

What factors impact on patient access and engagement with clubfoot treatment in low and middle-income countries?: a meta-synthesis of existing qualitative studies using a Social Ecological Model

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Objectives

Annually around 100,000 babies are born with feet twisted inwards, a condition known as ‘clubfoot’, which limits participation in everyday life. Clubfoot can be treated using non-surgical manipulation, but many guardians in low and middle-income countries (LMICs) do not present for treatment. The Social Ecological Model specifies five inter-related factors that may impact on this: intrapersonal, interpersonal, institutional, community or socio-cultural factors and public policy. Using this model, we conducted a systematic synthesis of previous research to identify factors that impact on treatment seeking for clubfoot and community level interventions to improve engagement in LMICs.

Methods

A search of five databases was conducted and articles screened using six criteria. Quality was appraised using the Critical Appraisal Skills Programme (CASP) checklist. Eleven studies were identified for inclusion. Analysis was informed by a Social Ecological Model.

Results

Intrapersonal barriers experienced were a lack of income and additional responsibilities. At the interpersonal level, support from fathers, the extended family and wider community impacted on treatment seeking. Institutional or organisational factors included long distances to treatment centres, insufficient information about treatments and challenges following treatment. Guardians’ beliefs about the causes of clubfoot shaped behaviour. At the level of public policy, two-tiered healthcare systems made it difficult for some groups to access timely care. Interventions to address these challenges included counselling sessions, outreach clinics, brace recycling and a range of education programmes.

Conclusions

This study identified factors that impact on access and engagement with clubfoot treatment across diverse settings and strategies to address them.

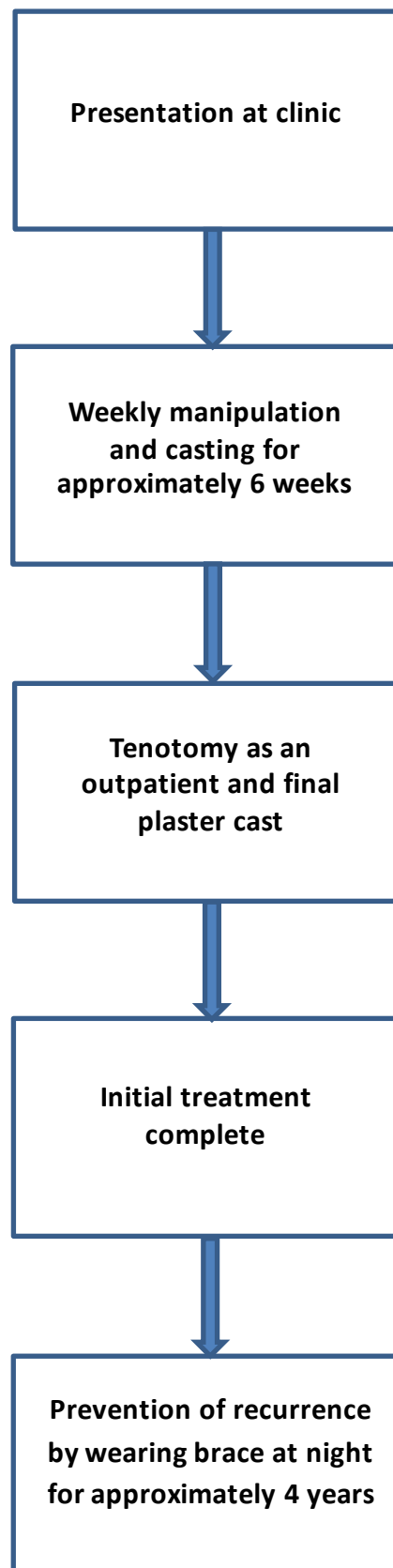
Keywords

Qualitative research; Meta-synthesis; Social Ecological Model; Low and middle-income countries; Clubfoot; Paediatric orthopaedics

Introduction

Worldwide, around 100,000 children are thought to be born annually with clubfoot and of those with clubfoot, 80% live in low and middle-income countries (LMICs) [1]. Babies born with clubfoot have a congenital disability that affects one or both feet. In medical terms this is described as 'equinovarus', which means that feet point downwards at the ankle and are twisted inwards. This does not spontaneously resolve. Untreated clubfoot can be painful, impair function including mobility, limit participation in everyday life and in some cases, result in ostracism from the community [2]. Clubfoot can be corrected surgically, or conservatively, using the Ponseti method, by stretching and manipulating the affected leg or legs [3]. This involves approximately six progressive manipulations of the foot at weekly intervals, using a plaster cast between manipulations to maintain correction. After these manipulations there is usually a need for a small outpatient operation to release the tight Achilles tendon, and one more plaster cast that is worn for three weeks. At this point the original twist will have been resolved, and initial treatment is complete, but as there will be tendency for the twist in the leg to recur, braces are recommended to be worn at night for approximately four years once a child is walking (for a full description of the treatment see Ponseti IV, 1966) [4]. An outline of the main stages of the Ponseti method is presented in Figure 1. A number of studies have indicated that the Ponseti method is an effective treatment for clubfoot, in the short and long term [5-7], and that untreated clubfoot or clubfoot that has 'relapsed' after treatment may benefit from more invasive surgical procedures [8]. Treatment for clubfoot is ostensibly widely available. 113 of 193 United Nations member countries have established clinics that provide Ponseti treatment, including high volumes of such clinics in LMICs [9].

Figure 1: Outline of the main stages of clubfoot treatment using the Ponseti method



However, a number of factors have been identified that act as barriers to service delivery including financial constraints of the service [10-12], a related lack of resources [11] and a lack of training for physicians [11, 13, 14]. In addition, it is thought that the majority of patients with clubfoot in LMICs do not come forward for treatment, and that many of those who do start treatment do not continue with it [12, 14-17]. Understanding factors that impact on access and adherence to clubfoot treatment may help to inform ways of encouraging uptake and engagement with services that have the potential to improve a child's wellbeing and ability to participate in a range of everyday activities.

