Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

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Thesis submitted in accordance with the requirements for the degree of Doctor of Philosophy

University of London

August 2018

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LONDON SCHOOL OF HYGIENE & TROPICAL MEDICINE

Funded by the Beit Trust, CBM, MiracleFeet, ZANE

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Declaration

I, Tracey Heather Smythe, confirm that the work presented in this thesis is my own. Where data and information have been derived from other sources. I confirm that these have been appropriately indicated in the thesis.

Signature: [redacted]

Date: 12/09/2018

Abstract

Background

Clubfoot is one of the most common congenital musculoskeletal birth defects. Untreated it leads to physical impairment and deformity, resulting in loss of mobility and function. The cause in most cases is unknown.

With early diagnosis and appropriate treatment functional impairment from clubfoot is avoidable. A minimally invasive technique as described in the Ponseti method is recommended for the treatment of the clubfoot deformity. In some resource-constrained settings, this treatment is being provided by non-specialised health workers, 'clubfoot therapists', trained to treat children with clubfoot.

This PhD thesis seeks to provide evidence to improve services for children with clubfoot in Africa using data from Zimbabwe as a case study.

Methods

Two systematic reviews were undertaken; first to investigate the birth prevalence of clubfoot in low-and middle-income countries, and second to determine and evaluate how results of clubfoot management in sub-Saharan Africa are reported.

A Delphi process with 35 experts (Ponseti technique trainers) from across Africa was used to determine (a) the criteria to assess clubfoot treatment and (b) to identify the indicators to evaluate the functionality of clubfoot clinics.

In a retrospective case series of 218 children with idiopathic clubfoot in Harare, Zimbabwe, the results of corrective treatment and the factors that affect outcome were analysed.

Using the results of the first Delphi exercise, a tool (the Assessing Clubfoot Treatment (ACT) score) was developed for clubfoot therapists to assess the results of Ponseti treatment in children of walking age in low resource settings. The tool was evaluated prospectively using the cohort from Harare, and also compared with other existing assessment methods.

From the second Delphi study - to obtain a consensus definition on indicators to assess the functionality of a clubfoot clinic in Africa - a questionnaire was developed (Functionality Assessment clubfoot Clinic Tool, FACT) and piloted in a cross-sectional study of service provision in 12 clubfoot clinics in Zimbabwe.

A prospective mixed methods (both quantitative and qualitative) evaluation was used to assess the feasibility of a training programme (delivered through the Africa Clubfoot Training project from 2015 - 2017) for clubfoot therapists in Africa.

Results

There is similarity of pooled estimates of birth prevalence of clubfoot in Africa, Eastern Mediterranean region, India and South East Asia (between 1.11 (95%CI 0.96 – 1.26)/1,000 live births and 1.21 (95%CI 0.73 -1.68)/1,000 live births).

Of 22 studies that report results of the Ponseti method in sub-Saharan Africa only 14 (64%) described a primary outcome. Clinical assessment was the most commonly reported outcome measure and the Pirani score was the most frequent tool used to assess clubfoot severity. Results were predominantly reported though case series.

The case series from Parirenyatwa Hospital demonstrated that the Ponseti method was successful in the majority (85%) of feet (defined as a Pirani score of 1 or less) up to completion of the corrective phase, with a relatively low loss to follow-up (8.9%).

The ACT score, which was developed as a result of the first Delphi exercise, included one simple clinical assessment and three parent reported outcomes. In the children who were followed up (n=68) in the cohort from Harare, 72% (49/68) achieved an acceptable outcome (defined as an ACT score of 9 or more). The ‘success’ of treatment defined by five different assessment tools varied between 56% and 93% in the cohort.

Using the FACT score, developed from the second Delphi exercise, the most common needs identified in the 12 clubfoot clinics in Zimbabwe were (a) a

standard treatment protocol, (b) a process for surgical referrals, and (c) a process to monitor drop out of patients.

Fifty-one regional trainers from 18 countries in Africa were trained over the two years of the Africa Clubfoot Training project. These regional trainers delivered the basic and advanced course to 113 participants in 3 countries (Ethiopia, Rwanda and Kenya). The mean participant confidence and the mean participant knowledge both increased substantially following the training. Participants expressed high acceptability of the training, which they attributed to its clear purpose and guidance, and the interpersonal interaction with the trainers.

Conclusion

Clubfoot services can be improved in Zimbabwe and probably the wider Africa region. It requires a health system-oriented approach. The evidence presented indicates that children with clubfoot can be effectively treated by trained clubfoot therapists (using the Ponseti method). To enable this there is a need to ensure that clubfoot clinics are appropriately equipped and clubfoot therapists are appropriately trained. Two tools have been developed to assist clubfoot therapists monitor their results (ACT score) and to enable programme managers to monitor the national clubfoot service provision (FACT). Studies to refine and test the ACT and FACT scores in other settings in Africa are required.

Format of the Thesis

The thesis for this PhD is presented in the 'research paper style' format, in accordance with the London School of Hygiene & Tropical Medicine research degree regulations. It includes a number of different but related journal articles that have been published in, accepted by, or submitted to peer-reviewed journals.

The chapters listed in italics in the Table of Contents are in the research paper format and include publication details in a cover sheet, including acknowledgement of the contributions of other people. The other chapters of the thesis are composed of 'linking material', which includes information or data not covered in the papers and makes the thesis a coherent body of work. I, Tracey H. Smythe, wrote the linking material.

This thesis is divided into four sections (A to D) with each section sub-divided into chapters.

Section A includes the introductory chapter on clubfoot epidemiology and clinical care (Chapter 1), and Chapter 2 on the study rationale, aim, hypothesis and research questions. Chapter 3 provides background information on the Zimbabwe Sustainable Clubfoot Programme and the Africa Clubfoot Training project.

Section B comprises nine research papers (Chapters 4 – 12), which describe the results of the PhD research study. Each chapter is introduced with a preamble that includes information on the rationale for the research. An epilogue follows the research paper and the associated appendices, and summarises the main findings and limitations of the study. The epilogue also includes supplementary material under the sub-title of 'additional information' that clarifies data published in the paper.

Chapters 4 and 5 comprise systematic reviews on birth prevalence of clubfoot in low- and middle-income countries, and the assessment of clubfoot treatment in sub-Saharan Africa, respectively.

Chapters 6 to 9 present the short and long-term results of clubfoot treatment, as described in the Ponseti method, in a cohort of children in one referral hospital in Zimbabwe. Chapter 6 reports the short-term results of clubfoot treatment in a case series. Chapter 7 proposes a tool for use by clubfoot therapists to assess success of treatment in children of walking age. The tool is developed and used to evaluate the cohort, and also compared to full clinical assessment in Chapter 8. Chapter 9 presents the long-term results of clubfoot treatment in the cohort and evaluates five tools for use by clubfoot therapists to assess success of treatment in children of walking age.

Chapter 10 outlines the indicators to assess clubfoot clinic functionality, and the extent to which these components are provided in Zimbabwe.

Chapters 11 and 12 present the formative research and short-term outcomes for the feasibility of a training programme for clubfoot therapists in Africa.

Section C is a general discussion of the findings from this PhD research study. It includes the implications of the findings of this body of work for future research, policy and clubfoot programmes. Chapter 13 includes an analysis of clubfoot service pathways to discern achievements and identify barriers to improving clubfoot services, using evidence identified in the previous chapters. A health system-oriented approach is used to make recommendations to improve clubfoot services in Zimbabwe. The strengths and limitations of this study are described in Chapter 14. Chapter 15 concludes the thesis with a summary of the findings, and implications for future research, policy and clubfoot programmes are considered.

Section D includes the appendices.

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Unless stated otherwise, all photographs included in this PhD thesis were taken by myself with permission from the participant

List of Abbreviations

ACT project: Africa Clubfoot Training project

ACT score: Assessing Clubfoot Treatment score

CI: confidence interval

CER: cost effectiveness ratio

CFT: clubfoot therapists

CTEV: congenital talipes equinovarus

DALY: disability adjusted life year

DOB: date of birth

FAB: foot abduction brace

FACT: Functionality Assessment clubfoot Clinic Tool

GCI: Global Clubfoot Initiative

GDP: Gross Domestic Product

HIV: human immunodeficiency virus

ICC: intra-class correlation coefficient

IMCI: Integrated management of childhood illness

JREC: Joint Research Ethics Committee for the University of Zimbabwe, College of Health Sciences and Parirenyatwa Group of Hospitals

LMIC: low- and middle-income countries

LSHTM: London School of Hygiene & Tropical Medicine

MoHCC: Ministry of Health and Child Care

MOOSE: meta-analysis of observational studies in epidemiology

MRCZ: Medical Research Council of Zimbabwe

NGO: Non-governmental organisation

PPP: purchasing power parity

PRISMA: preferred reporting items for systematic reviews and meta-analyses

RCT: randomised control trial

SD: standard deviation

THET: Tropical Health Education Trust

USD: United States Dollar

VAS: visual analogue scale

WHO: World Health Organisation

ZSCP: Zimbabwe Sustainable Clubfoot Programme

Acknowledgements

The contributions and support of many people have made this work possible.

I am grateful to my supervisors, Professor Allen Foster and Professor Chris Lavy; my work is embedded with lessons from your wisdom and integrity. Your constant encouragement and guidance has developed my penchant for problem solving. Allen, you have guided and honed my ability to wordsmith. Over these years I have experienced that ‘it will sort’, I am grateful for the opportunity. Chris, I owe this brilliant turn in my life to you, I am thankful for your unwavering support. You taught me to pause, and the value of hearing the quiet voices. I am so pleased to be able to continue to work alongside you both.

Professor Daniel Chandramohan, and in memory Jeroen Ensink. You encouraged my leap into further study and your voices are a reminder of perspective. Jeroen, I hope in some small way this work contributes to your legacy.

Thank you to my advisory committee. Professor Hannah Kuper, for your insightful feedback, and continued support. Professor Jayne Webster, for providing invaluable advice, and for shared laughter.

The ICED team. Thank you for your generosity of friendship. You provide an enriching and challenging environment in which to work. Jyoti Shah, project administrator extraordinaire, I appreciate your patience and willingness to help with small details that go unseen.

I thank the Beit Trust for funding this research study. Your generous financial support of my PhD studentship and project costs has made this work possible.

I thank CBM, MiracleFeet and ZANE for funding project costs.

Thank you to the children and caregivers in this research, for sharing a small part of your lives with me. I hope that this work can further your voices being heard.

I am deeply grateful to my family. You have given me the most. You encourage me to be curious and to question. Thank you for keeping me grounded whilst encouraging me to leap. I could not have accomplished this work without your unwavering support.

This work is dedicated to Mackenzie. May your sensitive and kind spirit discern how you can care for those less able to help themselves. Continue to shine my love.

Contributors to the research presented in this thesis

Contributors to the research project besides listed authors in the manuscripts

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SECTION A INTRODUCTION

Chapter 1. Clubfoot epidemiology and clinical care

Child with bilateral clubfoot

1.1 Definition of clubfoot

Birth defects, or congenital anomalies, are a leading cause of mobility impairment in children (1). Congenital talipes equinovarus (CTEV), known as clubfoot, is one of the most common musculoskeletal birth defects. The structural development of the bones and muscles in the foot is affected and the clubfoot is fixed in a rigid downward and inward position (Figure 1).

Figure 1 Child with bilateral clubfoot
(Credit: Marieke Driese)

Specifically, the clubfoot deformity is characterised by four components:

- cavus of the midfoot (a high arch in the sole of the foot);
- adductus of the forefoot (the front of the foot is positioned toward the opposite leg);
- varus of the hindfoot (the heel is fixed inward); and
- equinus of the hindfoot (the foot points downward) (Figure 2).

There is also an associated decrease in size of the calf muscle.

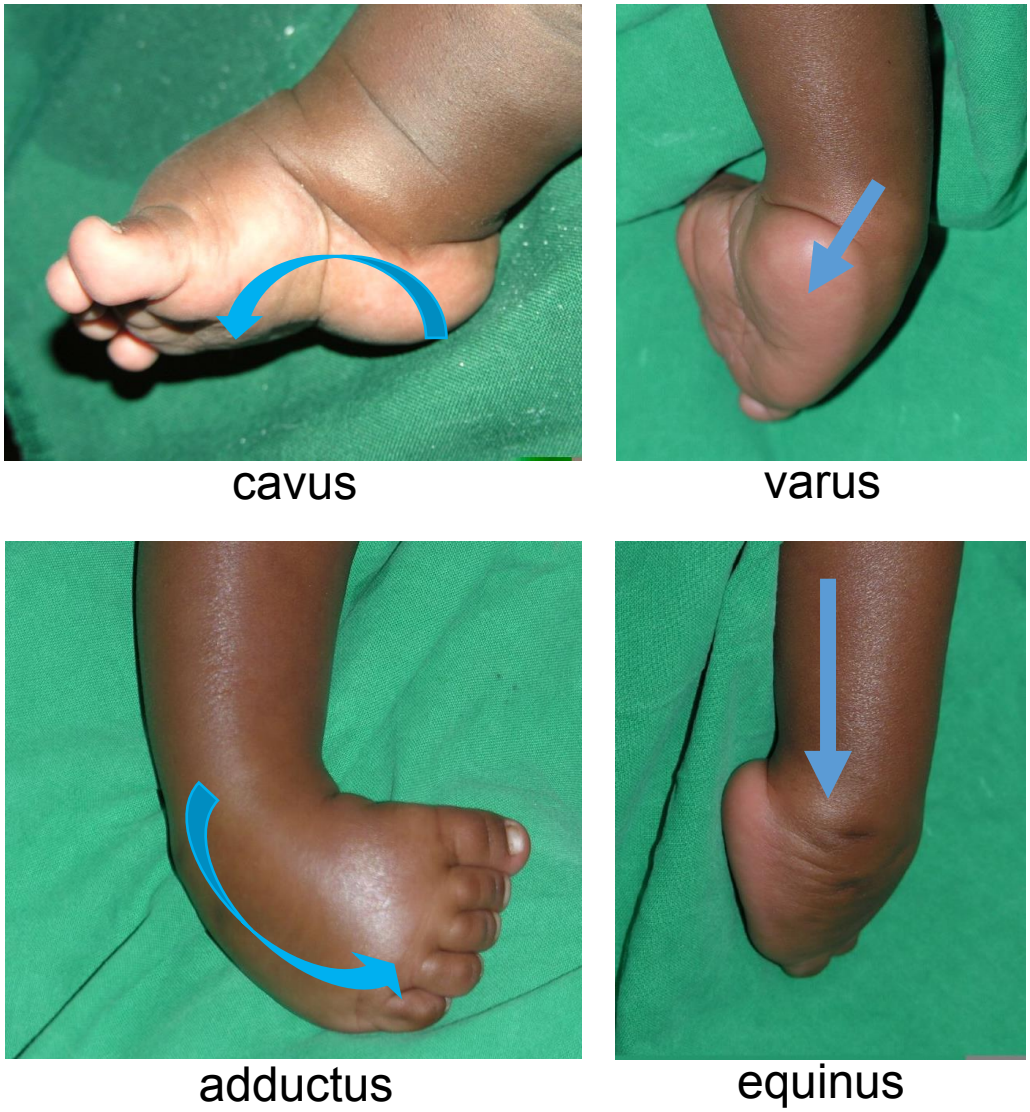


Figure 2 Three-dimensional deformity of a clubfoot
(Credit: Lynn Staheli)

1.2 Birth prevalence

A limited group of statistics is commonly reported for the birth prevalence of clubfoot. The estimates vary with ethnicity, from 0.39/1,000 live births in Chinese populations to 6.8/1,000 live births in Polynesian populations (2) and 1/1,000 live births in Caucasian populations (3). It is estimated that 80% of children born with clubfoot each year live in low- and middle-income countries (LMIC) (4) due to the increased birth rate in these countries.

The birth prevalence of clubfoot and the population birth rate allow estimates of the number of children born with clubfoot per year per million total population. This estimate can be used for planning purposes. For example, a birth rate of 10 live births/1,000 population per year extrapolates to 10,000 live births/million

population/year. When the birth prevalence of 1/1,000 cases of clubfoot is applied to this population, this gives a figure of 10 clubfoot cases per million total population.

