to perform the functions described in any particular level will probably be classified at or above that level; in contrast the gross motor functions of children who cannot perform the functions of a particular level will likely be classified below that level.

Reference: Dev Med Child Neurol 1997;39:214-223
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Gross Motor Function Classification System for Cerebral Palsy (GMFCS)

Before 2nd Birthday

Level I — Infants move in and out of sitting and floor sit with both hands free to manipulate objects. Infants crawl on hands and knees, pull to stand and take steps holding on to furniture. Infants walk between 18 months and 2 years of age without the need for any assistive mobility device.

Level II — Infants maintain floor sitting but may need to use their hands for support to maintain balance. Infants creep on their stomach or crawl on hands and knees. Infants may pull to stand and take steps holding on to furniture.

Level III — Infants maintain floor sitting when the low back is supported. Infants roll and creep forward on their stomachs.

Level IV — Infants have head control but trunk support is required for floor sitting. Infants can roll to supine and may roll to prone.

Level V — Physical impairments limit voluntary control of movement. Infants are unable to maintain antigravity head and trunk postures in prone and sitting. Infants require adult assistance to roll.

Between 2nd and 4th Birthday

Level I — Children floor sit with both hands free to manipulate objects. Movements in and out of floor sitting and standing are performed without adult assistance. Children walk as the preferred method of mobility without the need for any assistive mobility device.

Level II — Children floor sit but may have difficulty with balance when both hands are free to manipulate objects. Movements in and out of sitting are performed without adult assistance. Children pull to stand on a stable surface. Children crawl on hands and knees with a reciprocal pattern, cruise holding onto furniture and walk using an assistive mobility device as preferred methods of mobility.

Level III — Children maintain floor sitting often by "W-sitting" (sitting between flexed and internally rotated hips and knees) and may require adult assistance to assume sitting. Children creep on their stomach or crawl on hands and knees (often without reciprocal leg movements) as their primary methods of self-mobility. Children may pull to stand on a stable surface and cruise short distances. Children may walk short distances indoors using an assistive mobility device and adult assistance for steering and turning.

Level IV — Children floor sit when placed, but are unable to maintain alignment and balance without use of their hands for support. Children frequently require adaptive equipment for sitting and standing. Self-mobility for short distances (within a room) is achieved through rolling, creeping on stomach, or crawling on hands and knees without reciprocal leg movement.

Level V — Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent mobility and are transported. Some children achieve self-mobility using a power wheelchair with extensive adaptations.

Between 4th and 6th Birthday

Level I — Children get into and out of, and sit in, a chair without the need for hand support. Children move from the floor and from chair sitting to standing without the need for objects for support. Children walk indoors and outdoors, and climb stairs. Emerging ability to run and jump.

Level II — Children sit in a chair with both hands free to manipulate objects. Children move from the floor to standing and from chair sitting to standing but often require a stable surface to push or pull up on with their arms. Children walk without the need for any assistive mobility device indoors and for short distances on level surfaces outdoors. Children climb stairs holding onto a railing but are unable to run or jump.

Level III — Children sit on a regular chair but may require pelvic or trunk support to maximize hand function. Children move in and out of chair sitting using a stable surface to push on or pull up with their arms. Children walk with an assistive mobility device on level surfaces and climb stairs with assistance from an adult. Children frequently are transported when travelling for long distances or outdoors on uneven terrain.

Level IV — Children sit on a chair but need adaptive seating for trunk control and to maximize hand function. Children move in and out of chair sitting with assistance from an adult or a stable surface to push or pull up on with their arms. Children may at best walk short distances with a walker and adult supervision but have difficulty turning and maintaining balance on uneven surfaces. Children are transported in the community. Children may achieve self-mobility using a power wheelchair.

Level V Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent mobility and are transported. Some children achieve self-mobility using a power wheelchair with extensive adaptations.

Between 6th and 12th Birthday

Level I Children walk indoors and outdoors, and climb stairs without limitations. Children perform gross motor skills including running and jumping but speed, balance, and coordination are reduced.