There is an increasing recognition of the need to deliver 'person-centred care' in LMICs. According to the World Health Organisation, this involves prioritising patients' experiences and needs in the design of services, including those of their significant others and communities [18]. To do this for treatment of any condition, there is a need to identify and describe how people experience health and healthcare, their preferences for care and rationale for their decisions about healthcare access and use [19]. Once these are understood then services can be developed to meet their needs most appropriately. Qualitative studies are particularly adept at providing information about experiences of health and healthcare, and a number of qualitative studies have identified factors that impact on patient access to clubfoot treatment in LMICs and suggested community level interventions to address them [11, 16, 17, 20, 21]. Increasingly it is thought that synthesis of previous qualitative work can deliver results that are of relevance to multiple contexts [22]. With a growing body of qualitative literature focusing on uptake and adherence to care for clubfoot, we aimed to synthesise these findings to identify common themes or issues across a range of settings that could then inform models of healthcare provision.

Theoretical frameworks deriving from a range of disciplines have been constructed to understand influences on treatment-seeking behavior [23]. This study uses a Social Ecological Model since this model recognises the impact of multiple and inter-related factors on illness behavior [24]. This provides a counterpoint to individualistic models that see behavior as the result of personal characteristics or rational decision-making processes [25]. According to the Social Ecological Model, there are five inter-related determinants of illness behavior: 1) intrapersonal factors that refer to individual characteristics such as socio-economic status and occupation; 2) interpersonal processes or personal interactions with others; 3) institutional factors that are the processes embedded in organisations; 4) community level factors such as values and beliefs in the wider society and 5) public policy [24]. Ecological frameworks have been used to inform the development of public health

programs in LMIC settings[26] and could provide clinicians with a practical tool for initiating change in the management of clubfoot.

A number of methods have been developed to synthesise qualitative and mixed methods research[27, 28]. These can be divided into approaches that aim to integrate findings such as meta-synthesis[29] and interpretive approaches such as meta-ethnography that aim to generate new theories from existing literature[30] although they are best understood as part of a continuum[31]. Meta-synthesis has been selected since it allows us to combine studies that use a range of methodologies[27].

The aim of this study is to use a meta-synthesis to synthesise findings from existing qualitative research to explore factors that impact on patient access and adherence to clubfoot treatment in LMICs and community level interventions to address them. A Social Ecological Model is used to structure analysis[24]. This may help healthcare professionals develop services to better meet the needs of patients in these settings.

Methods

A meta-synthesis of existing literature exploring patient engagement and adherence to clubfoot treatment and strategies to address these challenges was conducted in four stages: identifying studies for inclusion, appraising quality, data extraction and synthesis and reporting findings.

Identifying studies for inclusion

This involved a comprehensive review of relevant studies using Ovid MEDLINE, PsycINFO, Embase, Global Health and CINAHL. We originally intended to use the 'SPICE' framework (**S**etting, **P**articipants, **I**ntervention, **C**omparison and **E**valuation), to define the parameters of the qualitative synthesis and identify relevant search terms[32]. However, due to the paucity of qualitative research conducted on clubfoot services and treatments this did not appear to be of great value and we instead broadened the parameters of the search to ensure maximum capture of relevant publications using a combination of keyword searches and thesaurus terms or subject headings (Table 1). All the search terms were systematically applied to each database. A qualitative search filter based on that developed by the School of Public Health at the University of Texas was then

applied[33]. The search syntax for each database is detailed in Tables 2 and 3. Databases were searched in October 2015.

Table 1: Search terms and qualitative filters used to explore patient experiences of accessing services to correct clubfoot

Patient	Intervention
Clubfoot	Service*
“Club-foot”	Ponseti
Talipes	Surg*/ Surgic*
Equinovarus	Correction
“Idiopathic clubfoot”	Treatment
	Therapy
	Tenotomy

The lead researcher manually screened articles to identify studies which fulfilled the following criteria:

1. The study was focused on clubfoot services or treatments
2. The population included patients, parents, guardians, their close friends and family or healthcare professionals involved in the organisation or delivery of clubfoot treatment
3. The care setting was in low or middle income countries as defined by the World Bank [34]
4. The study evaluated patient engagement with services
5. The study was published in the last 10 years
6. The study used qualitative research methods
7. The research output was either an article or a report

Our initial objective was to limit ourselves to studies with patients, parents, guardians or their close friends and family. However, we found that a number of studies with healthcare professionals also reported perceived patient experiences. A decision was therefore made to include them in the review because the findings included within them were likely to provide valuable information about service use. Since the study aims to evaluate current or recent service provision, articles were limited to the last ten years.

Bibliographies of relevant articles identified were then hand searched for additional material.

Appraising quality

Quality appraisal was based on the Critical Appraisal Skills Programme (CASP), a 10 point framework constructed to facilitate evaluation of the quality and usefulness of qualitative studies[35]. To do this, we grouped the items of the CASP framework into three domains: 1) CASP items about the study's aims and appropriateness of methodology (items 1 and 2); 2) CASP items about study design and conduct, including research design, recruitment, data collection, relationship with researcher, ethics and analysis (items 3-8); and 3) CASP items about clarity of findings and value of the research (items 9-10). We then considered the characteristics of each study in relation to these items and domains. Based on these, the articles identified were classified as 'fully address CASP items', 'mainly address CASP items' or 'partially address CASP items'. Studies that fully addressed CASP items attended fully to *all* of the items; studies that mainly addressed CASP items attended fully to *most* of the items; studies that partially addressed CASP items attended fully to *some* of the items. The distinction between those considered to mainly address items and those that partially addressed items, were based on judgement rather than absolute criteria.

Articles were independently appraised by an experienced qualitative researcher and member of the research team (RGH). Both quality appraisers had independently arrived at the same judgements about the 13 articles, in which three studies were deemed to fully address items, four mainly address items and four partially address items. However, as all 13 articles contributed valuable insights into the phenomena, a decision was made to include all of them in the review. This accords with advice from Campbell and colleagues on the use of appraisal frameworks in qualitative syntheses[36]. Descriptions of the characteristics of each study in relation to the three CASP domains along with quality assessments are detailed in Table 4.