Data for a population of one million people are presented in Table 1. This information demonstrates how the estimate of the number of children born with clubfoot per million population will vary with birth rate, and with two hypothesised clubfoot birth prevalence rates, 1/1,000 and 6/1,000 live births respectively.

Table 1 Estimated clubfoot cases/million population

Birth rate/1,000	Projected births/mill pop	Clubfoot cases/mill pop for CFBP of 1/1,000	Clubfoot cases/mill pop for CFBP of 6/1,000
10	10,000	10	60
20	20,000	20	120
30	30,000	30	180
40	40,000	40	240

Legend: mill = million, pop = population. CFBP = clubfoot birth prevalence

Applying clubfoot birth prevalence rates of 1/1,000 and 6/1,000 live births to the population of Zimbabwe (16.15 million), with a crude birth rate of 33 live births/1,000 population (5), estimates 532 to 3,198 children born with clubfoot per year.

1.3 Aetiology and risk factors

Most cases of clubfoot occur as an isolated birth defect and are known as ‘idiopathic’ because the cause is not known. The remaining 20% of cases are associated with other structural conditions such as arthrogryposis, syndromes and disorders of the nervous system, for example spina bifida (6).

Many theories for the cause of idiopathic clubfoot have been proposed. It is likely that genetic and environmental factors are responsible (6). Transcriptional pathways on genes have been explored (6, 7), however the exact mechanisms by which genetic abnormalities cause clubfoot remain unknown. Male sex is consistently associated with an increased risk of clubfoot (8-11); clubfoot affects twice as many boys as girls.

Studies of environmental factors are inconsistent and hypotheses include seasonal variation (12) and intrauterine immobility (13). Associations with ethnicity are not clear. Other risk factors that have been reported are maternal smoking (9-11, 14-16), maternal diabetes (9, 14) and early amniocentesis (17). The underlying pathogenesis for these factors remains uncertain.

Idiopathic clubfoot is bilateral in half of the cases (18).

1.4 Early diagnosis and screening

The relationship between the foot and leg develops by the eleventh week of pregnancy and ultrasound screening can detect the presence of clubfoot after this period (19). Diagnosis at this stage is dependent on the skills of the examiner, the quality of the equipment and the methodology used (20), with a false positive rate of approximately 1 in 5 (18%) (21). In settings where ultrasound is not routinely available, clubfoot can be recognisable at birth due to the distinct fixed structure of the foot.

1.5 Assessment and classification of clubfoot deformity

The severity of clubfoot can vary from mild deformity to a rigid foot that is resistant to manipulation or movement (22). The most widely used severity classifications are the Pirani (23) and Dimeglio (24) scores, which are based on physical assessment of the foot.

The Pirani Score

The Pirani score grades six components of the deformity, three components in the midfoot and three in the hindfoot. Each component may score 0, 0.5 or 1. A total score of 6 is the most severe deformity and 0 is a normal foot. The score of the six components of the deformity is determined with either observation, measurement or palpation, and the foot can be assessed within one minute. This scoring system uses different views of the foot to assess the deformity and no technical equipment is required.

The midfoot components that are scored with the Pirani score are the medial crease, the curvature of the lateral border and the lateral head of talus (Figure 3).

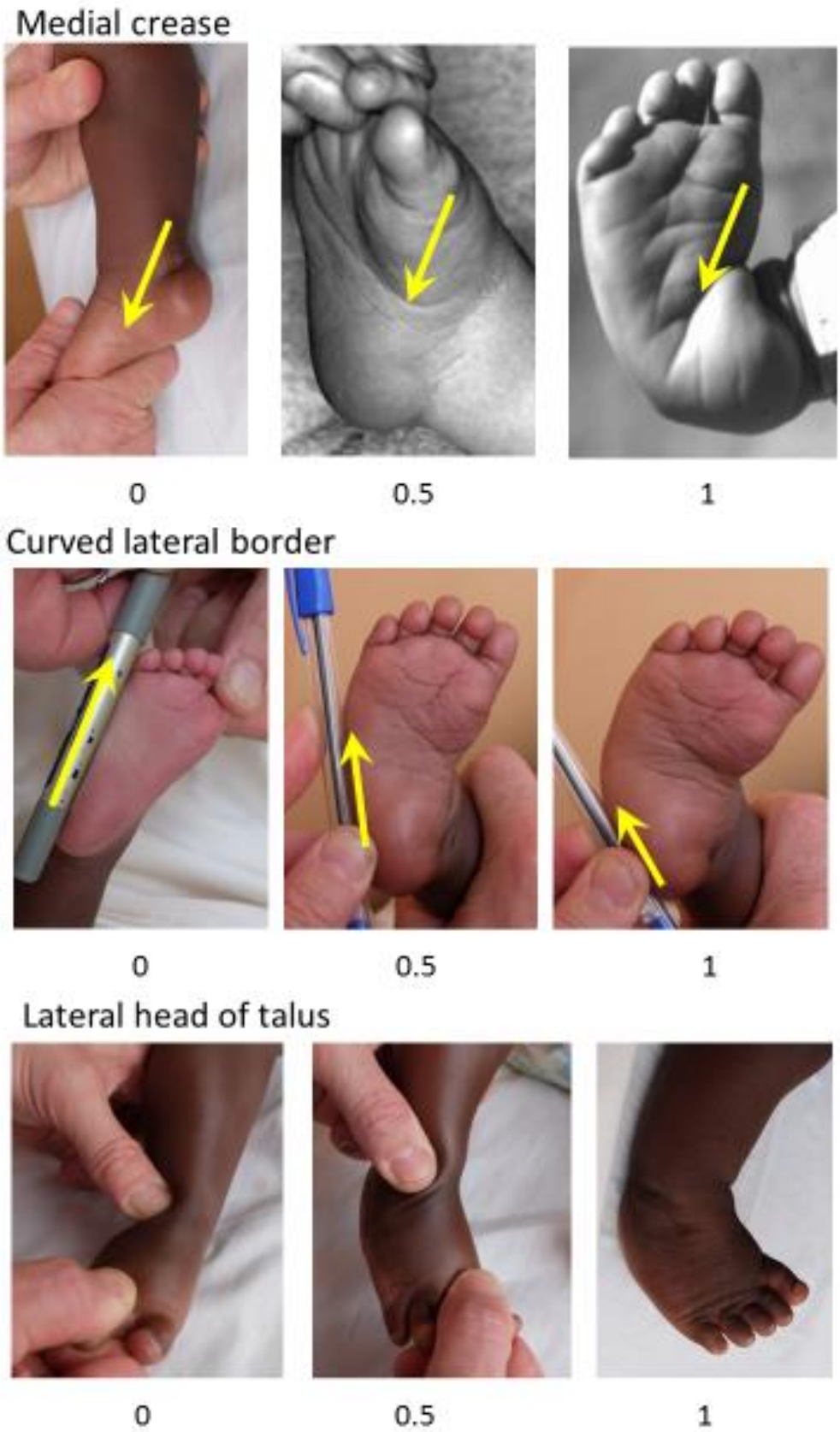


Figure 3 The Pirani score: midfoot assessment

(Credit: ACT training)

The hindfoot components that are scored are the rigid equinus, posterior crease and empty heel (Figure 4).

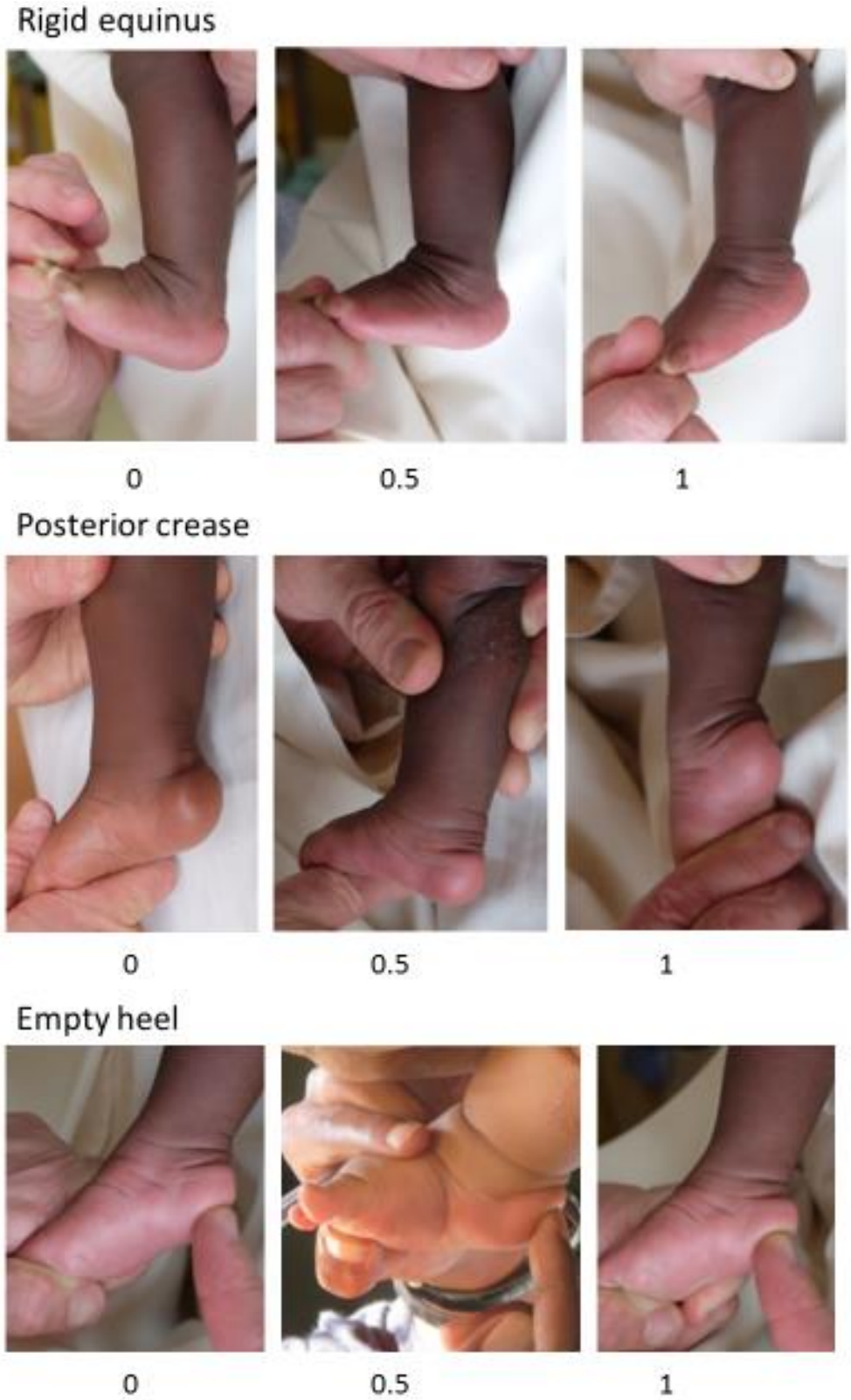


Figure 4 The Pirani score: hindfoot assessment

(Credit: ACT training)

The Dimeglio score

The Dimeglio score has a maximum of 20 points and includes eight assessments, one of which is muscle function. The method of assessment is based on a checklist and diagrams (Figure 5) (25) and training material includes an audio-visual package. It requires a specialised instrument for the precise measurement of angles, known as a goniometer.

Classification			Assessment of Clubfoot by Severity Scale			
Classification grade	*Type*	*Score*	*Characteristics: Deformity*	*Points (pts)*	*Characteristics: Other parameters*	*Points (pts)*
I	Benign	(<5)	90-45°	4	Posterior crease	1
II	Moderate	(=5<10)	45-20°	3	Medial crease	1
III	Severe	(=10<15)	20-0°	2	Cavus	1
IV	Very severe	(=15<20)	<20 to –20°	1	Poor muscle condition	1

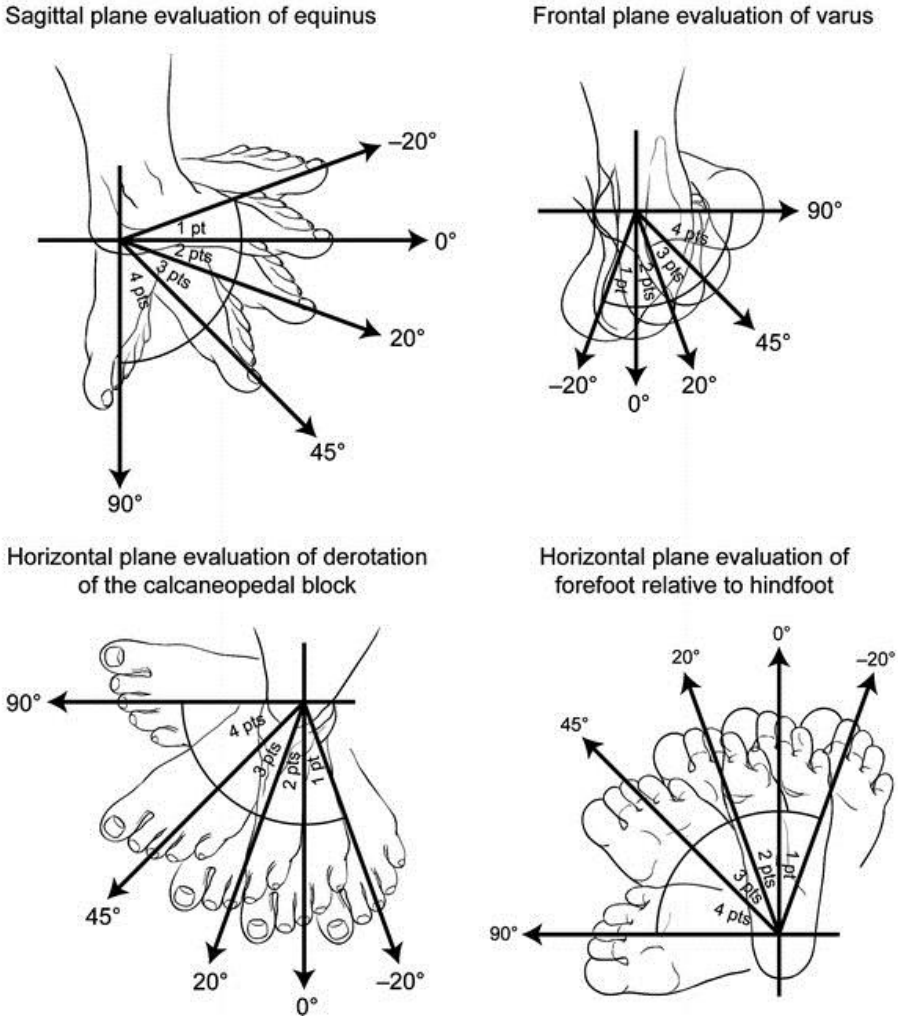


Figure 5 The Dimeglio score

(Credit: Benson M, Fixsen J, Macnicol M, Parsch K. Children's orthopaedics and fractures: Third Edition. 2010.)

The angle of deformity is assessed in four planes of movement. These components are scored 1 – 4, with 4 the most severe. The presence of four additional elements are observed and scored as 1 (or 0 for absence). The elements are: posterior crease, medial crease, cavus and poor muscle function. The higher the score, the more severe the deformity and the total score is graded as benign, moderate, severe or very severe.

Measurement of recurrence

The clubfoot deformity has a strong tendency to recur after corrective treatment. This is because the factors that initiate the deformity remain active as the child grows (26). Recurrence of the deformity is less common after the child is four years old as growth of the foot decreases in speed. There is no agreed assessment of severity of recurrence and elements of the deformity that recur are typically noted under clinical examination and observation of function.

1.6 Clinical management of clubfoot

Surgical management

The approach to management of clubfoot has changed over the years as understanding of the deformity has improved. Extensive open surgery to obtain a foot with a 'perfect' shape began in the late 1800s (27). Case series published between 1971 and 1985 estimate a 25% failure rate in extensive open clubfoot surgery (28). Complications included wound scarring, foot and leg weakness, pain in the foot and loss of normal ankle motion with age.