Level II Children walk indoors and outdoors, and climb stairs holding onto a railing but experience limitations walking on uneven surfaces and inclines, and walking in crowds or confined spaces. Children have at best only minimal ability to perform gross motor skills such as running and jumping.

Level III Children walk indoors or outdoors on a level surface with an assistive mobility device. Children may climb stairs holding onto a railing. Depending on upper limb function, children propel a wheelchair manually or are transported when travelling for long distances or outdoors on uneven terrain.

Level IV Children may maintain levels of function achieved before age 6 or rely more on wheeled mobility at home, school, and in the community. Children may achieve self-mobility using a power wheelchair.

Level V Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At level V, children have no means of independent mobility and are transported. Some children achieve self-mobility using a power wheelchair with extensive adaptations.

Distinctions Between Levels I and II

Compared with children in Level I, children in Level II have limitations in the ease of performing movement transitions; walking outdoors and in the community; the need for assistive mobility devices when beginning to walk; quality of movement; and the ability to perform gross motor skills such as running and jumping.

Distinctions Between Levels II and III

Differences are seen in the degree of achievement of functional mobility. Children in Level III need assistive mobility devices and frequently orthoses to walk, while children in Level II do not require assistive mobility devices after age 4.

Distinctions Between Level III and IV

Differences in sitting ability and mobility exist, even allowing for extensive use of assistive technology. Children in Level III sit independently, have independent floor mobility, and walk with assistive mobility devices. Children in Level IV function in sitting (usually supported) but independent mobility is very limited. Children in Level IV are more likely to be transported or use power mobility.

Distinctions Between Levels IV and V

Children in Level V lack independence even in basic antigravity postural control. Self-mobility is achieved only if the child can learn how to operate an electrically powered wheelchair.

- This work has been supported in part by the Easter Seal Research Institute and the National Health Research and Development Program.
- Distribution of the Gross Motor Function Classification System for Cerebral Palsy has been made possible by a grant from the United Cerebral Palsy Research and Educational Foundation, USA.

Want to know more? Contact:

CanChild
Centre for Childhood Disability Research

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E-mail: canchild@mcmaster.ca
Website: www.fhs.mcmaster.ca/canchild

J



National Perinatal Epidemiology Unit

ORECI

Oxford Register of Early Childhood Impairments

23 September 2005

Health professional
name & address

Dear

Re: «ReturnedForename» «ReturnedSurname» DoB: «DOB» Study Id No:
«ReturnedAddressLine1», «ReturnedAddressLine2», «ReturnedPostcode»

We are conducting a research study to determine the reliability of family-assessment for the **Manual Ability Classification System** (MACS) for children with cerebral palsy. The family of the child named above has provided your name as one of their health professionals and consented for us to contact you.

You may have participated in our similar study of the Gross Motor Function Classification System.

We would be grateful if you could read the enclosed leaflet and indicate the level of the MACS Level that best represents «ReturnedForename»'s manual ability. Please also indicate whether you made this rating by observing the child or by reviewing their clinical record, or you used both methods.

I have rated this child's manual ability as **MACS Level** (please circle only one level)

I **II** **III** **IV** **V**

I made this rating using (please tick one)

- ☐ Direct observation
- ☐ Reviewing their clinical records
- ☐ Mixture of both

If you had trouble choosing between levels or any other comments please explain:

Thank you very much for your time

Tel: 01865 226706
Fax: 01865 227002
christopher.morris@perinat.ox.ac.uk

Chris Morris, NPEU, Dept of Public Health, Old Road Campus, University of Oxford, OX3 7LF

K

unique to each child. Lots of families wrote comments on the forms that provide additional information. This may be presented in reports but the sources of the information will remain entirely confidential.

What's happening now?

We still have quite a number of families who did not respond to our invitation or reminder letters. We have been granted permission by the Research Ethics Committee in Oxford to write once more to these families, this time sending only the single sheet questionnaire for the Gross Motor Function Classification System. Those of you who have taken part in the survey have already helped to show that family ratings of this system are reliable. This second survey will help us to better gauge the range and profile of severity of movement disability in all children with these kinds of movement disabilities in the Thames Valley area. We are also seeking approval to invite the small number of families who have moved outside the Thames Valley to participate in the survey.