Data extraction and synthesis

The full articles or project reports were imported into NVivo qualitative analysis software[37] and analysed by the Lead Researcher (SD) using a thematic approach. That is, coding the data to identify themes and subthemes in the articles[38]. Analysis included primary data such as participant quotes and field note extracts included in the articles, and the secondary interpretations of authors. An index of codes was generated. An abductive analysis[39] was then conducted such that codes were transposed onto the five levels of the Social Ecological Model[24]. 50% studies were double-coded

by a researcher from the wider study team (RGH) and the index of codes discussed and refined to reach a single code list.

Reporting findings

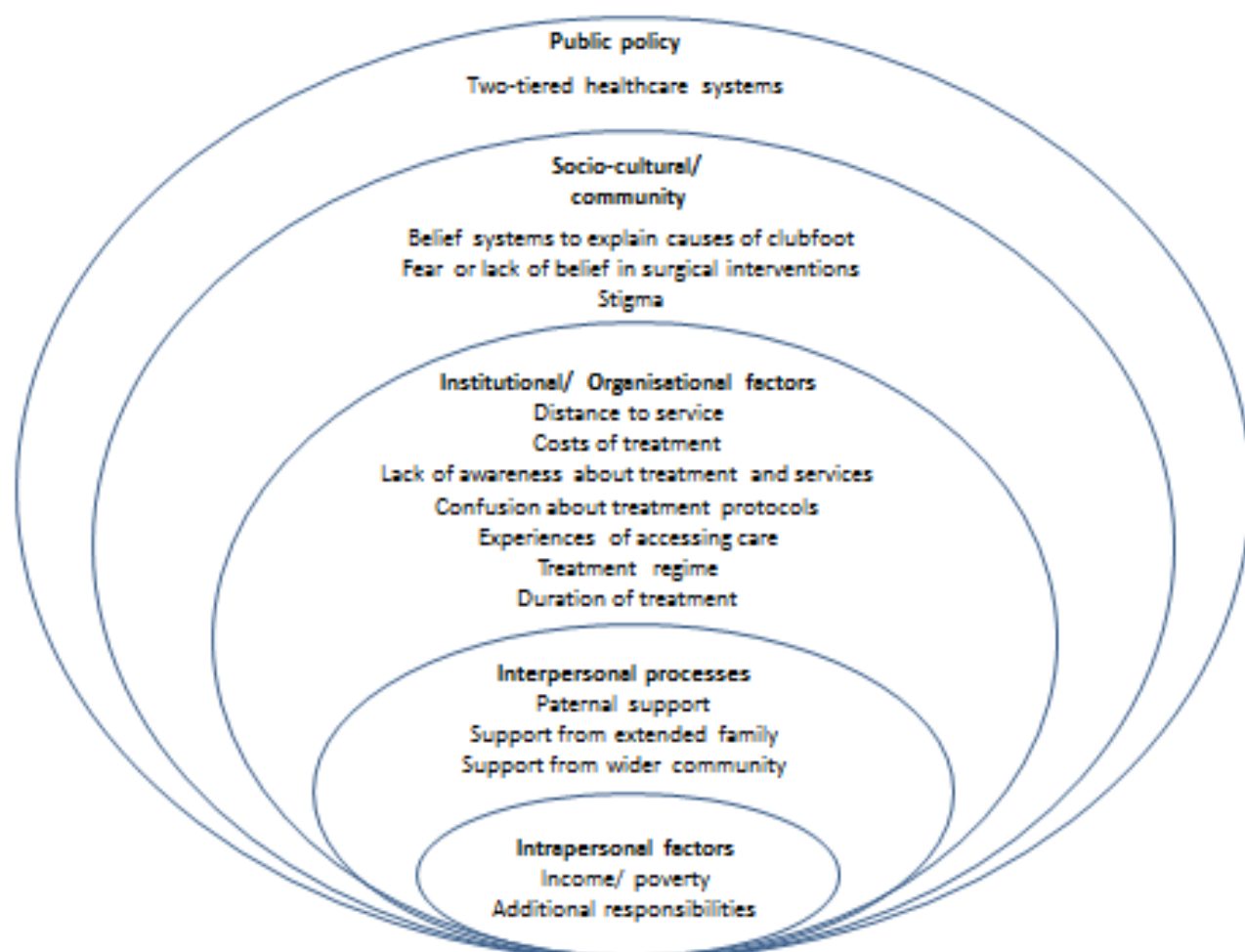
To facilitate transparency, the review was presented in accordance with ENTREQ guidelines, a 21 item list to improve the reporting of qualitative syntheses[22].

Results

77 papers were initially identified from the search criteria and 13 included in the review. The process of identifying studies relevant for inclusion is detailed in a PRISMA flow chart in Figure 2. Of these, two articles were identified as being part of the same study and another three employed mixed methods. Studies explored barriers and facilitators to treatment in the following care settings: Nigeria (1), Malawi (1), Uganda (1), Kenya (1), Latin America (2), India (1), Sri Lanka (1), China (1), Brazil (1), Vietnam (1). A summary of the characteristics of the studies are presented in Table 4. This includes care settings, methodology, study aims and quality appraisal. Articles derived from the same study are grouped and indicated in blue. Articles derived from the same study are cited only once to ensure they do not appear to be over-represented in the analysis.

Factors influencing treatment seeking behaviour were experienced throughout the care pathway from initial presentation, adherence to casting regime and bracing and a number of community level interventions to address these challenges identified. A summary of the main factors influencing treatment seeking and their relation to the five levels of the Social Ecological model[24] are presented in Figure 3.

Figure 3: Factors influencing treatment seeking behaviour: codes identified and their relation to the 5 levels of the Social Ecological Model



Below we explore factors impacting on service use and treatment in more detail, using the five levels of the Social Ecological Model to inform findings. We also identify community level interventions to address these challenges. It is important to note that as a review article we are providing a distillation of the work of others, rather than endorsing that work.