Current indications for surgery include when non-operative treatment reaches a plateau and elements of the clubfoot deformity remain (29), in the treatment of residual muscle imbalance (for example, a tendon transfer) (30), and in cast resistant feet of older children with clubfoot (31).

Conservative management

A second approach to clubfoot management is conservative treatment. Conservative techniques achieve correction of the clubfoot deformity gradually, by slowly stretching tight structures through manipulation and then fixation, either with casting materials or strapping. This allows time for soft tissue to remodel and for the position of the bones in the foot to re-align. Treatment

ideally begins soon after birth. Conservative treatment developed by Dr J.H. Kite in the 1930s involved manipulation, where each component of the deformity was corrected separately instead of uniformly, and the corrected position was then held with plaster casts (32). The process takes many cast changes over months as the method of manipulation of the foot prevents the hindfoot deformity from resolving, and the below knee casts are unable to maintain the position of the foot adequately (33).

Another conservative technique involves daily manipulation and stretching of the foot, stimulation of underactive muscles and taping (34), and is known as the ‘French‘ or ‘functional physical therapy’ method. It is human resource intensive and relies on daily dedication of the parents over months.

1.7 The Ponseti method of conservative clubfoot management

In the late 1940s, after extensive anatomical studies, Dr I.V. Ponseti of the University of Iowa devised a minimally invasive technique that included a correction and a maintenance phase. The technique is started as soon as possible after birth. The aim of clubfoot treatment as recommended by Ponseti is to provide children with

> *“a functional, pain-free, normal-looking foot, with good mobility, without calluses, and requiring no special shoes.” (27)*

The corrective phase of treatment involves the simultaneous correction of three components of the clubfoot deformity, with the equinus (downward pointing of the foot) corrected last. The manipulated foot is held in a series of long leg (toe to groin) plaster of paris (POP) casts, with the knee at 90 degrees. Two people are required to perform the treatment, one to manipulate the foot and maintain the corrected position, and the other to apply the POP. The casts retain the degree of correction and allow the soft tissue time to remodel. The corrective phase usually takes 4-8 weeks and the baby/infant is seen weekly for treatment (Figure 6).

Figure 6 Positions of the foot and cast after weekly manipulations
(Credit: Lynn Staheli)

An outpatient procedure to cut the Achilles tendon (heel tendon), known as a tenotomy, is usually needed to correct the downward position of the foot. An Achilles tenotomy is performed when the cavus, adductus and varus have been corrected, but the rigid equinus remains. The procedure can be undertaken in the outpatient clinic under local anaesthetic (35) and can be performed by a trained medical officer or orthopaedic clinical assistant. The decision of when to refer for tenotomy typically rests with the treating clubfoot therapist. The picture on the right side of Figure 6 demonstrates the foot position after a tenotomy. The final cast remains for 3 weeks to allow the Achilles tendon to re-grow in this lengthened position.

The corrected foot is then maintained in a foot abduction brace (FAB) immediately after the removal of the final cast (Figure 7), with the aim to prevent recurrence of the deformity. The wear schedule of the FAB is recommended as 23 hours a day for the first three months and then only at night until the age of four years (26). The FAB size is changed as the child's foot grows. Both phases are equally as important for success of clubfoot management.

Figure 7 The FAB used in the maintenance phase

1.8 Evidence for clubfoot treatment

There is some evidence from a small number of randomised controlled trials (RCTs), before-and-after treatment studies and case studies to suggest that the minimally invasive Ponseti method is the treatment of choice.

In 2009 Zwick et al (36) conducted a RCT of the Ponseti method versus surgical treatment with 19 infants (28 feet). The trial was stopped for ethical reasons, before the sample size that was calculated *a priori* was reached, because interim analysis revealed that the surgical group had worse outcome. Minimum follow-up was 3.3 years. In the study, the Ponseti method resulted in better parental satisfaction and passive mobility of the corrected foot when compared to surgery. These results are supported by other observational studies. A cohort study of 45 patients (73 feet) that had undergone clubfoot surgery found a correlation between the extent of clubfoot surgery and the degree of functional limitation (mean follow-up of 30 years) (29). A second cohort study of 24 patients reported that the patients treated with extensive open foot surgery experienced more foot pain, weakness and limited foot range of motion when compared to 48 age matched controls (mean follow-up 21 years) (37). A retrospective study comparing surgery (n=24) against the Ponseti method (n=18) with a control group of healthy young adults (n=48) (38) found an increased range of motion in the foot, greater strength and less arthritis in the adults treated with the Ponseti method compared to surgery. The surgical group had the lowest ankle power generation (strength) compared with the control subjects.

A Cochrane review of clubfoot treatment in 2014 (39) included 14 trials (607 children) and concluded that the Ponseti technique produced better short-term foot alignment compared to other non-operative methods. Recurrence of the deformity following the Kite method led to extensive open foot surgery more often than recurrence following the Ponseti technique.

A systematic review of treatment of clubfoot in children under 2 years old (12 studies, 852 children) in 2017 (40) found that the Ponseti technique required fewer casts, and had a shorter duration to achieve correction and a lower recurrence rate when compared to other techniques. The majority of included studies were case series.

The Ponseti technique for clubfoot treatment has replaced the previous methods as the first line of management for clubfoot in many countries (it is used in 113 of the 193 United Nations member states (41)), and has been shown to reduce the need for extensive open foot surgery (42, 43).

1.9 Untreated clubfoot

Children with untreated or partially treated clubfoot walk on the side or top of their foot because the bones and joints of their foot become fixed in the deformed position (Figure 8).

Figure 8 Child with untreated clubfeet

Untreated clubfoot results in decreased mobility, pain, skin breakdown, callus formation and the inability to wear standard shoes (22, 44), all of which contribute to disability. The World Report on Disability (2011) (45) identifies children with disabilities as being amongst the most socially excluded and vulnerable. Children with neglected or partially corrected clubfoot are therefore likely to experience less access to education, social exclusion and discrimination because of the physical deformity (46). The personal, social and economic consequences for untreated clubfoot extend beyond that of the individual, to their families and wider community, and the cycle of disability and poverty is well recognised (47). A recent systematic review of poverty and

disability in LMIC (48) found evidence for a positive association between physical impairment and poverty in 78% (14/18) of included studies.

The concept of ‘access’ describes the relationship between a child with clubfoot and the family, and the health system. Penchansky and Thomas (1981) (49) present access as the “degree of fit” between the population and the health system. The authors define access as a general concept that includes the dimensions of availability, accessibility, accommodation, affordability and acceptability. Factors to consider for access include the supply features of the health system and the demand features of the population (50). Levesque et al (51) define access to healthcare as the opportunity to have healthcare needs fulfilled. The authors expand on the definition of Penchansky and Thomas (1981) (49) and propose that patient centred access includes approachability, acceptability, availability, affordability and appropriateness of the service, with the population able to perceive, seek, reach, pay and engage in services.

Studies that investigate access to clubfoot care in LMIC include evaluation of carer and service provider barriers. A study in four countries in 2015 (Argentina, the Netherlands, Indonesia and South Africa), to identify factors that influence initiation of clubfoot treatment, undertook semi-structured interviews with caregivers of children with clubfoot (n=51) and practitioners treating clubfoot (n=11) (52). The study found that cost of transport, long duration of travel, poverty, lack of awareness and missed working days form a barrier toward the start of treatment. A systematic review and meta-synthesis in 2016 of factors that affect patient access to clubfoot treatment in LMIC included 11 qualitative studies (53). The study used the five levels of the Social Ecological Model (intrapersonal factors, interpersonal processes, institutional/organisational factors, social-cultural/community and public policy) (54) to inform the findings of the meta-synthesis. The review found that caregivers experienced barriers at all five levels. At the intrapersonal level, these included limited access to finance and additional responsibilities within the home. Interpersonal influences were lack of support from fathers, extended family and the wider community. Institutional or organisational factors included long distances to treatment centres, and insufficient information about treatments and bracing protocols. Sociocultural influences included stigma associated with the impairment. At the

level of public policy, caregivers who were dependant on social services or limited insurance policies experienced longer delays in acquiring braces than those accessing comprehensive private care.

1.10 Cost effectiveness of clubfoot treatment

The Ponseti technique is cost-effective in low resource settings when compared to treating other conditions (55) and surgical correction (56). Grimes et al (55) found the average cost of the Ponseti treatment to be USD167 per patient using data from 12 countries in sub-Saharan Africa. The average number of disability-adjusted life years (DALYs) averted was 7.42. The cost of averting one year lost to disability due to clubfoot (cost-effectiveness ratio (CER)) (57) was USD22.46 per DALY averted. This is similar to the median CER of standard vaccinations (USD12.96–25.93 per DALY) (58). In Pakistan, Hussein et al (56) found that the average cost for the Ponseti method was USD349 compared with USD810 for children treated surgically.

1.11 Task-sharing and task-shifting in clubfoot management

In high-income settings, some clubfoot clinics have adopted a task-sharing approach between physiotherapists and orthopaedic surgeons. A retrospective cohort study in a tertiary hospital in Toronto, Canada, found that the Ponseti method of casting was equally as effective when directed by a physiotherapist as compared to a surgeon, with fewer recurrences and a less frequent need for additional procedures in the physiotherapist-directed group (59). In settings where the number of trained surgeons is limited, non-specialised healthcare providers can carry out clubfoot treatment. For example, the orthopaedic clinical officer programme in Malawi has produced healthcare workers competent to manage clubfoot deformity by conservative treatment, and perform minor soft tissue surgery such as tenotomy, in small rural district hospitals (60). Of 100 consecutive cases treated by orthopaedic officers, 98 of the feet achieved plantigrade (a position where the child can stand with feet flat on the floor) (61).

1.12 Research framework for clubfoot management

This PhD thesis proposes a theory of change to guide the generation of data to plan, implement and improve health care services for children with clubfoot in Africa. This model was developed to understand the clubfoot service within the

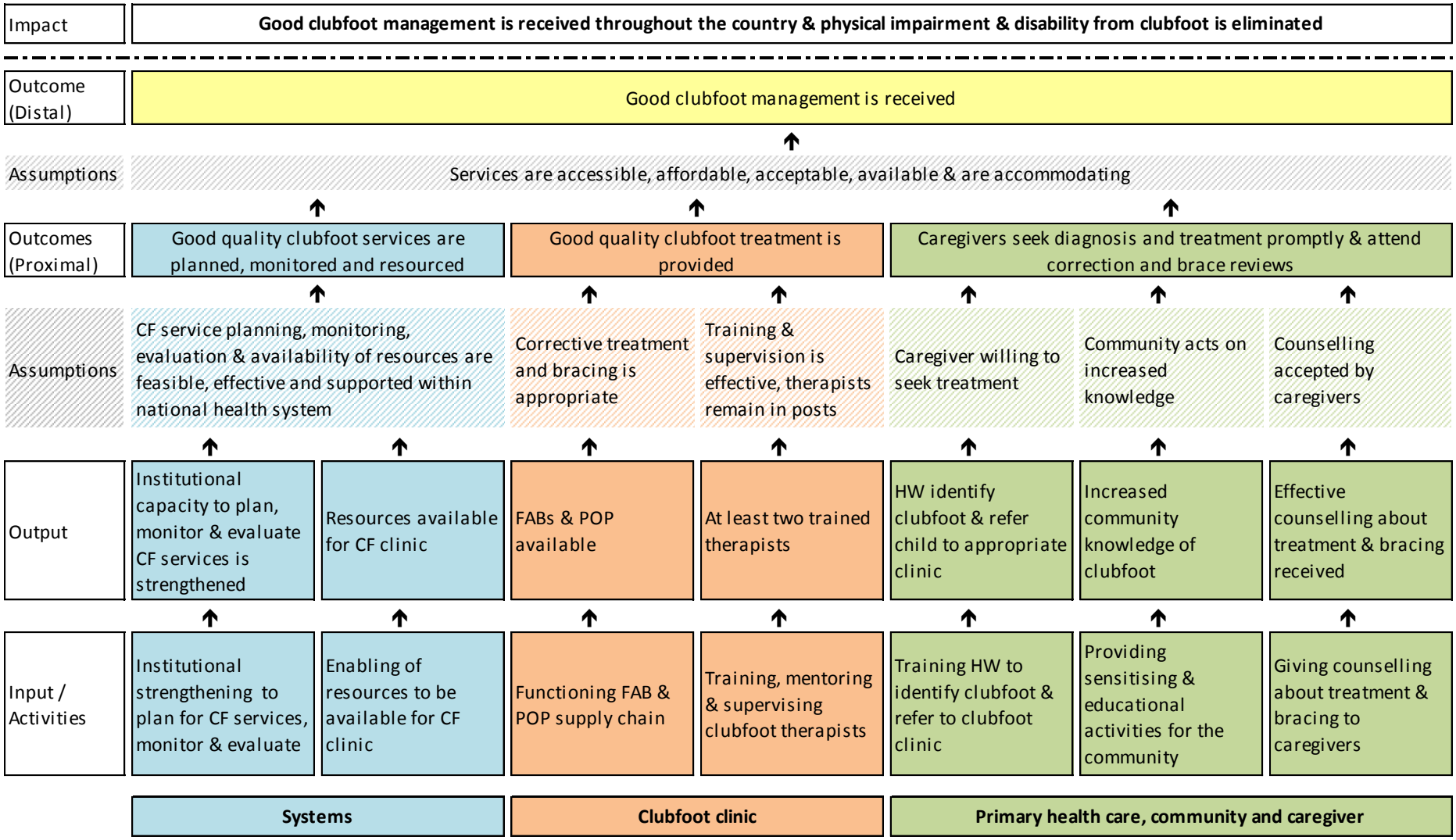
health system of Zimbabwe, and the pathways that determine the extent to which the delivery and receipt of clubfoot treatment (Ponseti technique) is successful. For the purpose of this PhD thesis, a ‘good clubfoot service’ is defined as a clubfoot service that is appropriate to the needs of the population given the available resources in the country and that meet national standards.

A theory of change is a conceptual model. It illustrates hypothesised causal pathways. For example, this model contains key elements that, in theory, change what occurs in clubfoot services for good clubfoot management to be received. It links outcomes with activities to explain how and why the desired change is expected to come about (62).

This theory of change was refined through the course of the PhD to reflect on-going understanding and research findings. Consensus on the hypothesised causal pathways was reached through two workshops. The workshops included experts in clubfoot service provision in Africa, health service provision in low resource settings and experts in evaluation of complex interventions.

The base of the design illustrates the activities that are required to achieve three proximal outcomes of **(i) good quality clubfoot services are planned, monitored and resourced; (ii) good quality clubfoot treatment is provided;** and **(iii) caregivers seek diagnosis and treatment promptly and attend correction and brace reviews** (Figure 9). The hypothesised causal pathways are denoted by arrows, which indicate the relationship between an activity and the output that is produced from the activity. The three proximal outcomes are preconditions to achieving the distal outcome of **‘good clubfoot management is received,’** which is positioned at the top of the design. Assumptions of context and processes are noted in shaded boxes.

A dashed line is drawn between the distal outcome and the impact of ‘good clubfoot management is received throughout the country and physical impairment and disability from clubfoot is eliminated.’ This is the ceiling of accountability and is the level at which the distal outcome may contribute to impact, but will require additional factors, for example, ‘a just society.’ (63)



Legend: CF = clubfoot; HW = health worker; FAB = foot abduction brace; POP = plaster of paris

Figure 9 The theory of change for clubfoot services in Zimbabwe

The theory of change is one method that provides a framework for evaluation of services. For example, the theory of change considers the interdependence between caregiver demand for clubfoot services, and the supply of the services by the clubfoot clinic and the broader health system. The extent of the caregiver's knowledge and understanding of clubfoot may influence their decision to seek treatment, whilst the diagnosis of clubfoot and appropriate referral from a health worker will influence the extent to which early intervention at a clubfoot clinic can be provided. Indicators can be established for these hypothesised causal pathways (e.g. proportion of children identified with clubfoot within a catchment area; extent of caregivers' understanding of clubfoot treatment after diagnosis). Both quantitative and qualitative data can be generated from the indicators and measured over time to assess the extent of improvement.