Where can I find out more about this?

You can go to our website at www.npeu.ox.ac.uk/morris or contact us directly using the details below.

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Old Road Campus,
Headington
OX3 7LF

Tel: 01865 226706
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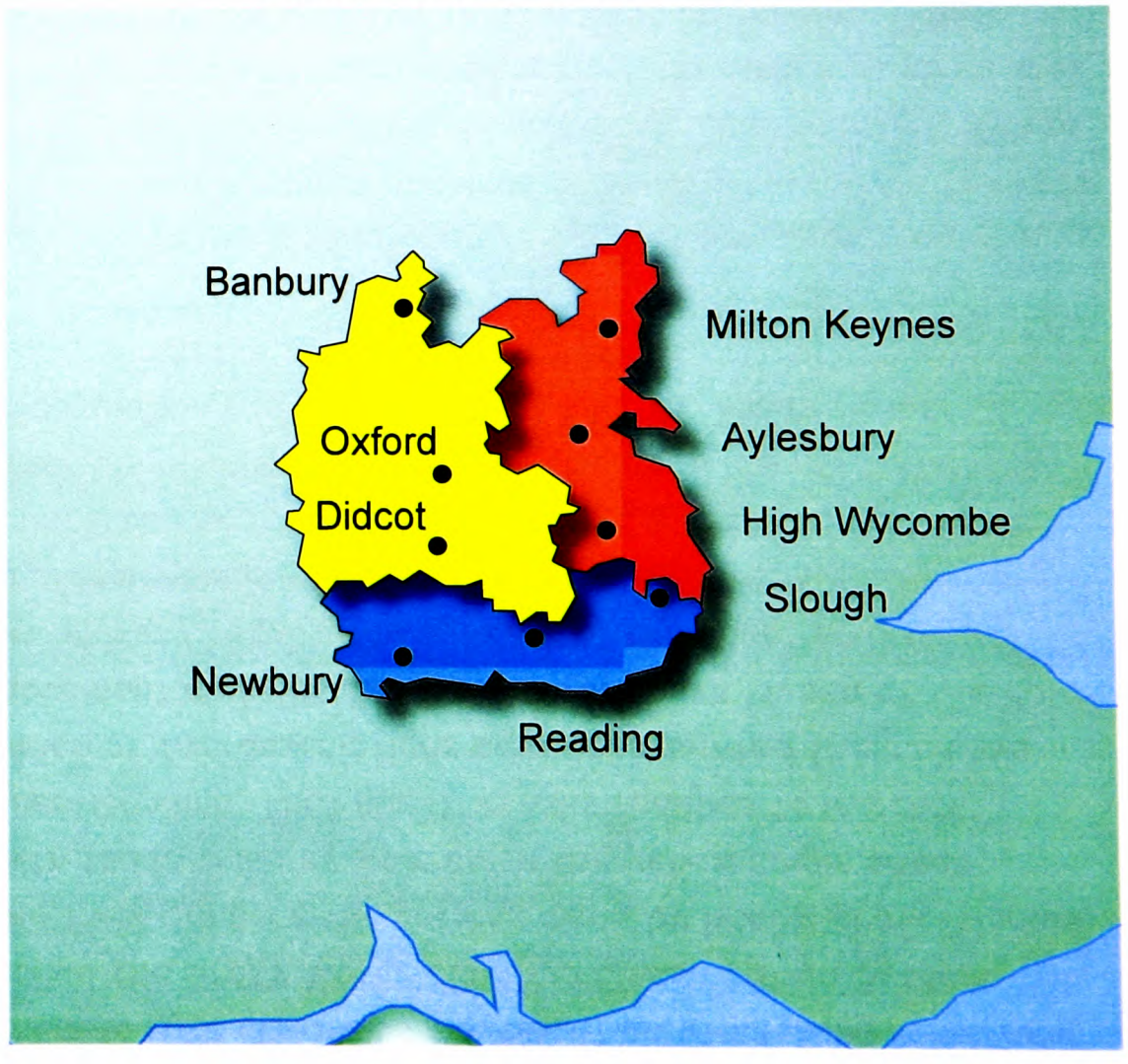
ORECI