Intrapersonal factors

The income of carers presented a potential barrier to treatment in all studies. For the poorest, a lack of access to resources presented an almost insurmountable challenge. ‘Hidden costs’ of accessing treatment included travel expenses[11, 12, 40-44] and money lost taking time off from economically productive activity[10-12, 40, 43]. For some, a commitment to adherence meant prioritising treatment at the expense of other essentials such as household amenities and education [40, 41]. In

Peru and Kenya, carers staying near the treatment centre incurred the cost of accommodation [42, 43].

Additional responsibilities were a factor since parents were often forced to manage commitments to work[11-13, 40-42] and within the home[13, 40, 41]. Treatment seeking sometimes became a secondary issue that was superseded by other, more immediate problems such as acute illness or hunger[11, 41]. Practitioners in Kenya thought treatment was delayed when parents were planning to have more children[43].

Interpersonal processes

The role of social support networks in influencing access to treatment was identified in just under half of the studies[10, 12, 13, 40, 41]. Paternal support was seen to have a major influence since fathers were a potential source of emotional and practical support[12, 40, 41]. In some contexts, women did not seek care for their children because of fathers' decisions about access and use of family finances[40, 41, 44]. In Kenya, one study described how 'competition' with multiple wives meant mothers may be reluctant to seek support from their husbands[43]. Elsewhere, fathers were more likely to provide support[41] and accompany their children to clinics[11].

The potential influence of the extended family and wider community varied. For societies with a strong convention of respect for elders, treatment decisions could be made by older members of the extended family[10, 40, 41] or influenced by the advice of community leaders such as pastors [40, 41, 44]. Some guardians also received practical support from the extended family[40, 45] or friends[10, 13, 40, 41].

Institutional/organisational factors

In almost all of the studies, many guardians were seen to face long journeys to treatment centres[10, 12, 40-42, 44], resulting in high transport costs[41-44] and long periods away from other responsibilities[12, 13, 42, 44]. Accessing reliable transport was viewed as challenging[12, 13, 40, 41]. Since the majority of treatment sites existed in urban areas, this tended to make treatment seeking more difficult for those living rurally[10-13, 40-42, 45].

Treatment costs impacted on treatment seeking behaviour. Whilst in some care settings free treatment facilitated access[21, 45], in others, carers faced substantial costs such as purchasing materials for casting and braces[13, 14, 41-45]. In Uganda, although healthcare services state that they provide free treatment for clubfoot, carers sometimes had to purchase materials when clinics ran out or were asked by healthcare professionals to pay for treatment[41]. The existence of regional variation in costs was identified[10, 13, 14, 41].

Healthcare professionals thought that some guardians were not aware that the condition could be treated and that services existed to do so[10-12, 40-43]. Some guardians did not know that the bracing portion of the treatment was an ongoing element of care and stopped using the brace after casting[11, 13-15, 40, 42]. Both guardians and healthcare professionals described confusion about the bracing protocol, such as when braces should be worn and for what duration [10, 11, 41, 43, 45].

Negative experiences in accessing care meant guardians were less likely to pursue treatment. In Uganda, issues included healthcare professionals' requests for payment, the difficulty of travelling to different regions for casting and bracing[41] and healthcare professionals not arriving to provide care at clinics[41]. In other areas, guardians encountered crowded clinics, long waiting times[10, 11, 15] and there were reports of intimidating or aggressive professionals[40]. Some clinics ran out of materials meaning that treatment was either delayed or carers were forced to pay[14, 15, 42]. Carers were also less likely to consent to casting if they felt that their children had been treated incorrectly previously[10].

The treatment regime presented a number of challenges. These included practical issues such as difficulties in holding and bathing a child wearing a brace[10, 11, 14, 41, 43] and concerns about discomfort[10-14, 40, 41, 45]. Duration of treatment was also problematic as it meant that any barriers had to be negotiated over a number of years[10, 40, 41]. For instance, although some parents were able to afford initial clinic attendance, they were unable to maintain adherence due to cumulative costs[13] or in the case of those living in a position of financial instability, unable to ensure they could regularly acquire the money needed[41].

Socio-cultural/ community factors

The impact of socio-cultural factors was explored in just over half of the studies and differed widely between care contexts. Factors included belief in biomedical models, faith in alternative belief

systems that provided explanation of causes of clubfoot such as witchcraft or God[40, 41, 43, 44], fear or a disbelief in surgical interventions[10, 11, 41, 43] and stigma associated with the condition[10, 11, 13, 40, 41, 44].

Although there was not a direct relationship between beliefs and treatment-seeking behaviour[40], these helped to shape decisions in nuanced ways. For instance, beliefs about causation and non-biomedical systems meant carers often sought traditional medicine instead, or as well as, biomedical interventions[11, 40, 41, 43]. In some cases it was reported that carers were reluctant to intervene if they saw clubfoot as 'God's will'[41] or a 'gift'[10]. The stigma associated with clubfoot impacted on treatment-seeking in different ways. For some, it provided a powerful impetus to seek a cure[10, 40], whilst for others it was a hindrance since they did not want to 'advertise' that their child was different to others[11, 13, 40, 41, 44].

Public policy

Participants in the included studies tended to focus on factors at the individual and organisational levels. In Latin America and Brazil, the two-tiered healthcare system presented a barrier[13, 14, 42]. Guardians who were dependant on social services or cheap insurance policies experienced longer delays in acquiring braces than those accessing comprehensive private care [14], along with higher treatment costs as some insurance policies did not cover the costs of congenital conditions[13, 42]. This made it more challenging for these groups to access timely and effective care for their children.

Community level interventions to address challenges

Authors and a number of participants identified interventions at the community level to address the challenges identified. To enhance interpersonal support, it was suggested that clubfoot appointments should be arranged on the same day by clinicians, preferably in organised clinics, to enable carers to provide each other with emotional and practical support [12, 13, 40, 41]. Observing the progress of other children was also thought to enable parents and guardians to continue to bring children for treatment and carry on with treatment protocols at home [12, 13, 40, 41]. Building counselling into treatment was viewed as an opportunity to provide carers with emotional support and to enable practitioners to help parents and guardians with use of treatment [40, 41]. It was suggested that involving fathers in this process may help strengthen paternal support[40, 41].