The activities on the pathways to achieve good clubfoot management being received are identified as:

- Institutional strengthening to plan for, monitor and evaluate clubfoot services within the national health system;
- Enabling resources to be made available for clubfoot clinics;
- A functioning FAB and POP supply chain;
- Training, mentoring and supervising of clubfoot therapists;
- Training of health workers to identify children with clubfoot and to refer to clubfoot clinics;
- Providing sensitising and educational activities for the community; and
- Giving counselling about treatment and bracing to caregivers.

This PhD thesis aims to address the following proximal outcomes of the model:

1. **Quality of treatment provided:**

   (i) through the evaluation of short and long-term results of clubfoot treatment in a cohort, and factors that influence the outcome;

   (ii) through the development of a tool (Assessing Clubfoot Treatment (ACT) score) for clubfoot therapists to monitor their treatment and decide on need for referral for further intervention;

(iii) through evaluation of the feasibility of a training programme for clubfoot therapists in Africa that will support the implementation of the Ponseti technique.

2. **Clubfoot service planning and monitoring:**

   (i) through the development of a tool (Functionality Assessment clubfoot Clinic Tool (FACT)) to assess the functionality of clubfoot clinics in Zimbabwe, and identify the strengths and weaknesses leading to recommendations for improvement.

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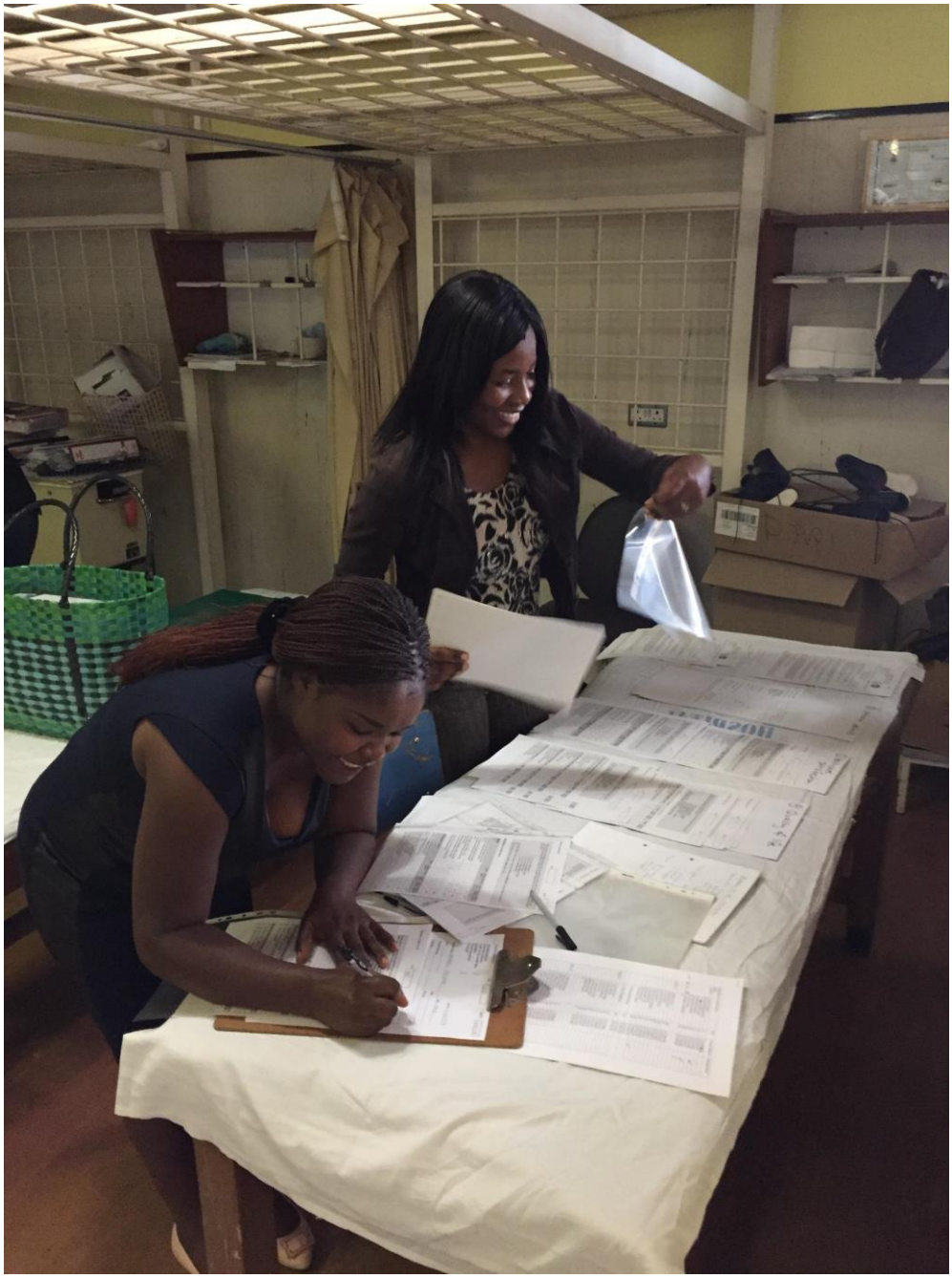
Chapter 2. Study rationale, aim, hypothesis and research questions



Members of the physiotherapy team at Parirenyatwa Hospital, Harare

2.1 Development of ideas for this thesis

As a paediatric physiotherapist working in Zimbabwe, I treated children with clubfoot using the Ponseti method. From 2011 – 2013 I trained health care workers and other physiotherapists to do the same. We worked in busy clinics on a 'first come first served' basis; there was little time to evaluate our results and training was on an *ad hoc* basis. I noticed that the clubfoot deformity responded well to corrective treatment in most cases but not in a few cases. I did not know what factors influenced these outcomes in our clinics, or how to evaluate our results in older children. I worked with committed clinicians and believed that the care being provided to children with clubfoot was good, but I had no method for testing this hypothesis.

Concern for my patients and for the health care workers that I was mentoring informed my decision to try and understand clubfoot services, and to identify opportunities for improvement in Zimbabwe. Work included in this PhD thesis follows the completion of my MPH (Developing Countries) in September 2015.

2.2 Study rationale

First, in order to plan and implement services to reduce impairment and disability from clubfoot in Africa information to estimate the magnitude of clubfoot is required.

Second, although there is a trend in Africa toward using the Ponseti technique for clubfoot treatment, there is no published review of the results in sub-Saharan Africa. In order to monitor, evaluate and improve clubfoot services in African countries it is essential to know what results can be expected and what factors influence the outcome.

Third, clubfoot services are delivered in designated clubfoot clinics in Zimbabwe; however, a tool to assess the functionality of these clinics and the areas that require improvement is not available.

Fourth, Zimbabwe is implementing strategies to improve maternal and child care; this includes policies that target improved recruitment and retention of experienced healthcare providers, and task-shifting some services to non-specialist health care workers (1). If clubfoot services are to be delivered by

specifically trained ‘clubfoot therapists’ it is important to explore the content and feasibility of a training programme for these clubfoot therapists, to optimise the task-shifting and task-sharing roles.

There is limited published data to inform these requirements. The body of work produced in this thesis is needed to address these gaps in evidence for the provision of care for children with clubfoot in Africa.

2.3 Aim

This thesis aims to determine how services for children with clubfoot in Africa can be improved using Zimbabwe as a case study.

2.4 Hypothesis

Children with clubfoot in Zimbabwe can be effectively treated by trained clubfoot therapists, using the Ponseti method, in appropriately equipped clubfoot clinics.

2.5 Research Questions

1. How many children are born with clubfoot per million population in African countries each year?
2. What are the reported results of clubfoot treatment (Ponseti technique) in Africa?
3. How can the results of Ponseti treatment be monitored and assessed by clubfoot therapists in Africa?
4. What are the short and long-term results of Ponseti treatment in a cohort of children with clubfoot in Zimbabwe and what factors influence the outcome of treatment?
5. What indicators are required to assess the functionality of clubfoot clinics in Africa?
6. What are the strengths and weaknesses of current clubfoot clinics in Zimbabwe; how can the services be improved?
7. What is the feasibility of a training programme for clubfoot therapists in Africa?

2.6 Components of the thesis

This PhD thesis contains the following components:

- Two systematic reviews. The first includes birth prevalence studies of clubfoot in LMIC to assess the magnitude of the condition. The second is a systematic review of the results of the Ponseti method as reported in sub-Saharan Africa.

- A Delphi process. This was used with experts in clubfoot treatment in Africa to establish the key criteria for measuring success of treatment.

- A cohort study of 218 children with clubfoot, treated in one clinic in Harare, Zimbabwe. This was used to document the short and long-term outcomes of clubfoot treatment and to identify the factors that influence the outcome.

- A second Delphi study. This was used to identify the indicators for assessing the functionality of a clubfoot clinic and service in Africa; the indicators were piloted in a tool to evaluate the Zimbabwe clubfoot clinics.

- A prospective mixed methods (both qualitative and quantitative) study. This evaluation of a training programme for clubfoot therapists informed the content of the programme and the feasibility of the training in Africa.

2.7 Research methodology

The use of both quantitative and qualitative methods, with a cohort study, a cross-sectional clinic service provision study and a pre-post evaluation of training aimed to provide multiple perspectives to determine ways in which to improve services for children with clubfoot (Table 2).

This research contributes to evidence to improve clubfoot services in Africa by employing the following methods:

Table 2 Research methodology

Research Question (2.5)	Method (2.6 and 2.7)	PhD thesis chapter
1	Systematic review and meta-analysis	4
2	Systematic review and integrative review	5
3	Delphi study	7
4	Case series: retrospective analysis of clinic records	6
4	Cohort study: prospective follow-up	8
4	Diagnostic accuracy study of five outcome measures	9
5	Delphi study	10
6	Cross-sectional clinic service provision study	10
7	Mixed methods (qualitative and quantitative) evaluation of training development	11
7	Feasibility study of training course for clubfoot therapists	12

2.8 Research timescale

The time plan and activities of this PhD thesis are outlined in Table 3.

Table 3 PhD thesis timescale

	2015	2016				2017				2018	
Activity	Sept-Dec	Jan - Mar	Apr - June	July - Sept	Oct - Dec	Jan - Mar	Apr - June	July - Sept	Oct - Dec	Jan - Mar	Apr - June
PhD Registration (25th Sept 2015)											
Systematic review and meta-analysis of clubfoot birth prevalence											
Analysis of baseline Zimbabwe cohort data											
Pilot clubfoot treatment training in Ethiopia											
Delphi process in Ethiopia											
Systematic review of results of Ponseti method											
Analysis of Delphi process											
Select, develop and translate evaluation tools and questionnaires											
Pilot data collection tools in Zimbabwe											
Pilot clubfoot treatment training in Rwanda											
Review of Zimbabwe pilot cohort data collection											
Zimbabwe cohort data collection											
Zimbabwe facility data collection											
Zimbabwe cohort data analysis											
Zimbabwe facility quality score analysis											
Zimbabwe cohort data synthesis											
Data analysis of ACT training											
Write up of PhD thesis											

2.9 Implementation of the research

I, Tracey H. Smythe, am the Principle Investigator (PI) in this research.

To evaluate the cohort and the Zimbabwe clubfoot programme, I worked with the Zimbabwe Ministry of Health and Child Care (Cecelia Nleya, Director of Disability and Rehabilitation Services), the Zimbabwe Sustainable Clubfoot Programme (Ryan Bathurst, Director) and the Parirenyatwa orthopaedic and clubfoot clinic team (Maxman Gova and Rumbidzai Muzarurwi respectively). The Research Assistant was Debra Mudariki, and Memory Mwadziwana and Mediatrice Mutsambi provided logistics support, undertook informed consent with the cohort and collected demographic data at the Parirenyatwa clubfoot clinic, Harare. I collected and extracted the data from clubfoot clinic charts.

The cohort was followed through the bracing phase and Debra Mudariki and I collected data.

The Zimbabwe clubfoot clinic functionality data were collected Debra Mudariki.

Andrew Wainwright provided advice on research study design for the Delphi studies and I collected data for the Delphi study.

I co-authored the Africa Clubfoot Training (ACT) course materials and participated in the design of project implementation. I conveived the framework for evaluation of the ACT project, and the ACT project team (Linda Hansen, Grace Le and Rosalind Owen) and I collected data for the feasibility study. Rosalind Owen collated and analysed the qualitative data from the ACT project.

I undertook the data cleaning and analysis, with statistic advice and support for the meta-analysis provided by David Macleod.

Professor Hannah Kuper (advisory committee) and Professor Jayne Webster (advisory committee) provided advice on this research.

Professor Allen Foster and Professor Christopher Lavy provided supervision and mentorship throughout the implementation of this research.

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Chapter 3. The Zimbabwe Sustainable Clubfoot Programme and the Africa Clubfoot Training project

Participants of the first Ponseti method training course in Parirenyatwa Hospital, Harare, March 2011

3.1 Research setting: Zimbabwe

Zimbabwe is a landlocked country situated in Southern Africa with an area of 390,757 square kilometres. The national census undertaken in 2012 estimated the population as 13.1 million (1). The World Bank estimated the population as 16,150,362 in 2016 (2). The capital city is Harare and Zimbabwe is divided into eight administrative provinces and two cities with provincial status (Figure 10), with 67% of the total population living in rural areas (3).

Figure 10 Map of Zimbabwe with administrative districts
(Credit: Ezilon maps)

Clubfoot services are provided in each administrative district and Table 4 provides information on services and estimates of population data from the 2012 Census (3). Figure 11 demonstrates the geographic spread of the clubfoot clinics in Zimbabwe.

Table 4 Clubfoot services provided in Zimbabwe

Province	Population*	Clubfoot service	No on map
Harare	2,123,132	Parirenyatwa Group of Hospitals, Harare	1
		Harare Central Hospital, Harare	2
		Chitungwiza Central Hospital, Chitungwiza	3
Mashonaland Central	1,152,520	Bindura Provincial Hospital, Bindura	4
Mashonaland West	1,501,656	Chinhoyi Provincial Hospital, Chinhoyi	5
Mashonaland East	1,344,955	Marondera Provincial Hospital, Marondera	6
Midlands	1,614,941	Gweru Provincial Hospital, Gweru	7
Manicaland	1,752,698	Mutare Provincial Hospital, Mutare	8
Masvingo	1,485,090	Masvingo Provincial Hospital, Masvingo	9
Matabeleland North	749,017	Tsholotsho Rehabilitation Village, Tsholotsho	10
Bulawayo Province	1,200,337	United Bulawayo Hospital, Bulawayo	11
		Mpilo Central Hospital, Bulawayo	12
Matabeleland South	683,893	Gwanda Provincial Hospital, Gwanda	13

*Population data from the 2012 census

Figure 11 Map of Zimbabwe with Ponseti clubfoot clinics (May 2018)
(Permission: Library of Congress)

Demographics

The crude birth rate for Zimbabwe in 2016 was estimated by the World Bank as 33/1,000 population (2). The 2015 Demographic and Health Survey estimates the national fertility rate in Zimbabwe is four children/mother and the under-five mortality rate is 69 deaths/1,000 live births (4).

Health system

The primary health care system in Zimbabwe is a pyramidal referral model; clinics and district hospitals provide local services for uncomplicated medical cases and refer patients with more serious or complicated conditions to the seven provincial hospitals (Mutare, Bindura, Marondera, Chinhoyi, Gwanda, Masvingo and Gweru) and six central hospitals. Orthopaedic services are offered in five of the six central hospitals (Harare Central, Parirenyatwa, Chitungwiza, Mpilo, United Bulawayo Hospitals) (5), however out-of-pocket expenses remain high, with hospitals charging on average USD50 for an x-ray (6). The majority (89%) of the population do not have health insurance (4). In 2012, 45% of orthopaedic positions were vacant, with 20% vacancy in both nursing and physiotherapy positions (5).