May 2004

npeu
National Perinatal Epidemiology Unit

MEASURING CHILDREN'S ABILITIES

A research study investigating the movement abilities of a geographically defined population of children with cerebral palsy



Welcome

It is now nearly a year since we began inviting families to take part in this survey. We would like to thank you again for your interest in this research and provide you with some feedback as to how the work is progressing. This will be one of several newsletters to keep you informed as we are still in the process of collating and analysing the results.

How many people took part?

The families of 282 children, with problems with co-ordination of movement, living in the Thames Valley Health Authority were invited to participate.
The invitations were initially mailed to families in Oxfordshire and West Berkshire at the beginning of the school summer term of 2003. Invitations were then mailed to families living in East Berkshire and Buckinghamshire during the autumn term. We also sent reminder letters as we are never sure whether our letters were received without a response. So far 133 families agreed to participate in the survey, and 112 families have already returned completed questionnaires. There were 31 families who informed us that they did not wish to take part and 6 invitation letters were returned undelivered by the Post Office.

What is happening to the information that was collected?

We conducted a further postal survey with the health professionals whose names families gave us. We asked them to rate children's movement abilities using something called the Gross Motor Function Classification System. We will then compare the ratings made by health professionals with those families gave us on the yellow single sheet questionnaire. That question asks families to indicate which of five descriptions best represented their child's movement abilities. So far when we have looked at the preliminary results the agreement between families and professionals is excellent. This work therefore helps to reinforce the importance of families' judgments of what their children are able to do. The work also demonstrates the reliability of family report for the Gross Motor Function Classification System.



We are also entering all the information from the booklet questionnaires, the Activities Scale for Kids and the Lifestyle Assessment Questionnaire. These are standardised measures that indicate what children are doing physically and how this impacts on families. These will be analysed together with the Gross Motor Function Classification System to investigate what effect different levels of movement ability have on the lives of children and families. The Activities Scale for Kids was not appropriate for children with very limited abilities and this in itself is an important if predictable finding.

We would like to reassure you that nobody is identified in any of part of the research. The findings from each questionnaire are brought together using only the identifying number that was at the top of each form, and

What is happening now?

We are about to contact the few families who moved away from the Thames Valley area to invite them to participate. We will also offer those families who have not responded one more opportunity to take part. The full results from all the questionnaires will then be ready for analysis. Then we will find out what everyday activities children in each GMFCS Level perform regularly and what impact that has only family life.

Of course the assessing movement ability is only one aspect of measuring children's abilities. A team in Sweden have developed the Manual Ability Classification System (MACS) to describe children's ability to handle objects. Because of our success with the original study, we have been offered the opportunity to help in the development of this new measure by testing whether family-assessment is reliable for the MACS. Information from the MACS will also help us understand the findings from the other questionnaires.

Other news

The Oxford Register of Early Childhood Impairments has been collecting information about children born to residents of Berkshire, Buckinghamshire, Northamptonshire and Oxfordshire since 1984. The register has recently changed its name to ***4Child*** Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children. More details are available on the website www.npeu.ox.ac.uk/4Child.