To mitigate the barriers identified at the institutional or organisational level, it was felt that introducing outreach clinics in rural areas[40, 41], or training professionals in these areas where this was not possible[44], could address the long journeys carers face in accessing treatment. It was thought costs of care could be diffused by creating brace recycling programmes [11, 13, 14, 41] or by procuring financial assistance from non-governmental organisations[43]. Another author suggested relationships between clinicians and carers may be improved by introducing protocols to help clinicians provide care[40]. Monitoring of practice would help ensure that standards and consistency were maintained[41]. It was suggested that duration of treatment, although largely unavoidable, may be addressed by the introduction of the accelerated Ponseti method involving shorter periods between casts[40]. However, this would only reduce the casting portion of the treatment.

Authors and participants thought that education programmes should be introduced to promote awareness of treatment and provide information to guardians accessing care. The general public could receive information through a range of media such as newspapers, radio, posters, leaflets [11, 21, 41] or the internet[11, 42]. Community centres such as schools may also be used alongside influential community leaders such as pastors[40, 41]. The importance of using appropriate media and taking into account levels of literacy and affluence of groups in society was emphasised in a number of studies[10, 41, 42]. Educating healthcare professionals in other settings about the service to enable them to refer patients[10, 40, 41, 43, 45] and encouraging guardians to promote it to other carers was also viewed as important[40, 42, 45]. The necessity of explaining the treatment protocol fully to patients was emphasised, especially during the bracing phase of treatment[14, 40, 41][44][12, 13]. It was also felt this should begin at the treatment outset[14, 43]. Employing designated professionals to do this was viewed as a means of 'freeing up' other professionals responsible for this role and ensuring that guardians received this aspect of care[14]. Leaflets may be used to provide information to take home and refer to should any questions about protocol arise [10, 11, 13, 14, 43]. At the level of public policy, the recognition of the Ponseti method as the gold standard of treatment and the implementation of national clubfoot programmes was seen as a way of standardising treatment processes and facilitating the organisation of nationwide publicity campaigns[12-14, 42, 43].

Discussion

The study has identified and explored a range of factors that impact on presentation and adherence to clubfoot treatment across a range of care settings in LMICs and identified community level

interventions to address them. Results showed that people experienced impacts at the five levels outlined in the Social Ecological Model: the intrapersonal, interpersonal, institutional or organisational, socio-cultural or community levels and to a lesser extent, the level of public policy[24]. At the intrapersonal level, these included access to finance and additional responsibilities within the home. Interpersonal influences were paternal support and support from the extended family and wider community and this was particularly influential in societies where men had more control over decisions and family finances or those where respect for elders was emphasised. Institutional or organisational factors made it difficult for parents or guardians to seek care in all the studies reviewed and included long distances to treatment centres, insufficient information about treatments, knowledge about bracing protocols and challenges with the treatment regime. Socio-cultural or community influences were only identified in half of the studies and included faith in biomedical models of care, alternative belief systems to explain the causes of clubfoot and the stigma associated with the impairment. However, as would be expected when culture and community are seen as complex processes, these were seen to impact on individuals in different ways. Factors at the level of public policy were only identified in Latin America and Brazil where participants felt that the two-tiered healthcare systems presented barriers to treatment seeking. Interventions to address these challenges included counselling sessions, outreach clinics, brace recycling and a range of education programmes.

Strengths and weaknesses

We completed an exhaustive search of the literature using established methods for identifying and synthesising qualitative studies. Although we cannot be sure that we captured all the relevant studies, involvement of members from the wider study team including a clinician experienced in the delivery of clubfoot services, to refine our search terms and identify relevant databases provides us with confidence that this was probably achieved. The studies identified were independently appraised by two members of the research team and data double coded, helping to ensure that quality assessments were consistent and that findings reflect the data under study[46]. To facilitate transparency, the review was presented in accordance with ENTREQ guidelines, a 21 item list to improve the reporting of qualitative syntheses[22].

The review findings were limited on account of studies identified. Using the CASP qualitative appraisal framework[35], three studies were deemed to fully address CASP items, four mainly address CASP items and four partially address CASP items. Studies that partially addressed CASP

items tended to lack clarity about methods of data collection and analysis. Those that fully addressed CASP items included more detail on these processes. As described in the methods section, we classified the quality of each study based on how well or fully it addressed the CASP items. However, the distinction between studies that were considered to ‘mainly address CASP items’ or ‘partially address CASP items’ was based on the quality assessment of two assessors rather than an absolute quality threshold. All included papers contained information about access to clubfoot services that were of relevance to the review and all had value, even where findings could have been presented more clearly or when value was not explicitly stated by the study authors. Some papers contributed more information to themes than others and this is reflected in the findings where these studies are cited more frequently.

Due to the focused nature of the articles they did not include nuanced characterisations of practices and societal norms. Therefore, we acknowledge that the synthesis conveys findings as described in the included studies rather than more detailed information about context. In addition, in three of the studies, patients’ views were identified only by healthcare professionals and there may be discrepancies between clinicians’ views and those of patients. This may account for the emphasis on organisational or institutional barriers rather than interpersonal or socio-cultural factors. The studies identified also tended to focus on barriers to treatment seeking rather than enablers, accounting for the emphasis on barriers in this review. Also, the studies only represent a limited number of LMICs. However, given the homogeneity of experiences, we are reasonably confident that these findings are transferable to other care settings[47].

The Social Ecological Model[24] enabled us to explore the issues under study by highlighting the inter-related factors that impact on treatment-seeking behaviour for the guardians of children with clubfoot. To manage the tension between undertaking an abductive approach[39] whilst ensuring that the data was not ‘forced’ into pre-defined constructs, an inductive thematic analysis[38] was conducted before data were then transposed onto the theory. This meant that any factors that did not ‘fit’ within the model would have been identified. A further theme—potential interventions to enhance care— was identified using the thematic approach.