Zimbabwe as an African case study

There are several reasons for choosing Zimbabwe as the case study location. I was involved in the establishment of the Zimbabwe clubfoot programme and have in-depth knowledge and understanding of the background and decision-making processes that were undertaken to develop the programme. I mentored clubfoot clinic staff between 2011 and 2013 in Parirenyatwa Hospital, Harare, in a supervisory role that is similar to national co-ordinators of clubfoot programmes in Uganda and Tanzania. The clinic has well maintained records suitable for cohort analysis. The country size and distances between clubfoot clinics allowed data collection from all clinics within the study timeframe as each clinic has a supervisory visit at least twice a year. The findings of the research study may be applicable to clubfoot services in Africa where the challenges faced by most clubfoot therapists and caregivers of children with clubfoot are similar, such as limited human resources and specialised equipment.

Differences and similarities of national health expenditure also need to be considered for generalisability of research findings. Table 5 presents gross domestic product (GDP) (a measurement of a country's economy) and health expenditures of five countries in Africa, including Zimbabwe. The purchasing power parity (PPP) GDP allows comparison between countries by assessing similar goods, and health expenditure per capita PPP allows comparison of total health expenditure as a ratio of total population (7). The five countries spend $97 - $203 per person per year on health, with Zimbabwe spending $182.

Table 5 GDP and expenditure on health

Country*	PPP GDP ($)	GDP (USDmil)	Health Expenditure (%) of GDP (World Bank 2015)	Health expenditure/ capita PPP ($)
)Kenya	3,155	70,529	5.22	157
Tanzania	2,786	47,340	6.12	97
Uganda	1,819	24,079	7.30	139
Zambia	3,933	21,064	5.35	203
Zimbabwe	**2,027**	**16,620**	**10.32**	**182**

**In alphabetical order*

3.2 Clubfoot services in Zimbabwe

The Zimbabwe Sustainable Clubfoot Programme (ZSCP) was established in 2010. It is a non-government organisation (NGO) that is registered in Zimbabwe. Since 2012, the primary funder of ZSCP has been MiracleFeet, a registered non-profit organisation based in the USA. In Africa, MiracleFeet partners with clubfoot clinics in Guinea, Liberia, Madagascar, Senegal, Tanzania and Uganda. The aim of ZSCP is to eliminate clubfoot as a lifelong disabling condition in Zimbabwe by establishing an effective and sustainable countrywide clubfoot treatment program using the Ponseti method.

The Zimbabwe Ministry of Health and Child Care (MoHCC) initiated pilot clubfoot clinics that used the Ponseti method with the ZSCP in March 2011. Prior to this, the predominant technique used for clubfoot correction was the Kite method. I co-ordinated the ZSCP between 2011 and 2013, with the aim to set up clubfoot clinics with MoHCC at the Central and Provincial level that use the Ponseti method of treatment. To achieve this aim, local government physiotherapists and rehabilitation technicians were trained in the management of clubfoot with the Ponseti method and FABs were provided to clinics.

Two Ponseti treatment training courses were undertaken in Harare and Bulawayo in 2011. The programme expanded to Mutare and Gweru Provincial Hospitals in 2012. The training materials that were used were adapted from the Global Clubfoot Initiative (GCI) resources. In the first year, approximately 200 children were enrolled for treatment in the clubfoot clinics that used the Ponseti method.

The ZSCP continues to provide training, supervision and mentorship to local government clinic staff and, due to economic challenges in the past years, currently provides POP and underwrap for the casting process and FABs for the maintenance phase of clubfoot treatment. ZSCP also has a 'Ponseti guide' programme. The guides are trained to counsel caregivers of children with clubfoot and to assist with data collection in clinics.

3.3 The Africa Clubfoot Training project

The Africa Clubfoot Training (ACT) project was a two-year project that started in March 2015 with the aim to strengthen training and delivery capacity for clubfoot treatment in sub-Saharan Africa. The ACT project was a partnership between the Nuffield Department of Orthopaedics, Rheumatology and Musculoskeletal Sciences (NDORMS) at the University of Oxford, the Global Clubfoot Initiative (GCI), CURE Clubfoot and CURE Ethiopia Children's Hospital.

The GCI is a non-profit consortium of individuals and organisations with technical and organisational expertise in the Ponseti method of clubfoot management and with experience in establishing national clubfoot programmes in LMIC. The three main aims of GCI are to provide resources, create links and to collect and share information for clubfoot care in LMIC.

CURE International is a governing member of GCI. CURE International is a registered non-profit organisation based in the USA that was established in 1996 to provide treatment for children with physical disabilities in low resource settings.

The ACT project was funded by the UK Department for International Development (DFID) through a Health Partnership Scheme grant awarded by Tropical Health Education Trust (THET). The project proposed to:

i. Develop a training course for novice clubfoot treatment providers;
ii. Strengthen the training and delivery capacity for clubfoot treatment in sub-Saharan Africa; and
iii. Build capacity for clubfoot treatment training and mentoring.

The first pilot courses were undertaken in Addis Ababa, Ethiopia (September 2015, January and June 2016), Kigali, Rwanda (October 2016), and Nairobi, Kenya (January 2017) in CURE Hospitals.

3.4 Funding, collaborating institutions and ethical clearance

This PhD study was supported by the Beit Trust, through a research degree scholarship administered by LSHTM.

The Beit Trust, CBM, MiracleFeet and ZANE funded project costs.

Ethical approval was obtained though LSHTM and the Medical Research Council of Zimbabwe (MRCZ). In addition permission was granted by the Ministry of Health and Child Care (MoHCC), Joint Research Ethics Committee for the University of Zimbabwe, College of Health Sciences and Parirenyatwa Group of Hospitals (JREC), ZSCP and CURE Hospitals (Appendix 1).

3.5 Informed consent

In the cohort study, caregivers were contacted by telephone from information on their child's clubfoot clinic chart. Debra Mudariki, the ZSCP clinical supervisor, explained the study to the caregiver and the caregiver and child were invited to attend an assessment session. Informed written consent was obtained from the caregivers of children under the age of sixteen as per national guidelines, upon attendance of the assessment (Appendix 2). Two Parirenyatwa clinic physiotherapists explained the study to caregivers and children in Shona. Care was taken to explain the study objectives and procedures in light of the study involving a vulnerable population: children with, or with a history of, functional limitations. Children were referred to local services if an element of recurrence of the clubfoot deformity was evident on assessment.

Written consent was obtained from all participants prior to commencing training in the evaluation of the ACT project (Appendix 2).

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SECTION B RESULTS

Chapter 4. Birth prevalence of congenital talipes equinovarus in low- and middle-income countries: a systematic review and meta-analysis

The waiting room at the clubfoot clinic, Parirenyatwa Hospital, Harare

Preamble

Individual epidemiological studies of clubfoot report a variation in birth prevalence range from 0.39 – 6.8/1,000 live births and no quality assessment or analysis of these studies has been undertaken to date.

Accurate information about the magnitude of clubfoot is required to plan and implement services to reduce impairment and disability from clubfoot in Africa. Estimates of birth prevalence of clubfoot are therefore important to understand the actual need, so that the resources (human, equipment and financial) required to develop good health care services for children born with clubfoot can be planned and made available.

This is the first systematic review and meta-analysis of birth prevalence of clubfoot to be undertaken globally. The review includes 48 studies undertaken in LMIC between 1960 and 2015 with data from 13,962,989 children in 20 countries. Data are presented for each World Health Organisation (WHO) region as well as for India and China separately. The information is used to estimate how many children are born with clubfoot per million population in Africa each year.

This research paper was published in the journal Tropical Medicine & International Health after peer review in December 2016.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?	Tropical Medicine and International Health		
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Student Signature: [redacted] **Date:** 9/5/18

Supervisor Signature: [redacted] **Date:** 9/5/18

Tropical Medicine and International Health doi:10.1111/tmi.12833

VOLUME 22 NO 3 PP 269–285 MARCH 2017

Systematic Review

Birth prevalence of congenital talipes equinovarus in low- and middle-income countries: a systematic review and meta-analysis

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Abstract

OBJECTIVE Congenital talipes equinovarus (CTEV), or clubfoot, is a structural malformation that develops early in gestation. Birth prevalence of clubfoot is reported to vary both between and within low- and middle-income countries (LMICs), and this information is needed to plan treatment services. This systematic review aimed to understand the birth prevalence of clubfoot in LMIC settings.

METHODS Six databases were searched for studies that reported birth prevalence of clubfoot in LMICs. Results were screened and assessed for eligibility using pre-defined criteria. Data on birth prevalence were extracted and weighted pooled estimates were calculated for different regions. Wilcoxon rank-sum test was used to examine changes in birth prevalence over time. Included studies were appraised for their methodological quality, and a narrative synthesis of findings was conducted.

RESULTS Forty-eight studies provided data from 13 962 989 children in 20 countries over 55 years (1960–2015). The pooled estimate for clubfoot birth prevalence in LMICs within the Africa region is 1.11 (0.96, 1.26); in the Americas 1.74 (1.69, 1.80); in South-East Asia (excluding India) 1.21 (0.73, 1.68); in India 1.19 (0.96, 1.42); in Turkey (Europe region) 2.03 (1.54, 2.53); in Eastern Mediterranean region 1.19 (0.98, 1.40); in West Pacific (excluding China) 0.94 (0.64, 1.24); and in China 0.51 (0.50, 0.53).

CONCLUSION Birth prevalence of clubfoot varies between 0.51 and 2.03/1000 live births in LMICs. A standardised approach to the study of the epidemiology of clubfoot is required to better understand the variations of clubfoot birth prevalence and identify possible risk factors.

keywords clubfoot, congenital talipes equinovarus, birth prevalence, incidence, low income, developing country

Introduction

Congenital anomalies, also known as birth defects, are one of the leading causes of disability in children [1]. Clubfoot, or congenital talipes equinovarus (CTEV), is one of the most common congenital deformities that cause mobility impairment [2]. The structure and position of the foot are affected, and untreated clubfoot results in pain and reduced mobility, which potentially leads to participation restrictions and activity limitation [3].

Clubfoot forms in the early weeks of gestational development, and this may be part of specific syndromes or secondary to neurologic or systemic disease. However, the majority of cases occur in isolation and are termed 'idiopathic' [4], the cause of which is not fully understood [5]. Genetic factors have been implied [6, 7], while environmental factors, for example seasonal variation and intrauterine immobility, have been reported in some studies [5, 8]. Associations with ethnicity are not clear. Other risk factors that have been reported are male gender [9–11], maternal smoking [10–15] and maternal diabetes [10, 13]. However, the underlying pathogenesis for these factors remains a matter of scientific debate. A multifactorial aetiologic model that involves both environmental and genetic factors is likely [8].

Epidemiological studies consistently report higher birth prevalence [16] of idiopathic clubfoot in males and in firstborn children [17]. The condition is bilateral in half of the cases [18]. Typically, a small set of statistics are routinely cited for birth prevalence of clubfoot with reports of 0.39 per 1000 births in Chinese populations, 1.1 per 1000 in Caucasian and 6.8 per 1000 in Polynesian populations [19]. Overall, it is estimated that 80% of children born with clubfoot each year live in low- and middle-income countries (LMICs) [18].

Accurate collection of data on population birth rate and prevalence of birth defects is essential to plan,

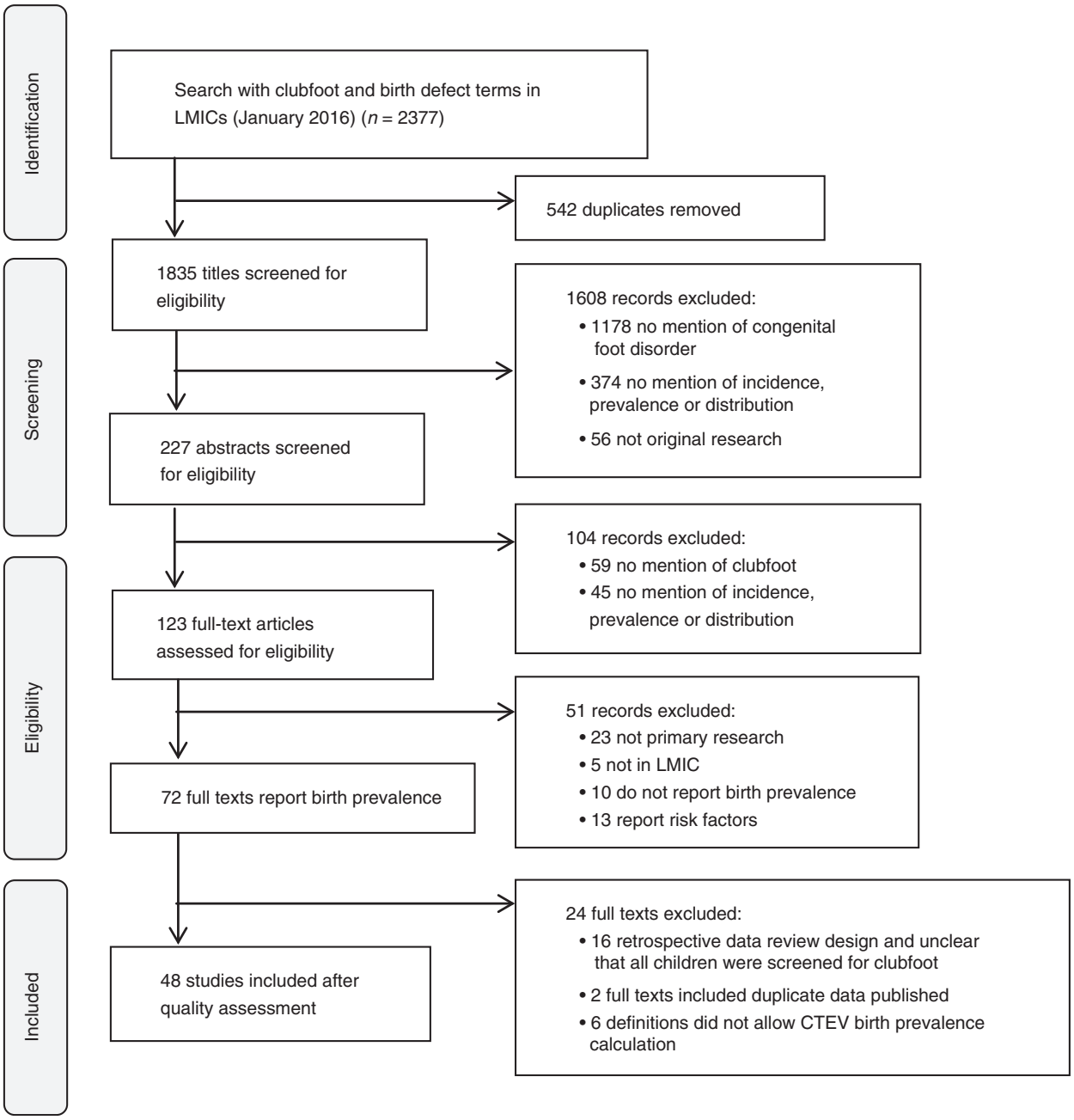

Figure 1 Search strategy with PRISM flow diagram.