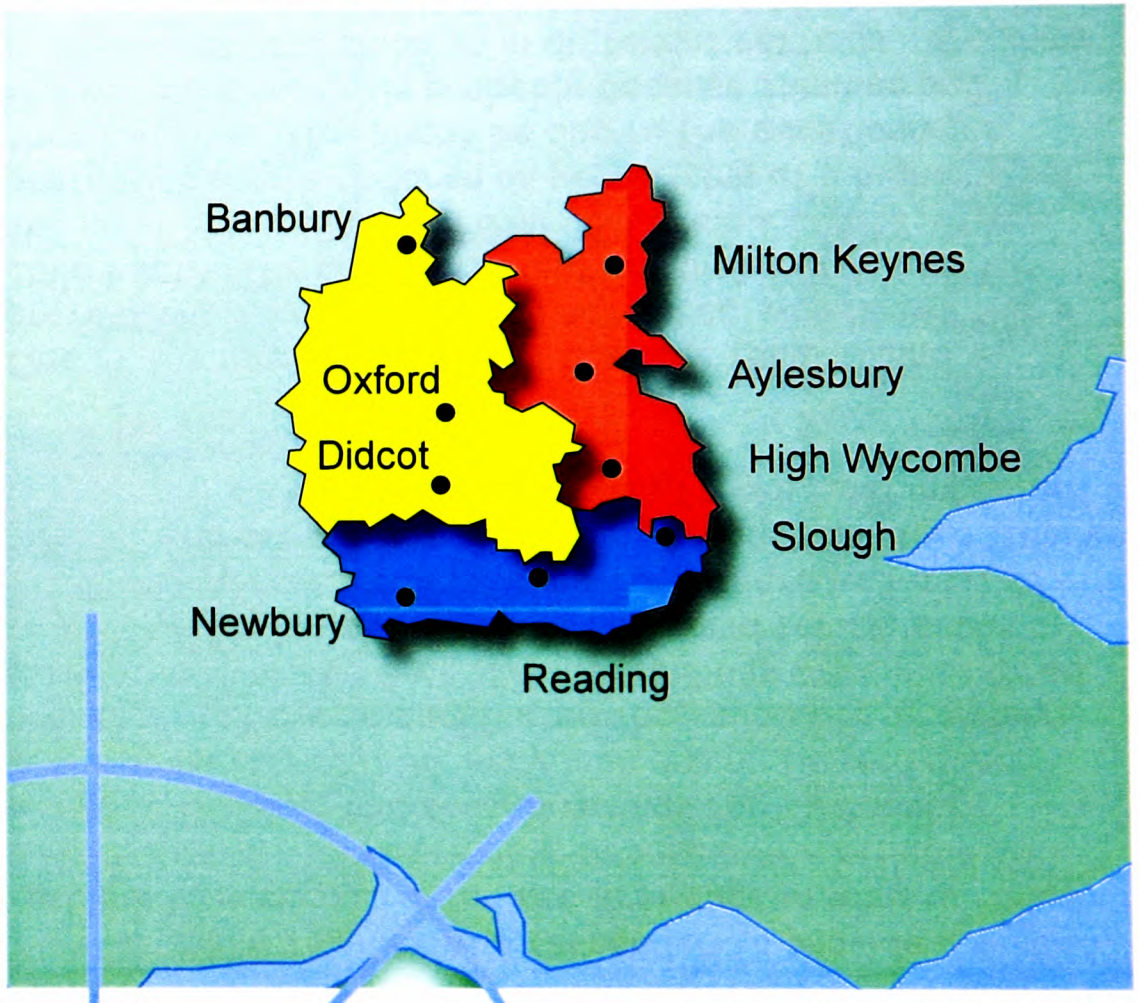
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AQREC A02.073.newslOct04

MEASURING CHILDREN'S ABILITIES

A research study investigating the movement abilities of a geographically defined population of children with cerebral palsy



October 2004

4Child
Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children

npeu
National Perinatal Epidemiology Unit

Welcome

Welcome to this second newsletter for the "Measuring Children's Abilities" study. We would like to thank you for your continuing interest in this research. Much progress has been made since our last newsletter in May and this leaflet aims to keep you informed on what is being done with the information you gave us.

What is the Gross Motor Function Classification System?

Until recently there has not been a satisfactory overall measure of the movement abilities of children with cerebral palsy. Words such as mild moderate and severe mean different things to different people, and also to families and professionals. In 1997 a team of researchers called CanChild published the Gross Motor Function Classification System (GMFCS). The GMFCS classifies children's ability to sit stand and walk, at different ages, into five levels. The GMFCS has been widely adopted by health professionals in research studies and helped to increase our knowledge about cerebral palsy, but has so far required a professional to make the assessment.