Further study

Further research is now needed to explore the structure, policy and current implementation of clubfoot treatment services in these settings. Doing so would provide information about how these services can be successfully implemented in practice.

Conclusions

This study has successfully used a meta-synthesis to synthesise findings from existing qualitative research on factors that impact on patient access to clubfoot treatment in LMICs and identified interventions at the community level to address them. Using the Social Ecological Model[24] to inform findings highlights the inter-related factors that influence treatment seeking. These findings may help professionals across a range of LMICs develop services to better meet the needs of patients.

Declaration of interests

SD, CL and RGH have no conflicts of interest to report.

Authors' contributions

SD, CL and RGH all contributed to the analysis, or interpretation of data. All authors contributed to drafting this work and revising it for important intellectual content and all gave final approval for the version to be submitted. RGH is guarantor.

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Table 2: Search syntax used for Ovid MEDLINE, PsycINFO, Embase and Global Health

Search Number	Searches
1	Clubfoot/
2	clubfoot.ti,ab.
3	"club foot".ti,ab.
4	talipe*.ti,ab.
5	equinovarus.ti,ab.
6	1 or 2 or 3 or 4 or 5
7	service*.ti,ab.
8	exp Tenotomy/ or exp Treatment Outcome/ or exp Achilles Tendon/ or exp Orthopedic Procedures/ or exp Manipulation, Orthopedic/
9	Ponseti.ti,ab.
10	surger*.ti,ab.
11	surgic*.ti,ab.
12	clinic*.ti,ab.
13	correction*.ti,ab.
14	treat*.ti,ab.
15	therap*.ti,ab.
16	tenotom*.ti,ab.
17	7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16
18	qualitative research/
19	exp interviews as topic/ or narration/
20	"focus group*".ti,ab.
21	qualitative.ti,ab.
22	ethnograph*.ti,ab.
23	fieldwork.ti,ab.

24	"field work".ti,ab.
25	"key informant".ti,ab.
26	("semi-structured" adj3 interview*).ti,ab.
27	("semi-structured" adj3 discussion*).ti,ab.
28	("semi-structured" adj3 questionnaire*).ti,ab.
29	((semistructured or unstructured) adj3 interview*).ti,ab.
30	((semistructured or unstructured) adj3 discussion*).ti,ab.
31	((semistructured or unstructured) adj3 questionnaire*).ti,ab.
32	((informal or "in-depth" or indepth) adj3 interview*).ti,ab.
33	((informal or "in-depth" or indepth) adj3 discussion*).ti,ab.
34	((informal or "in-depth" or indepth) adj3 questionnaire*).ti,ab.
35	("face-to-face" or structured or guide) adj3 interview*).ti,ab.
36	("face-to-face" or structured or guide) adj3 discussion*).ti,ab.
37	("face-to-face" or structured or guide) adj3 questionnaire*).ti,ab.
38	18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37
39	6 and 17 and 38
40	limit 39 to updatelrange="prmz(20160108120620-20160125151951]"

Table 3: Search syntax used for CINAHL

Search Number	Searches
1	(MH "Equinus Deformity") OR (MH "Clubfoot")
2	TI clubfoot OR AB clubfoot
3	TI talipe* OR AB talipe*
4	TI equinovarus OR AB equinovarus
5	S1 OR S2 OR S3 OR S4
6	TI service* OR AB service*
7	TI ponseti OR AB ponseti
8	TI (surger* OR surgic*) OR AB (surger* OR surgic*)
9	TI clinic* OR AB clinic*
10	TI correction OR AB correction
11	TI treatment OR AB treatment
12	TI therapy OR AB therapy
13	TI tenotomy OR AB tenotomy
14	S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13
15	S5 AND S14
16	TI (("semi-structured" or semistructured or unstructured or informal or "in-depth" or indepth or "face-to-face" or structured or guide) adj3 (interview* or discussion* or questionnaire*)) or (focus group* or qualitative or ethnograph* or fieldwork or "field work" or "key informant") or interviews as topic/ or focus groups/ or narration/ or qualitative research/) OR AB (("semi-structured" or semistructured or unstructured or informal or "in-depth" or indepth or "face-to-face" or structured or guide) adj3 (interview* or discussion* or questionnaire*)) or (focus group* or qualitative or ethnograph* or fieldwork or "field work" or "key informant") or interviews as topic/ or focus groups/ or narration/ or qualitative research/)
17	S5 AND S14 AND S16

Figure 2: PRISMA flow chart detailing process of identifying studies relevant for inclusion