Table 1 Studies of congenital talipes equinovarus (CTEV) birth prevalence included in systematic review, 1960–2015*

Reference												
Primary Author	Year	Country	Study time	Period of study	Population	Setting	Method of case ascertainment	Population *N*	Clubfoot *N*	Birth prevalence per 1000	Wilson (score)	Clubfoot definition
Africa												
Simpkiss [42]	1961	Uganda	1956–1957	10 months	Live births	1 Hospital	Clinical evaluation by medical student or midwife	1927	1	0.52	0.09–2.93	Musculoskeletal system: talipes equinovarus
Lesi [43]	1969	Nigeria	1966–1967	1 year	Births	1 Maternity hospital	Clinical evaluation within 12 h of birth by primary author and assistant nurse	16 720	19	1.14	0.73–1.77	Congenital defects found according to system' bony: talipes
Pompe van Meerdervoort [44]	1976	South Africa	Not defined	About 3 years'	Live births	1 Hospital	Physical examination day after delivery by paediatric/ orthopaedic registrar. Positive confirmation by clubfoot specialist.	10 000	35	3.50	2.52–4.86	Excluded spina bifida, arthrogryposis and CP. Noted CTEV: fixed deformity
Delport [45]	1995	South Africa	1986–1989	3 years	Live births	1 Urban academic hospital	Clinical evaluation within 24 h of birth by paediatrician or medical officer	17 351	8	0.46	0.23–0.91	Talipes equinovarus ICD-9 code 75450
Venter [46]	1995	South Africa	1989–1992	3 years 6 months	Live births	1 Rural hospital	Physical examination by trained nurse	7617	19	2.49	1.60–3.89	Musculoskeletal system: talipes equinovarus
Mkandawire [47]	2004	Malawi	2000–2002	22 months	Births	1 Hospital	Physical examination of all neonates, when and by whom not outlined	16 877	34	2.01	1.44–2.81	Definition of idiopathic and secondary
Mathias [48]	2010	Uganda	2006–2007	20 months	Live births	8 Regional hospitals	Physical examination by delivery room staff and confirmation by clubfoot specialist	110 336	130	1.18	0.99–1.40	Clear definition of clubfoot: positional, idiopathic or syndromic
Orimolade [49]	2014	Nigeria	2014	6 months	Live births	1 Tertiary hospital	Physical examination after delivery	1551	5	3.22	1.38–7.52	Idiopathic clubfoot variety

Table 1 (*Continued*)

Reference												
Primary Author	Year	Country	Study time	Period of study	Population	Setting	Method of case ascertainment	Population *N*	Clubfoot *N*	Birth prevalence per 1000	Wilson (score)	Clubfoot definition
Americas												
Monteleone-Neto and Castilla [30]	1994	Brazil	1982–1985	4 years	Live births	3 Maternity hospitals in Cubatao	Physical examination, prospective collection	10 218	21	2.06	1.34–3.14	ECLAMC definition of limb deformity: 'talipes'
Lopez-Camelo [37]	1996	Latin America	1967–1989	32 years	Births	Large database 24 geographic regions of Latin America	ECLAMC (Latin American Collaborative Study of Congenital Malformations)	2 159 065	3769	1.75	1.69–1.80	Pes equinus
Guardiola [39]	2009	Brazil	2000–2005	5 years	Births	Large database	ECLAMC	26 588	41	1.54	1.14–2.09	Clubfoot as defined by ECLAMC
Pachajoa [40]	2011	Columbia	2004–2008	4 years 7 months	Births	Review of data monitoring system in 1 tertiary care	Hospital birth defects monitoring system	32 995	58	1.76	1.36–2.27	ICD-10 and ECLAMC version 2002
South-East Asia												
Simatupang [50]	1977	Indonesia	1970–1975	5 years	Live births	1 Hospital [General RSUPP Medan]	Prospective, physical examination 'soon after birth'	17 241	19	1.10	0.71–1.72	Talipes
Kalra [51]	1984	India	Not specified	14 months	Births	1 Obstetric department, [SN medical college, Agra]	Physical examination within 28 h of birth, no description of by whom	2720	4	1.47	0.57–3.78	'Musculoskeletal system' talipes equinovarus
Bahadur [52]	1986	India	1980–1984	5 years	Live births	Not stated	Examination of infants after birth	13 321	21	1.58	1.03–2.41	Congenital talipes equinovarus
Chaturvedi [53]	1989	India	1986–1986	12 months	Births	1 Hospital	Physical examination within 48 h of birth	3014	3	1.00	0.34–2.92	Musculoskeletal talipes
Choudhury [54]	1989	India	1986–1987	12 months	Live births	1 Hospital, West Bengal	Physical examination	10 415	4	0.38	0.15–0.99	No numerator for clubfoot. Noted as talipes
Agarwal [55]	1991	India	1981–1984	2 years 6 months	Single births	1 Hospital	Physical examination during the early neonatal period	9405	30	3.19	2.24–4.55	Talipes equinovarus
Singh [56]	1991	India	1984–1987	4 years	Live births	1 Hospital, level II care to neonates	Physical examination by neonatal consultant	7015	32	4.56	3.23–6.43	CTEV

Table 1 (*Continued*)

Reference												
Primary Author	Year	Country	Study time	Period of study	Population	Setting	Method of case ascertainment	Population *N*	Clubfoot *N*	Birth prevalence per 1000	Wilson (score)	Clubfoot definition
Taksande [57]	2010	India	2005–2007	2 years 7 months	Live births	1 Rural medical college hospital	Physical review by consultant at time of birth	9194	11	1.20	0.67–2.14	Musculoskeletal system: 'talipes'
Chotigavanichaya [58]	2012	Thailand	2009	4 months	Births	1 Hospital	Clinical examination within 24 h	3396	8	2.36	1.19–4.73	Idiopathic clubfoot used Dimeglio classification
Pujari [59]	2012	India	Not defined	1 year	Live births	1 Hospital	Physical examination within 24–48 h of birth	4280	6	1.40	0.64–3.06	CTEV
Agrawal [60]	2014	India	2010–2011	1 year	Births >28 weeks	1 Tertiary care hospital	Physical examination for musculoskeletal defects within 24 h of delivery	7268	15	2.06	1.25–3.40	Talipes equinovarus noted these were not positional
Sachdeva [61]	2014	India	2010	4 months	Births	1 Government hospital	Clinical evaluation by paediatrician soon after birth. Data collection by 30 trained residents	2862	8	2.80	1.42–5.51	Talipes equinovarus
Baruah [62]	2015	India	2010–2013	2 years 9 months	Live births	1 Hospital	Physical examination within 24 h	17 052	23	1.35	0.90–2.02	ICD-10 classification
Europe												
Say [63]	1973	Turkey	1969	10 months	Live births >28 weeks	1 Hospital	Physical examination within 48 h by specialists	9947	22	2.21	1.46–3.35	Musculoskeletal system 'clubfoot'
Tuncbilek [26]	1999	Turkey	1993–1994	12 months	Births >20 weeks	22 university hospitals	Physical examination by paediatrician	21 907	43	1.96	1.46–2.64	ICD-10 was used 'pes equinus'
Eastern Mediterranean												
Akhtar [64]	1970	Pakistan	1965–1968	3 years	Live births	Obstetric department in 1 hospital	Obstetric dept. reported data, reviewed by research team	3570	3	0.84	0.29–2.47	Specific report of CTEV in Pakistan
Khrouf [65]	1986	Tunisia	1983–1984	9 months	Births	1 Teaching hospital	Clinical evaluation within 24 h of birth by paediatric staff	10 000	26	2.6	1.77–3.81	Musculoskeletal system, 'clubfoot' with additional deformities

Table 1 (*Continued*)

Reference												
Primary Author	Year	Country	Study time	Period of study	Population	Setting	Method of case ascertainment	Population *N*	Clubfoot *N*	Birth prevalence per 1000	Wilson (score)	Clubfoot definition
Bittar [66]	1998	Lebanon	1991–1993	2 years 6 months	Live births	1 Hospital	Physical examination within 24 h by senior resident and pre-discharge examination by paediatrician	3865	7	1.81	0.88–3.73	Musculoskeletal 'equinovarus'
Ali [67]	2008	Iran	2003–2006	2 years 8 months	Live births	1 Hospital	Physical examination after birth by primary author	4660	15	3.22	1.95–5.30	Musculoskeletal 'clubfoot'
Karbasi [23]	2009	Iran	2003–2004	8 months	Live births	All maternity hospitals in Yadz	Physical examination by Paediatrician	4800	19	3.96	2.54–6.17	ICD-10
Delshad [24]	2009	Iran	2005–2007	2 years	All births	Maternity wards in 6 government hospitals	Physical examination by paediatrician	61 112	62	1.01	0.79–1.30	ICD-10
Al-Ani [68]	2012	Iraq	2010–2011	1 year	Births	1 Tertiary hospital	Examined by neonatal specialists	5864	8	1.36	0.69–2.69	Specified difference between idiopathic and secondary
Golalipour [69]	2013	Iran	2007	1 year	Live births	1 Hospital	Physical examination by a paediatrician	6204	5	0.81	0.34–1.89	ICD-10 classification 'musculoskeletal system clubfoot'
El Koumi [70]	2013	Egypt	2011	1 year	Live births	1 Hospital	Physical examination within 24 h	2517	6	2.38	1.09–5.19	ICD-10 'musculoskeletal minor talipes'
West Pacific												
Emanuel [25]	1972	China	1965–1968	3 years	Singleton live and stillborn >28 weeks	6 Hospitals in Taipei	Examination by 1 of 4 study physicians, two public health nurses follow-up	25 549	19	0.74	0.48–1.16	ICD 1965 version, 'no reducible defect'
Boo [71]	1990	Malaysia	1988	4 months	Births	1 Maternity hospital,	Routine examination by Doctor, referred to research team	8369	4	0.48	0.19–1.23	Calculated structural clubfoot
Thong [27]	2005	Malaysia	2002–2003	14 months	Births	All health centres and hospitals in Kinta District	Population-based birth defect register: physical examination by trained nurse	17 720	23	1.30	0.87–1.95	ICD-10
Li [31]	2008	China	1997–2007	10 years	Births	Guangdong Province, 21 cities	Hospital-based surveillance programme	150 357	152	1.01	0.86–1.18	ICD-9 and ICD-10

Table 1 (*Continued*)

Reference												
Primary Author	Year	Country	Study time	Period of study	Population	Setting	Method of case ascertainment	Population *N*	Clubfoot *N*	Birth prevalence per 1000	Wilson (score)	Clubfoot definition
Hoang [28]	2013	Viet Nam	2010	1 year	Live births	127 Commune health stations	Physical examination within 24 h of birth	13 954	17	1.22	0.76–1.95	ICD-10
Li [41]	2013	China	2008–2010	2 years	Live births	4 Counties in Hengyang Province	Cluster sampling survey	52 307	50	0.96	0.73–1.26	Clubfoot
Yi [32]	2013	China	2001–2010	10 years	Births	Large database review	Data from birth defects monitoring programme	8 273 382	4233	0.51	0.50–0.53	ICD-10 Q66.0
Yi [38]	2013	China	2008–2011	4 years	Births	Birth defects monitoring programme in Dongguan	Prospective collection of birth defects on hospital cards	556 282	537	0.97	0.89–1.05	Congenital clubfoot
Wang [33]	2014	China	2006–2013	8 years	Births	Database review in Haikou	Review birth defects registry forms and perinatal infants quarterly report	118 199	62	0.52	0.41–0.67	Talipes equinovarus
Wei Hong [34]	2014	China	2011–2013	3 years	Births	Hospital surveillance programme in Zhuhai City	Data from monitoring institutions	87 059	53	0.61	0.47–0.80	Talipes equinovarus
Yang [35]	2015	China	2003–2009	7 years	Births	Database review 26 medical institutions [Longgang District]	Review of birth defects surveillance network	191 017	137	0.72	0.61–0.85	Talipes equinovarus
Xia [36]	2015	China	1997–2011	15 years	Births	Large database 75 hospitals [Henan Province]	Population-based congenital anomalies surveillance system	1 815 920	757	0.42	0.39–0.45	ICD-10

*Studies are ordered by WHO region and year of publication.

initiate and develop healthcare services. The aim of this study was to conduct a systematic literature review to estimate the birth prevalence of clubfoot in different World Health Organisation (WHO) regions, in order to inform planning of services and programme management in LMICs.

Methods

The systematic review was planned, conducted and reported according to established MOOSE (Meta-analysis of Observational Studies in Epidemiology) guidelines [20] (Appendix S1). A systematic literature search was conducted in January 2016 for peer-reviewed articles that presented original research findings on the birth prevalence of clubfoot in LMIC settings.

Search strategy

EMBASE, Medline, Global Health, LLACS, Africa-Wide Information and the Cumulative Index to Nursing and Allied Health Literature (CINAHL) were examined for studies published between January 1960 and December 2015 to capture 55 years of data. It was hypothesised that studies that reported on several congenital birth deformities may not include clubfoot in the search terms. Consequently, to capture all relevant studies, a search was carried out using both birth defects and clubfoot terms, with LMIC keywords. Boolean, truncation and proximity operators were used to construct and combine searches for the key concepts as required for individual databases, and an example is available as Appendix S2.

The articles returned by the literature search were screened by one reviewer (TS) first by title and then by abstract. 10% of the abstracts were reviewed by a second reviewer (HK) to check for agreement. The full text was obtained for any paper that was included at abstract screening.

Studies of all languages were included and translated as required. The reference list of all included studies was examined for further relevant studies. All full texts were reviewed independently by two reviewers (TS and either AF, CL or HK), and differences were agreed by discussion. The search strategy is presented in Figure 1.

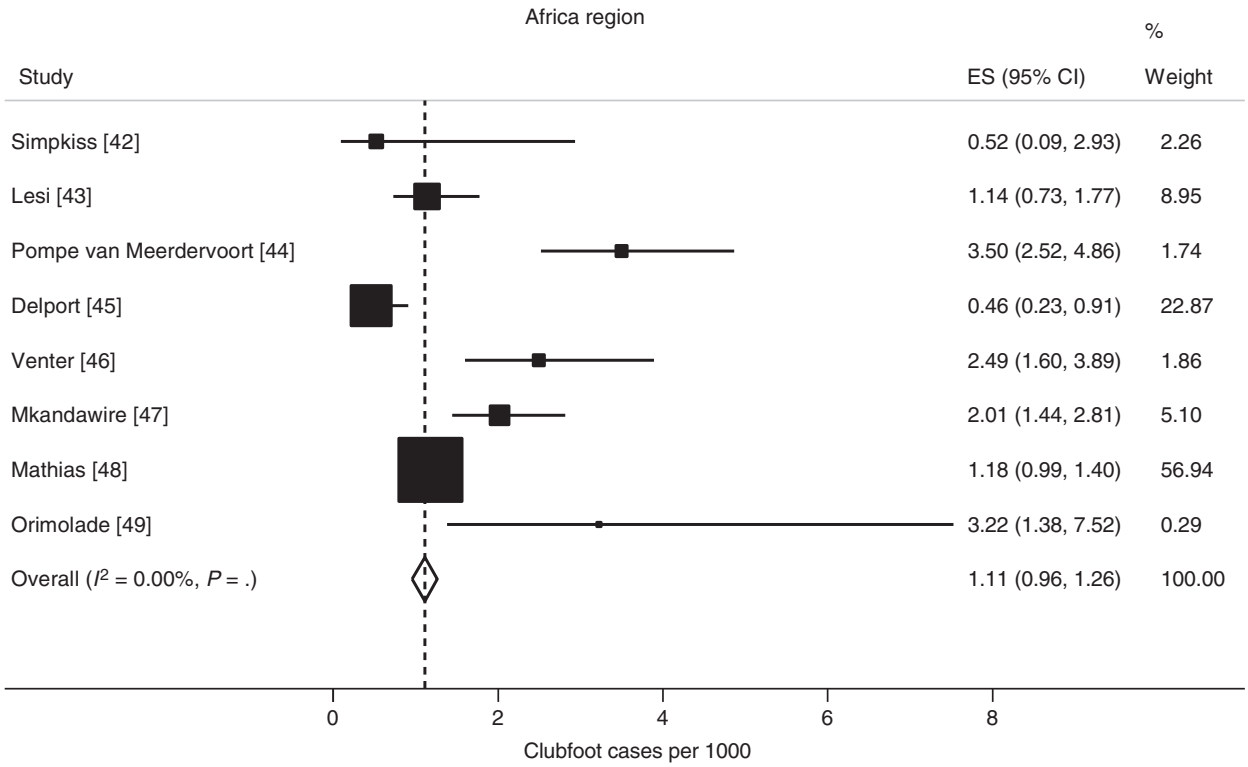

Figure 2 Birth prevalence of CTEV per 1000 births (Africa region). CTEV, congenital talipes equinovarus.

Study selection

Congenital talipes equinovarus (CTEV) was defined as a rigid deformity where the foot is fixed in a plantarflexed, supinated and adducted position. Studies were eligible if they met the following criteria: (1) original research that included CTEV; (2) results reported, or allowed calculation of, birth prevalence of clubfoot; (3) all children were screened for clubfoot; and (4) undertaken in a LMIC as defined by the World Bank country classification 2015. Exclusion criteria comprised the following: (1) full text unavailable, (2) unclear that all children were screened for clubfoot (e.g. large reviews of medical records), (3) unclear source population that prevented clear definition of the population denominator or (4) duplicate reports from the same study.