What has this study found out so far?

We thought that families could also make this assessment. One of the questionnaires that families in this survey completed (the yellow one) asked which of five descriptions best represented their child's movement ability. These descriptions were taken almost word for word from the GMFCS Levels for children between 6 and 12 years. We surveyed the child's health professionals for their assessment of the child's GMFCS and compared the results. The statistical method used to assess reliability produces a number between 0 and 1, where the closer the number is to 1 the better the reliability.

In this study we were able to compare the reliability of GMFCS assessments made by families with those made by physiotherapists, paediatricians and orthopaedic surgeons. The reliability between physiotherapists and families for 76 children was 0.94, which is very high. The reliability between paediatricians and families for 65 children was 0.91, similarly high, and between families and orthopaedic surgeons for 37 children was 0.84, still very good.

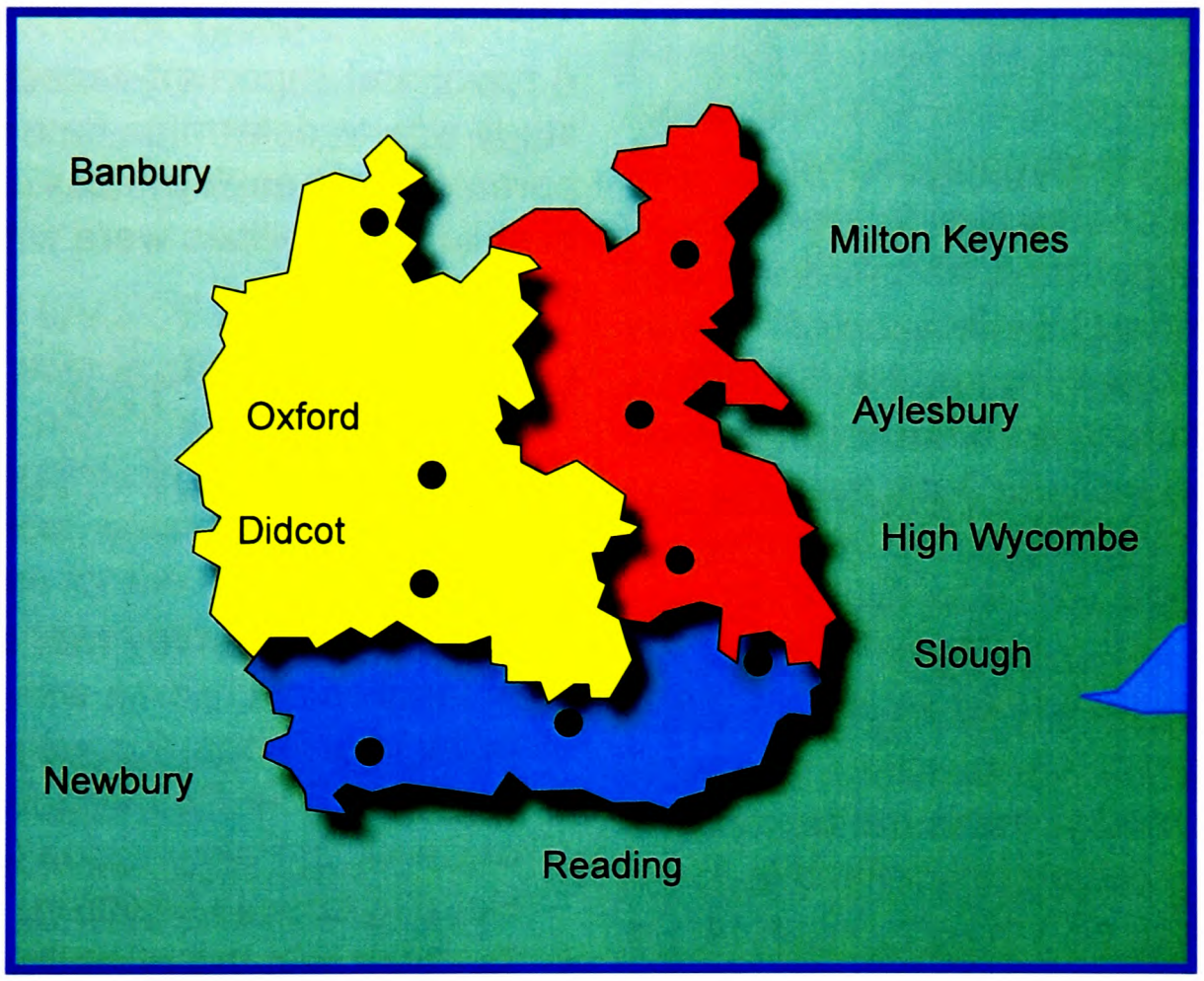
We also asked families who responded to provide a second rating 3 to 6 months after the first time in what is called a retest study. We were able to assess the reliability between the first and second ratings for 75 families in the same way as 0.98, very high again. So well done, together we have demonstrated the reliability of family-assessment for the GMFCS! The findings were presented to the largest forum in the world for cerebral palsy research last month and there was a lot of interest in what the findings represent, that family-self assessment can provide reliable and important information.