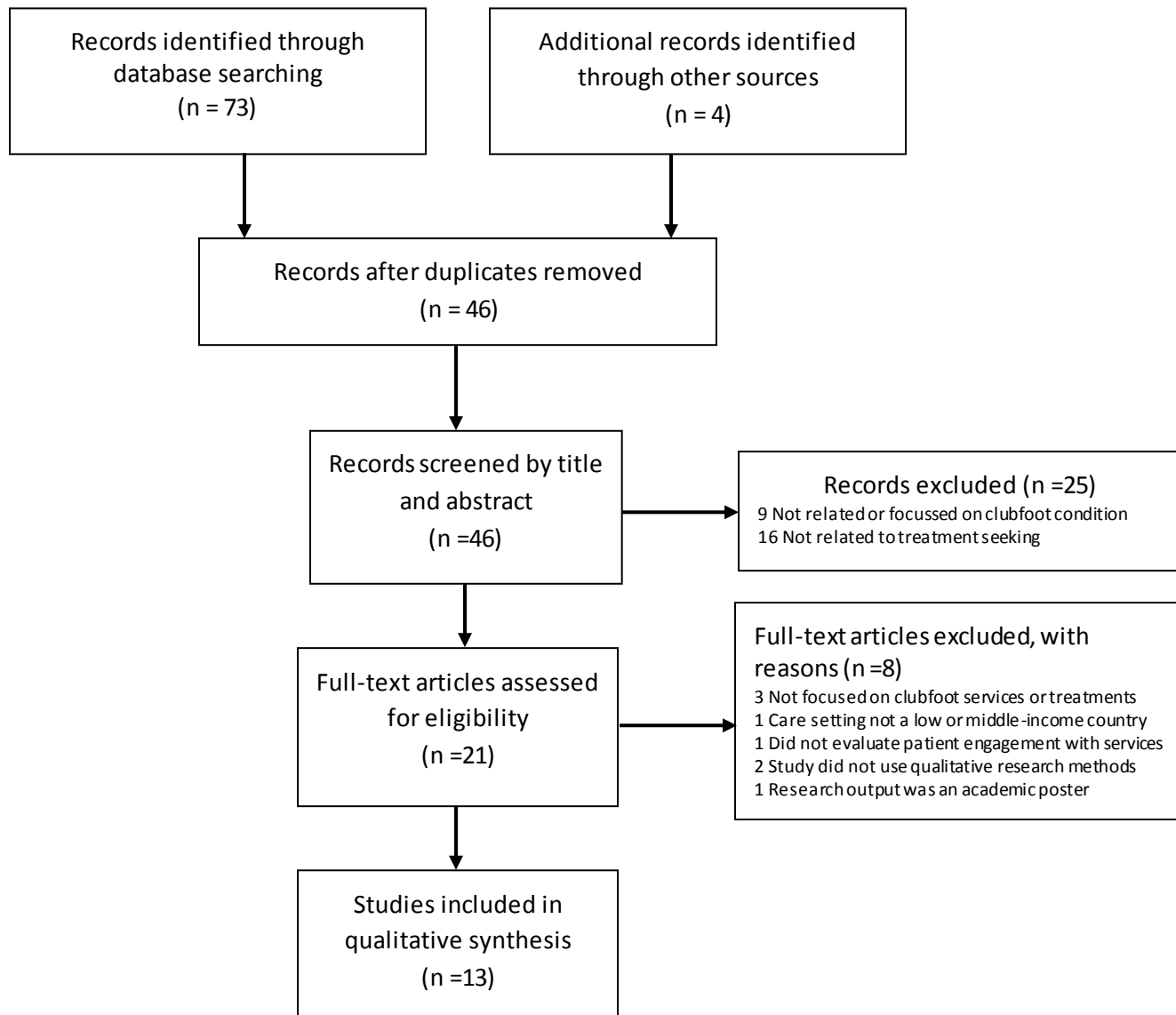


Table 4: Summary of the characteristics of papers included in the review

First author, year	Country	Participants	Methods	Aim	Characteristics in relation to three CASP domains	Extent it addresses CASP items
Aktintayo, O. A., 2012[43]	Nigeria	25 physicians practising Ponseti method, 6 newly trained practitioners, 42 parents of children with clubfoot	Semi-structured interviews, focus groups [As part of a mixed methods study]	To evaluate the initial impact of the Ponseti method and identify challenges to its diffusion and implementation.	<p>1) Aims and appropriateness Clear statement of the aims of the research.</p> <p>2) Design and conduct Range of research methods used. However, it was unclear what each method contributed to the study. No discussion of ethical considerations. Although data was sorted into themes, there was no description on how this process was undertaken.</p> <p>3) Clarity and value Findings were grouped into dominant themes that made them reasonably clear to understand. There was no evaluation of the strengths and weaknesses of the study. Value of research was emphasised with clear discussion of how findings may be applied to help develop services.</p>	Partially
Bedford, J., 2011[21]	Malawi	60 case studies with parents of children with clubfoot	Interviews, photographs, observation	To explore the treatment-seeking behaviour of guardians of patients undergoing treatment for clubfoot using the Ponseti method.	<p>1) Aims and appropriateness Clear statement of the aims of the research.</p> <p>2) Design and conduct Setting for data collection justified and methods clearly described.</p>	Fully

					<p>Ethical approval received from review board and informed consent sought. The process of thematic analysis undertaken is described in detail.</p> <p>3) Clarity and value Clear statement of research findings. Value of the research is emphasised, including discussion of how findings are being used in practice. Future areas of research identified.</p>	
Bedford, J. 2009[40]	Malawi	60 case studies with parents of children with clubfoot	Interviews, photographs, observation	<p>To explore perspectives on clubfoot and its treatment using the Ponseti method.</p> <p>To make recommendations to improve and develop services.</p>	Quality appraisal same as for Bedford 2011.	Fully
Boardman, A., 2011[13]	Chile, Peru, Guatemala	30 physicians practising the Ponseti method	Semi-structured interviews	To evaluate the impact and barriers to the diffusion of the Ponseti method.	<p>1) Aims and appropriateness Clear statement of the aims of the research.</p> <p>2) Design and conduct Semi-structured interviews appropriate for addressing research aims. Ethical issues have been taken into consideration, including details about the relevant ethical review board, process of obtaining informed consent and data storage.</p> <p>3) Clarity and value Presentation of findings slightly unclear as section seems to include</p>	Mainly

					author interpretations alongside findings. It is unclear what represents primary data. Findings are considered in relation to existing literature. The value of the research and its potential to improve patient care are briefly discussed.	
Gadhok, K., 2012[12]	India	15 orthopaedic surgeons practising Ponseti method, 15 parents of children receiving treatment with Ponseti method	Semi-structured interviews [As part of a mixed methods study]	To identify the challenges to the diffusion and implementation of the Ponseti method	<p>1) Aims and appropriateness Clear statement on the aims of the research.</p> <p>2) Design and conduct Study uses a range of research methods. However, it is not clear why these methods were selected or what each contributed to the study. Researcher describes how healthcare professionals have been sampled but not the patient population. The setting for data collection has been justified. Ethical issues have been taken into consideration including information on ethical review board and data storage and consent. There is very little description on the process of analysis.</p> <p>3) Clarity and value There is a clear statement of findings. Written descriptions are grouped into dominant themes and a table is included that distills the themes identified in the study. A discussion of strengths and weakness is included. However, there is limited discussion on how work contributes</p>	Mainly