Data extraction and analysis

Data were extracted from articles that met inclusion criteria according to The Centre for Reviews and Dissemination guidelines [21]. The following data were extracted:

- General study information, including title, author and year of publication
- Study design
- Study setting and dates conducted
- Population characteristics
- Primary research outcome, including case definitions and results.

All extracted values were examined by the second reviewer to ensure accuracy. Differences between the reviewers were discussed, and a consensus was reached on all papers. One author was contacted for further information.

Data reporting per 1000 births were assumed to be live births unless it was stated that stillbirths were included. Birth prevalence rates were calculated per 1000 live births with 95% confidence intervals (Wilson score intervals), on the basis of the binomial distribution using Stata 14.0 (StataCorp LP, College Station, Texas), from the reported study population and the number of babies identified with clubfoot. It was decided *a priori* that the populations of China and India would be analysed independently of their WHO region due to their large population size. Tests for heterogeneity were performed. Weighted summary measures were estimated for the six WHO regions, India and China with a random-effects model [22] in the meta-analysis. The relative weight that each study contributed was defined by the sample size of the study. The overall effect estimate is therefore a weighted combination of the studies that contribute to it. Summary measures were graphed with forest plots.

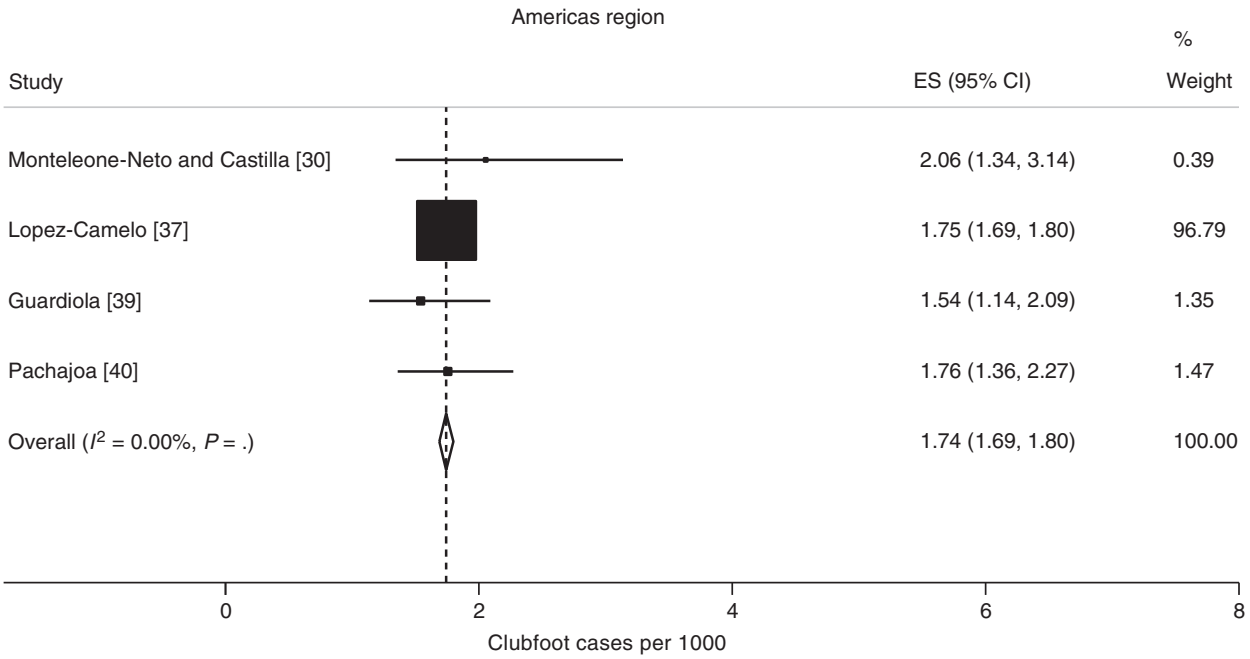

Figure 3 Birth prevalence of CTEV per 1000 births (the Americas region). CTEV, congenital talipes equinovarus.

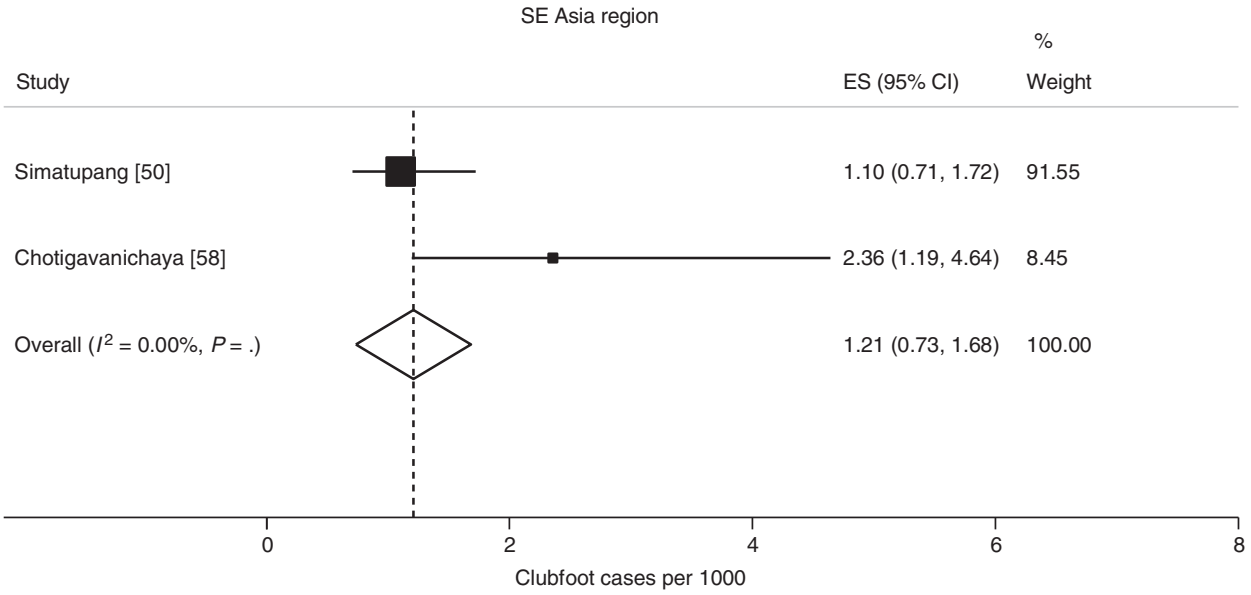

Figure 4 Birth prevalence of CTEV per 1000 births (South-East Asia region excluding India). CTEV, congenital talipes equinovarus.

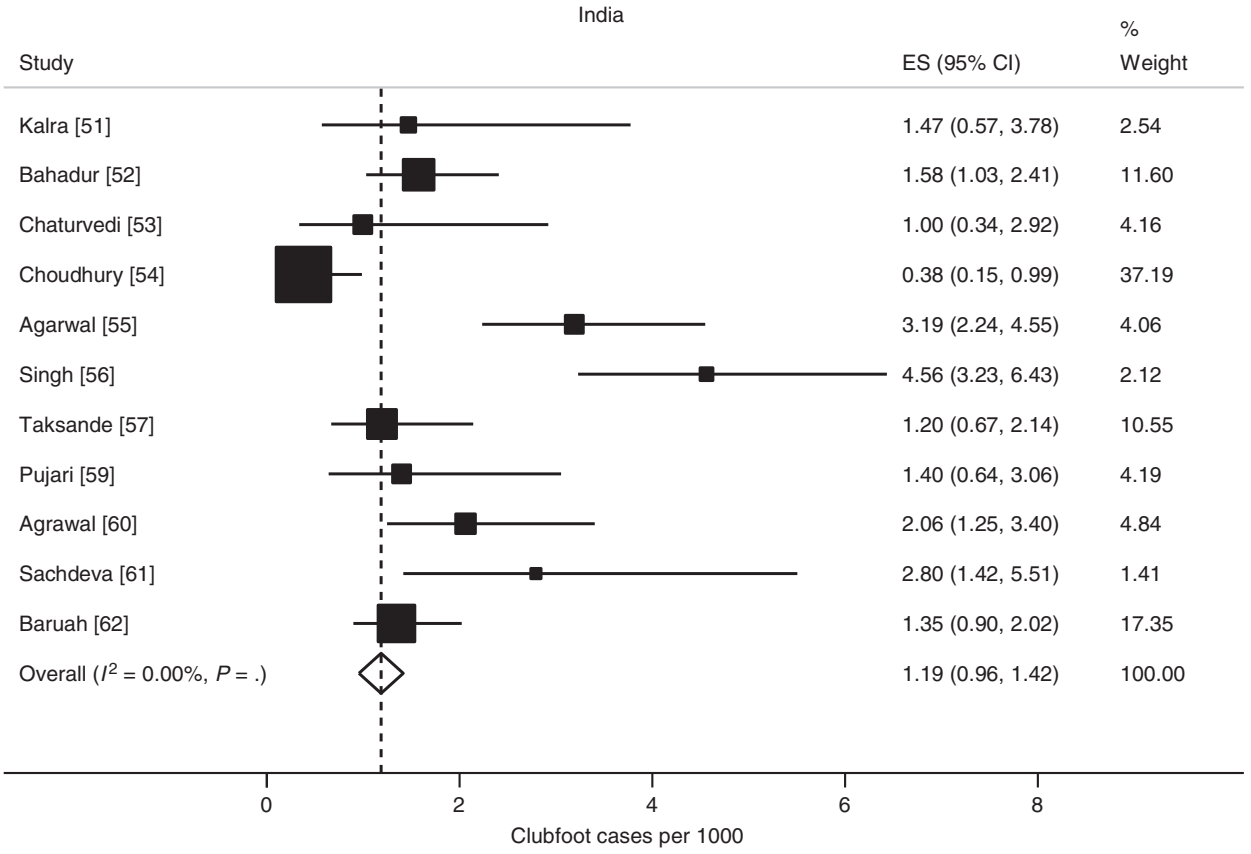

Figure 5 Birth prevalence of CTEV per 1000 births (India). CTEV, congenital talipes equinovarus.

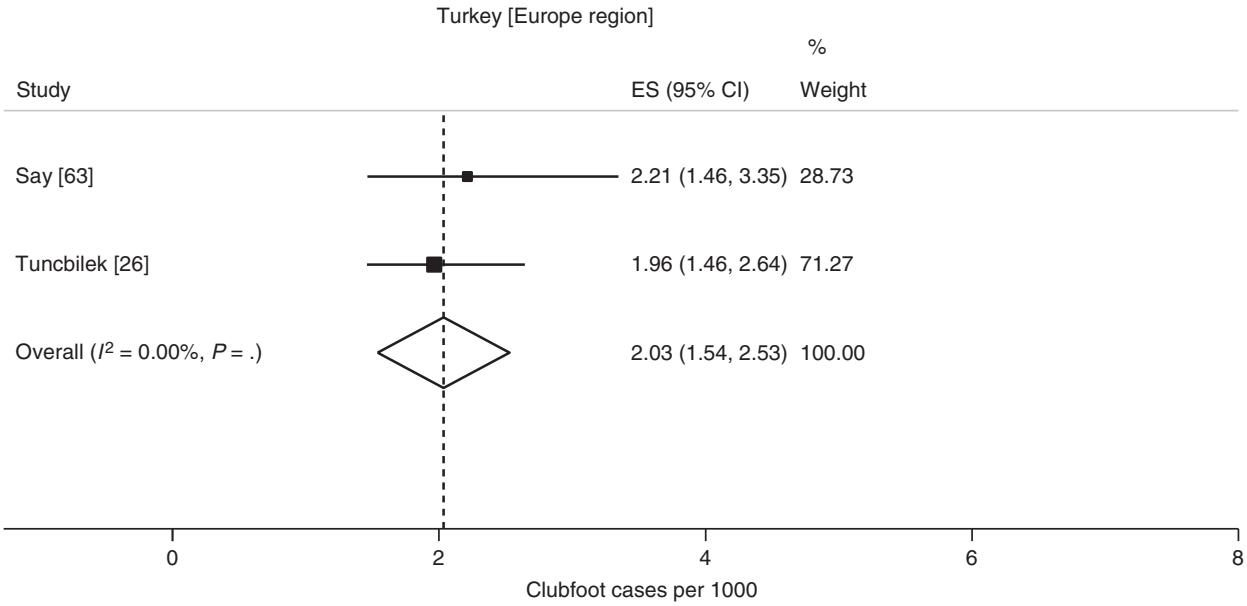

Figure 6 Birth prevalence of CTEV per 1000 births (Europe region). CTEV, congenital talipes equinovarus.

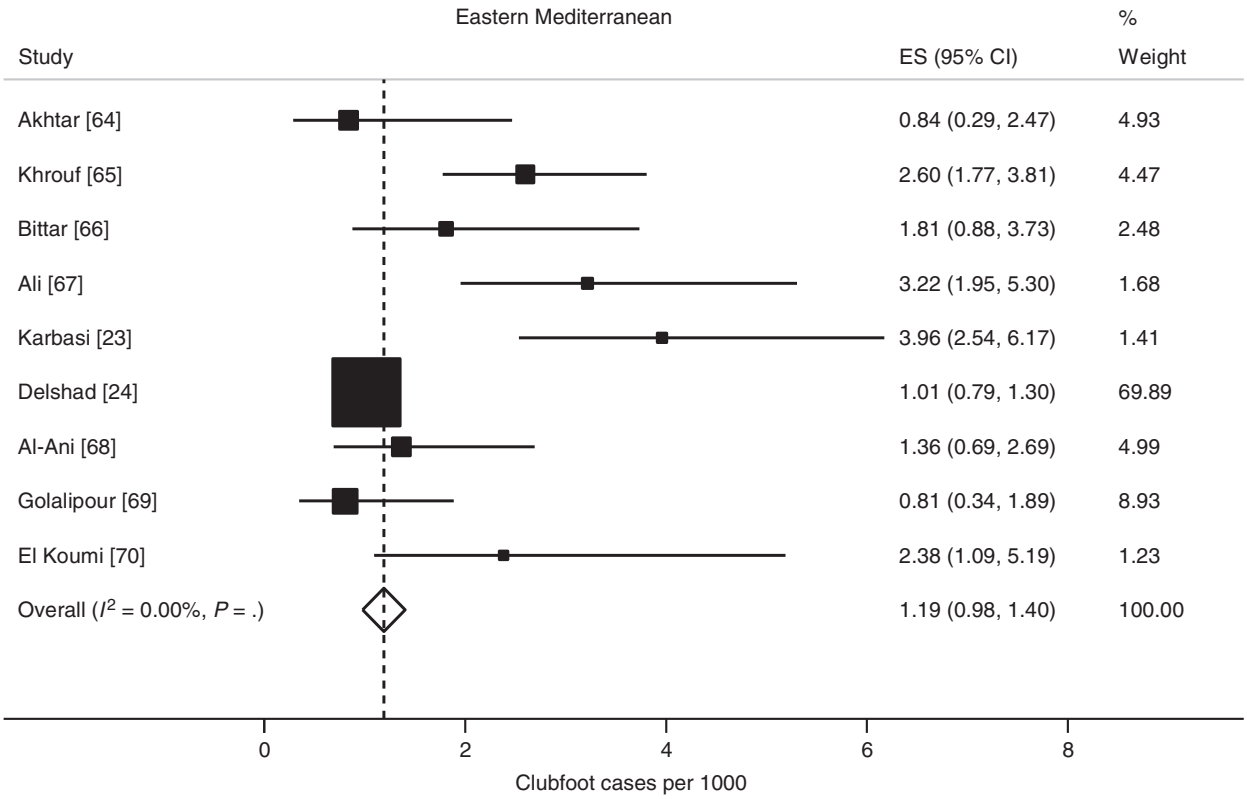

Figure 7 Birth prevalence of CTEV per 1000 births (Eastern Mediterranean region). CTEV, congenital talipes equinovarus.