One of the interesting issues is where families and professionals assessments do not agree, who knows the child best? The only consistent unreliability occurs for the most able children in GMFCS Levels I and II, where professional rate children as performing at a higher level than families. This might be due to the questionnaire but we think that it is probably because families and professionals see children in different settings. We know from other research that children use different mobility methods at home, at school and in the community. Despite the excellent findings, not every family found a description that they felt adequately described their child's abilities. There were 4 families in the first survey, and 7 in the second, that indicated that their child's movement abilities fell between the levels offered.

MEASURING CHILDREN'S ABILITIES

A research study investigating the movement abilities of a geographically defined population of children with cerebral palsy

Final Report



December 2005

4Child
Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children

University of Oxford

CanChild
Centre for Childhood Disability Research

npeu
National Perinatal Epidemiology Unit

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Why have I received this report?

The final report of this research study is being distributed to all the families who were invited to take part, and to the health professionals nominated by participating families. In addition, we are sending copies to organisations for which we believe there are implications for policy, such as the Department of Health and the Thames Valley Health Authority.

What was the study about?

The World Health Organisation has recently developed a new framework for thinking about "health" called the International Classification of Functioning, Disability and Health (ICF). The ICF distinguishes what might be right or wrong with our body from the way we get involved in life situations, by recognising that our 'participation' in life is affected by personal and environmental factors as well as how our bodies work.

This study aimed to measure the activities and participation (as defined in the ICF) of children with movement problems falling within the umbrella term 'cerebral palsy'. The number of children affected by these conditions has been observed for many years in several countries. However, as yet, we have not been able to reliably describe children's physical abilities in those populations because, until recently, there have not been adequate valid and reliable measurement tools.

Why is this study important?

Usually this type of research only involves families of children attending hospital appointments who might have more complicated or different issues than the ones who don't come to clinics very often. The study described in this newsletter invited families of a geographically defined population, which were the children with movement problems born to mothers resident in the Thames Valley between 1991 and 1997.

A novel and important aspect of this study was to determine how consistent parents' and health professionals' reports of children's abilities were with one another.

What were the objectives of this study?

- To conduct a survey with a population of families of children with cerebral palsy in collaboration with researchers at *4Child* Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children.
- To determine the reliability of families reporting their child's movement and manual abilities using the Gross Motor Function and Manual Ability Classification Systems (GMFCS and MACS).
- To measure and describe children's activities and participation using family-assessed instruments. A review of existing instruments undertaken as part of this study identified the Activities Scale for Kids (ASK) and Lifestyle Assessment Questionnaire (LAQ-CP).

- To examine associations between children's impairments and abilities (the 'severity' of their cerebral palsy) and their subsequent activities and participation as defined in the ICF.

How was information gathered?