					to the understanding of the topic and may be applied in practice.	
Jayawardena, A., 2013[45]	Sri Lanka	162 patients and healthcare practitioners involved with clubfoot care	Interviews, focus groups, observation	To evaluate the utility of a 'train the trainer' approach for disseminating the Ponseti method.	<p>1) Aims and appropriateness Clear statement of study aims.</p> <p>2) Design and conduct Research methods appropriate for addressing research aims and value of using multiple methods discussed. No discussion on why participants chosen to address study aims. Setting for data collection not justified. Methodology used is clearly stated and detail included on how interviews and focus groups carried out. Ethical considerations discussed including anonymisation and data storage. However, no information provided on why study granted exemption from ethical review. Lack of information on process of data analysis.</p> <p>3) Clarity and value Findings clear and organised into key themes. Strengths and weaknesses of study discussed. Contribution study makes to existing knowledge discussed. Areas of future research identified.</p>	Mainly
Kingau, N. W., 2015[44]	Kenya	10 service providers delivering clubfoot treatment, 10 caregivers of children with clubfoot.	Semi-structured interviews	To identify barriers experienced by service providers and carers to the effective management of clubfoot using the Ponseti method and surgical	<p>1) Aims and appropriateness Clear statement of study aims.</p> <p>2) Design and conduct Semi-structured interviews appropriate to address study aims.</p>	Fully

				interventions.	<p>Explanation of how participants were sampled and why they were most appropriate to address research aims. Detailed description on how interviews were carried out and data saturation discussed. No discussion of ethical considerations. Data analysis rigorous and detailed description of how process was conducted.</p> <p>3) Clarity and value Primary data included to support findings. Strengths and weaknesses of study discussed. Findings considered in relation to existing research and how they may contribute to service development.</p>	
McElroy, T., 2007[15]	Uganda	42 parents of children with clubfoot, 2 adults with clubfoot, 40 community leaders, 39 traditional healers, 38 practitioners treating clubfoot	Semi-structured interviews, focus groups, observation	To identify barriers to adherence to the Ponseti method of clubfoot treatment.	<p>1) Aims and appropriateness Clear statement of study aims.</p> <p>2) Design and conduct Research design appropriate to address aims and discussion of value of using multiple methods included. Information and justification of sampling strategy and of research setting. Detailed description of process of data collection. Ethical issues taken into consideration including review board, process of obtaining informed consent from participants speaking a range of languages, anonymisation of data and storage. In-depth description of analysis process.</p>	Fully

					3) Clarity and value Findings made clear and structured around dominant themes. Primary data included to support findings. No discussion of strengths and weaknesses of study. Discussion of how findings can be used in practice and areas of future research identified.	
Konde-Lule, J. 2005[41]	Uganda	42 parents of children with clubfoot, 2 adults with clubfoot, 40 community leaders, 39 traditional healers, 38 practitioners treating clubfoot	Semi-structured interviews, focus groups, observation	To explore knowledge, attitudes, beliefs, and practices about clubfoot across different regions in Uganda.	Quality appraisal same as for McElroy 2007.	Fully
Lu, N., 2010[11]	China	39 physicians practising the Ponseti method, 8 sets of parents of children receiving treatment with Ponseti	Semi-structured interviews, focus groups	To evaluate and identify barriers to the implementation of the Ponseti clubfoot program.	1) Aims and appropriateness Clear statement of study aims. 2) Design and conduct Research design appropriate and value of multiple methods explained. Explanation of why participations selected included. Research settings not justified. Fairly clear how data were collected. Ethical issues taken into consideration including ethical review board, informed consent, anonymisation of data and data storage. Process of data analysis described in detail. Codes discussed with wider study team to help validate findings. 3) Clarity and value Study findings clear. No discussion of	Mainly

					strengths and weaknesses of study. Implications of research for improving service design discussed and author makes series of recommendations for service development based on findings.	
Nogueira, M., 2013[14]	Brazil	45 orthopaedists trained in the Ponseti method	Semi-structured interviews	To identify barriers to bracing compliance.	<p>1) Aims and appropriateness Clear statement of study aims.</p> <p>2) Design and conduct No information on how participants sampled or why they were appropriate for addressing study aims. Setting for data collection not justified. Unclear how semi-structured interviews conducted. No information on how analysis undertaken.</p> <p>3) Clarity and value Clear statement of findings and weaknesses of study discussed. No clear statement on value of research but strategies suggested for addressing challenges identified.</p>	Partially
Palma, M., 2013[42]	Peru	25 physicians practising the Ponseti method	Semi-structured interviews	To evaluate how barriers to using the Ponseti method have changed in 2 years and to identify barriers to its diffusion.	<p>1) Aims and appropriateness Clear statement of study aims in the main body of the text although these are unclear in the abstract.</p> <p>2) Design and conduct Semi-structured interviews appropriate for addressing study aims. However, it is unclear how participants were sampled and setting for data collection not</p>	Partially

					<p>justified. Ethical issues taken into consideration including details of review board, anonymisation of data and data storage. Although thematic analysis undertaken, limited information on how this was conducted.</p> <p>3) Clarity and value Findings clear and grouped into dominant themes. No discussion on strengths and weaknesses of the study. Potential of research to improve service design discussed.</p>	
Wu, V., 2012[10]	Vietnam	12 practitioners practising Ponseti (physicians, nurses, physical therapists and case technicians), 99 parents of children with clubfoot and extended family	<p>Semi-structured interviews, focus groups, observation</p> <p>[As part of a mixed methods study]</p>	To evaluate the impact, progress and challenges facing Ponseti practitioners and patients' family.	<p>1) Aims and appropriateness Clear statement of research aims.</p> <p>2) Design and conduct Research design appropriate to address study aims. Value of using multiple methods discussed. Description of sampling strategy but no discussion of why these participants were most appropriate for addressing study aims. Setting for data collection not justified. No detail on how data collected and the contribution of each method to study findings is unclear. No discussion of ethical considerations. No information on process of analysis.</p> <p>3) Clarity and value Clear statement of findings and discussion of strengths and</p>	Partially

					weaknesses of the study included.. Implications of research for improving service design discussed and number of recommendations made based on findings.	
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