Participating families were mailed a package of questionnaires that included a single question asking which of five descriptions best represented their child's movement ability. This corresponded to the GMFCS Level. Some families classified their children a second time after a few months to examine whether there was any change in the child's classification.

The ASK and LAQ-CP include a variety of questions asking about the child's activities and participation in terms of physical independence, mobility and also any clinical, economic and social consequences for the family. Scores can then be calculated for each of these domains.

When a new classification system for describing children's ability to handle objects became available (MACS) participating families and the health professionals were then also asked to rate their child's manual abilities.

Families nominated their child's health professionals, such as paediatricians and physiotherapists, and they were also surveyed for their classification of the child's GMFCS and MACS Levels independent of what the parents said.

Who took part?

Families of 129/314 (41%) of children fully participated in the study, and the families of 175/314 (56%) children provided reports of their child's movement ability using the GMFCS (general physical ability) questionnaire. Fully participating children were shown to be broadly representative of the complete population in terms of their age, sex and characteristics of their impairments.

What did we find out?

Families' classification of their children's movement and manual abilities, using the GMFCS and MACS, was highly reliable (consistent) compared to ratings made by their health professionals. There was not perfect agreement between families and professionals but any disagreement was rarely by more than one level.

The best available instruments for families to report their child's activities and participation were identified and administered in a postal survey. The scores derived from the instruments described a wide range of involvement in life situations.

Children's physical independence and mobility scores were well explained by whole body movement, manual skills and intellectual ability, and also the presence of seizures and speech problems.

Social integration (including stress), clinical and economic burden scores were not well explained by children's abilities. These outcomes for the child and family are therefore likely to be more importantly influenced by personal and environmental factors.

Not many families in the survey indicated their child had access to occupational therapy.

What are the recommendations from this research?

Better measures of children's activities and participation are required to more completely reflect children's activities and participation as defined by the ICF. Any new measures should be general in what they ask about and not specific to particular diagnosis.

As families know how their child performs across a broader range of settings than do professionals, family assessment is likely to reflect the child's functioning more accurately than professional judgements. **Clinicians and researchers** should therefore use family assessment of children's abilities.

Commissioners and policymakers for health and social care must be aware that families of children of all levels of ability are prone to stress and social exclusion and therefore require support in the community.

We are not sure why some families chose not to take part in this study, although for some it may simply not have been of interest. Therefore researchers should engage with families to increase the relevance of studies undertaken and thereby improve the response.

On this last point we welcome your feedback on this study and, although we are unable to guarantee that your ideas will be possible to pursue in studies, any suggestions you might have for research you would like to see conducted in the future.

Academic publications from this research

Morris C, Kurinczuk JJ, Fitzpatrick R. Rosenbaum P. Do children's abilities explain their activities and participation? (Submitted to Developmental Medicine and Child Neurology)

Morris C, Kurinczuk JJ, Fitzpatrick R. Rosenbaum P. Families know their children best: family assessment of disability is highly reliable. (Submitted to Archives of Disease in Childhood)

Morris C, Kurinczuk JJ, Fitzpatrick R. (2005) Child or family self-assessed measures of activity performance and participation for children with cerebral palsy: a structured review. Child: Care Health and Development 31(4): 397-407.

Morris C, Galuppi B, Rosenbaum PL. (2004) Reliability of Family Report for the Gross Motor Function Classification System. Developmental Medicine and Child Neurology 46(7):455-60

Morris C, Bartlett D. (2004) Gross Motor Function Classification System: impact and utility. Developmental Medicine and Child Neurology 46(1):60-5.

The findings have also been presented at meetings of the European Academy of Childhood Disability and the American Academy for Cerebral Palsy and Developmental Medicine.

Chris Morris, the lead researcher, submitted his doctoral thesis at the University of Oxford in 2005 based on the work in this research study.

We are grateful to all the children, families and health professionals who made this work possible by participating in the survey.

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APPENDIX B

RELEVANT PUBLICATIONS,

ABSTRACTS, POSTERS

& LETTERS