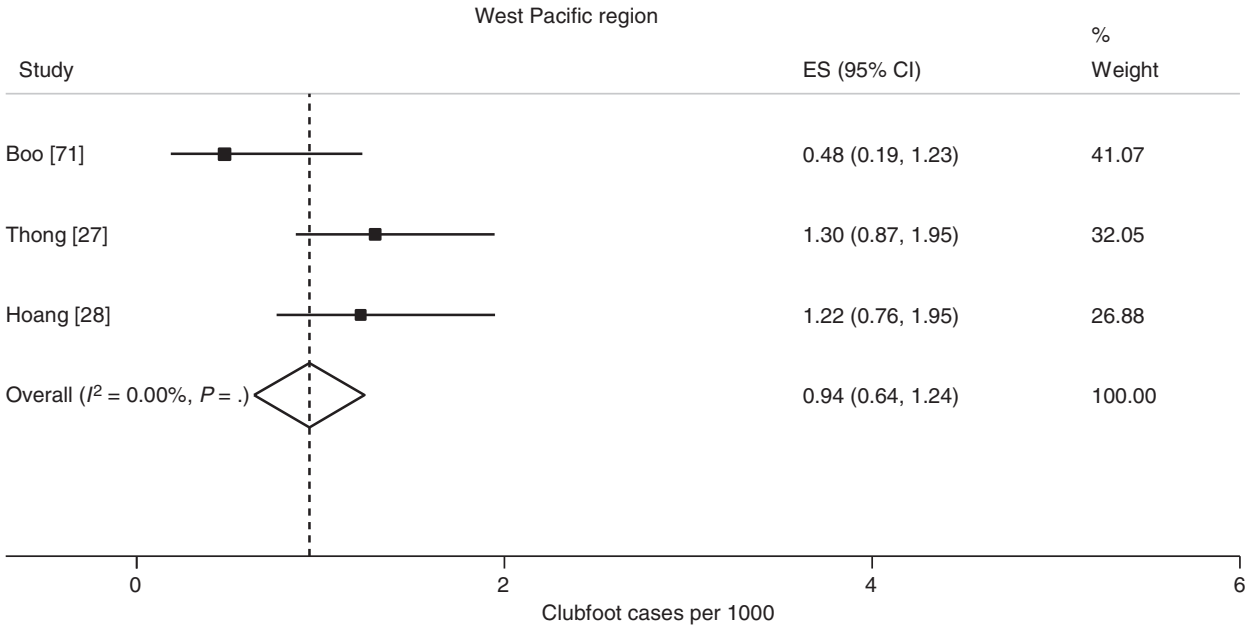

Figure 8 Birth prevalence of CTEV per 1000 births (West Pacific region excluding China). CTEV, congenital talipes equinovarus.

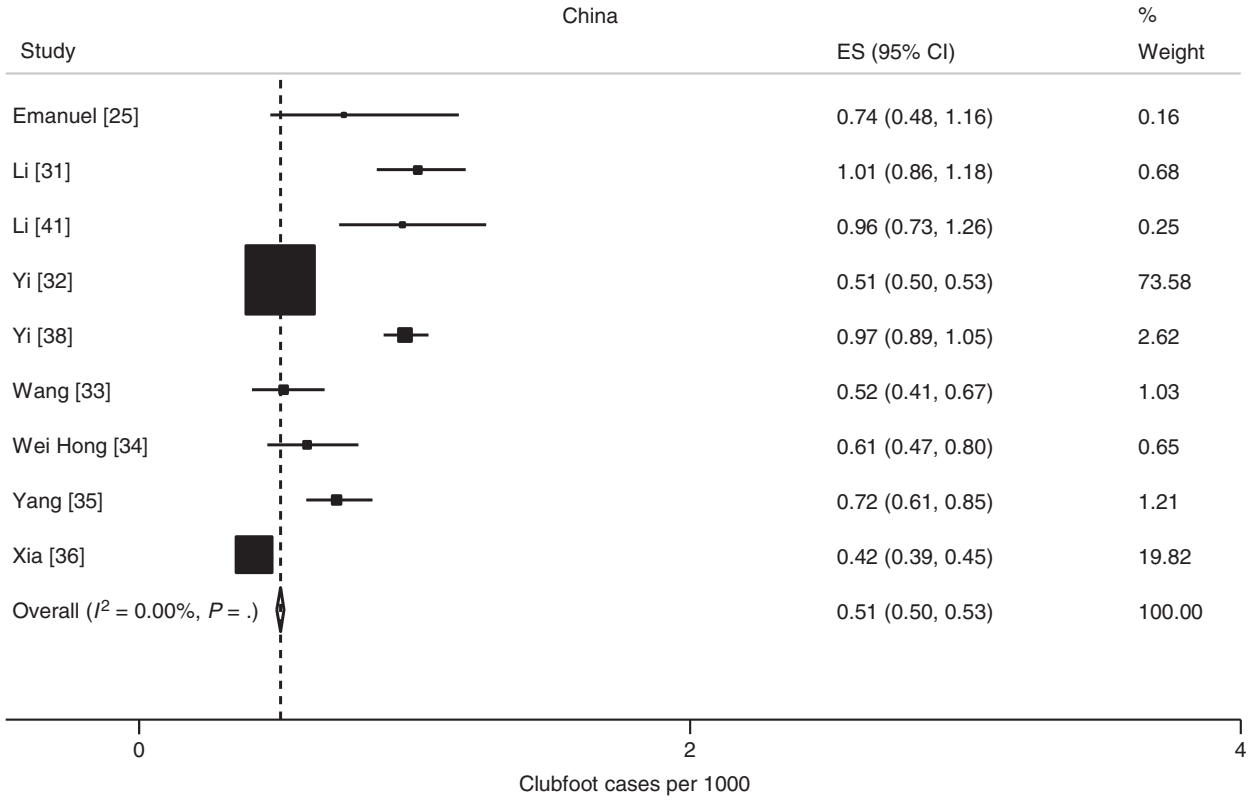

Figure 9 Birth prevalence of CTEV per 1000 births (China). CTEV, congenital talipes equinovarus.

As the time frame for the included studies is wide, an analysis was undertaken to identify whether the birth prevalence of clubfoot was different in the oldest estimates. A two-sample Wilcoxon rank-sum (Mann–Whitney) test was used to compare the birth prevalence in the time periods 1960–1985 and 1986–2015, consisting of 25 and 30 years, respectively.

Cases born per million total population per year were estimated according to regional clubfoot birth prevalence and crude birth rate per 1000 people. The Global Health Observatory data repository provided estimates of crude birth rate.

Results

A total of 1835 studies were retrieved for assessment (Figure 1). Of these, 72 studies reported on birth prevalence of clubfoot and provided data from 25 countries (Appendix S3). Twenty-four full texts were excluded, of which 16 papers were retrospective data collection and analysis and it was unclear whether all children were screened (Appendix S4 contains details on the studies excluded). Therefore, 48 studies were selected for inclusion and provided data from 13 962 989 children in 20 countries.

Table 1 summarises the characteristics of the studies eligible for inclusion. All the studies drew cases from a hospital setting. Eight of 37 studies (21.6%) that used a prospective design with physical examination were undertaken in more than one hospital [23–30]. Nine studies used a large database review in settings where there was systematic screening for clubfoot [31–39], one study analysed data from a single hospital defects monitoring system [40] and one study used a cluster sample survey [41]. Thirteen papers (27%) were from the South-East Asia region, with 11 papers in the region published from India. The West Pacific region consisted primarily of research undertaken in China and used large database reviews. Turkey was the only LMIC represented in Europe.

The pooled estimates for clubfoot birth prevalence for Africa (1.11 [0.96, 1.26]), South-East Asia (1.21 [0.73, 1.68]), India (1.19 [0.96, 1.42]) and the Eastern Mediterranean region (1.19 [0.98, 1.40]) are similar. The pooled estimate for clubfoot birth prevalence in LMICs within the Americas region is 1.74 (1.69, 1.80); in Turkey (Europe region) 2.03 (1.54, 2.53); and in West Pacific (excluding China) 0.94 (0.64, 1.24). The birth prevalence is lowest in China at 0.51 (0.50, 0.53).

Analysis of the birth prevalence of clubfoot reported in two date ranges (1960–1985 and 1986–2015) demonstrated no evidence of a difference over time ($P = 0.56$).

A meta-analysis by region was undertaken (Figures 2–9). The individual study results are displayed in the first column, identified under 'Study'. The summary birth prevalence is displayed in the final row with the test for heterogeneity denoted as I^2 (if $I^2 \leq 25\%$, studies are regarded as homogeneous). The second column visually displays the study results. The third column is the summary estimate of the birth prevalence of clubfoot, denoted by ES (95% CI) or effect size. This column gives the corresponding numerical results. The vertical line is the pooled estimate of birth prevalence, and the x-axis is the value of clubfoot cases per 1000 live births. The size of the box is directly related to the 'weighting' of the study in the meta-analysis, and the weight in % in the final column indicates the influence of the study on the overall results. The horizontal lines through the boxes depict the length of the confidence intervals. The diamond in the last row of the graph illustrates the overall result of the meta-analysis. The middle of the diamond sits on the value of the summary birth prevalence, and the width of the diamond depicts the width of the overall CI.

Based on the evidence since 1960, figures to plan for clubfoot management can be calculated for the eight populations given the birth rate per million population (Table 2). Population numbers are based on WHO region population birth rates. For planning purposes, regional estimates of birth prevalence should be applied to country specific birth rates.

Table 2 Projected clubfoot cases born per million total population/year

Region	Birth prevalence/1000	Crude Br/1000*	Clubfoot cases born per million total population/year
Africa	1.11 (0.96, 1.26)	38.3	43 (37–48)
Americas	1.74 (1.69, 1.80)	17.0	30 (29–31)
SE Asia	1.21 (0.73, 1.68)	19.9	24 (15–33)
India	1.19 (0.96, 1.42)	20.4	24 (20–29)
Turkey (Europe region)	2.03 (1.54, 2.53)	16.8	34 (26–43)
Eastern Mediterranean	1.19 (0.98, 1.40)	26.2	31 (26–37)
West Pacific	0.94 (0.64, 1.24)	14.7	14 (9–18)
China	0.51 (0.50, 0.53)	13.3	7

*Accessed WHO data, October 2016 (http://apps.who.int/gho/data/view.main.CBDR2040).

Discussion

This review summarises 48 studies of clubfoot birth prevalence from LMIC settings with data from 13 962 989 children in 20 countries. To our knowledge, this is the first systematic review of birth prevalence of clubfoot. The results demonstrate a range in birth prevalence from 0.51 (0.50, 0.53) per 1000 live births in China to 2.03 (1.54, 2.53) per 1000 in Turkey. Pooled estimates of birth prevalence rates appear to be similar in Africa, South-East Asia and Eastern Mediterranean regions and India. There was no evidence for a difference in clubfoot birth prevalence in LMICs between 1960–1985 and 1986–2015.

The case numbers and denominator population size differ in the individual studies included in the meta-analyses. The birth prevalence of clubfoot in China is strongly influenced by two large outlier studies [32, 36] that decrease the pooled estimate. Both studies were database reviews of data from hospitals that monitored birth defects through physical examination, and the data were collated on a congenital anomaly registration form. The authors note it is possible that cases were missed. Alternatively, the data may represent a unique feature of inheritance in the idiopathic clubfoot population of China. Only two papers contribute to the estimates of Turkey and the South-East Asia region with combined screened populations of 31 854 and 20 637 children, respectively.

Results compared to other studies

Many LMICs lack rigorous congenital anomaly surveillance programmes [72], which makes calculation of birth prevalence difficult. Current estimates range from 4 to 12 cases per 1000 births [73] in LMIC settings. These are likely underestimated due to stigma and exclusion [74] and are also reliant on case definition and robust screening methods. This analysis suggests some variation in the birth prevalence of clubfoot as previously indicated [75]; however, the range is not as large as reported by others [19]. Except for China, there were similar estimates across the regions.

Current data heterogeneity suggests the resulting variation in clubfoot birth prevalence in LMICs is likely influenced by study design and data collection methods and possibly by region and therefore ethnicity as well. Case definition, the case mix between tertiary and secondary facilities and the training of observers may affect prospective reporting of clubfoot. The true birth prevalence will be affected by risk factors, genetic and/or environmental, most of which are unknown.

Strengths and limitations

A strength of this study is the relatively large population denominator in several regions. It includes all categories of structural clubfoot (e.g. idiopathic or syndromic) as treatment is required in all cases although outcomes may differ. Data were excluded from clinics where it was not clear from the report how many babies were examined and did not have clubfoot, as birth prevalence cannot be calculated without a denominator. This has resulted in the exclusion of some studies [76, 77] that are regularly cited. This review is limited by the quality and representation of the available data from LMICs.

Implications

The estimated birth prevalence of clubfoot will be useful for the planning of services and to better estimate areas of need for country programmes. For instance, one equipped clinic in each district of 1 million people will be sufficient to handle clubfoot treatment if the new case load is up to 43 cases of clubfoot each year, as estimated by this review. Screening at birth for clubfoot is important, so that cases can be detected and treated early, when treatment is most effective. Scaling up appropriate services for screening and treatment remains a priority. Future studies should ensure that a clear case definition and robust screening methods are undertaken to allow comparison of epidemiological data.

Conclusions

Clubfoot is relatively common and should be detected at birth. There is no evidence for a large variation in birth prevalence between regions or of the folklore about a high Polynesian birth prevalence. Comparison of prevalence figures for congenital malformations reported from different parts of the world requires clear case definition and comparable methods of data collection. The published data over the last 55 years for clubfoot in LMIC suggest a birth prevalence in the range of 0.5 to 2.0 cases/1000 live births, which results in an estimated 7–43 cases of clubfoot/year/million population, dependent mainly on birth rate. The regional figures, for example in sub-Saharan Africa of approximately 43 cases/year/million population, provide useful information on planning treatment services for clubfoot in LMIC. A standardised approach to the study of the epidemiology of clubfoot is required to better understand the variations of the birth prevalence of clubfoot and possible risk factors.

Funding

This work was supported by The Beit Trust and CBM.

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Supporting Information

Additional Supporting Information may be found in the online version of this article:

Appendix S1. MOOSE Checklist [for Meta-analysis Of Observational Studies in Epidemiology].

Appendix S2. Search terms for clubfoot and birth defects and LMICs.

Appendix S3. Papers reporting CTEV birth prevalence published by year and WHO region prior to quality assessment.

Appendix S4. Full text excluded studies.

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<u>Web Appendix 1. MOOSE Checklist</u> [for Meta-analysis Of Observational Studies in Epidemiology]

Background

Problem definition

The birth prevalence of clubfoot or congenital talipes equinovarus (CTEV) is reported to vary in the literature due to inconsistent case definition and population denominators.

Hypothesis statement

Studies of CTEV in low and middle income countries (LMICs) report different birth prevalence estimates. Studies will be homogenised and the differences in birth prevalence will be examined in terms of WHO country definition and changes in time.

Study outcomes

- Birth prevalence of CTEV: Number of cases of congenital talipes equinovarus per 1,000 live births
- Generate a homogenous dataset that will allow for comparisons between LMICs and between the date ranges of 1960 – 1985 and 1986 - 2015.

Type of exposure or intervention used

- Geography was assessed using LMICs (World Bank 2005) in the WHO regions: African region, Region of the Americas, South East Asia Region, European Region, Eastern Mediterranean Region and the Western Pacific Region. India and China were estimated individually due to large population size.
- Changes over time were assessed in two time periods: 1960 – 1985 and 1986 – 2015
- The Global Health Observatory data repository provided estimates of regional crude birth rate to allow estimation of cases born per million total population per year.

Type of study design used

- Observational studies of CTEV

Study population

- All children in the study population were screened for clubfoot
- Clear definition of study population with a reliable estimate of the denominator population.
- If the population was well defined and birth prevalence given without the number of cases outlined, cases were calculated with the given information.

Reporting of search strategy

Qualifications of searchers

- Tracey Smythe has trained in systematic methods of literature searching as part of her PhD studies at the London School of Hygiene and Tropical Medicine
- Prof Christopher Lavy, Prof Allen Foster and Dr Hannah Kuper have participated in many systematic reviews.

The researchers were guided by:

- Jane Falconer, librarian at LSHTM and Fellow of the Higher Education Academy
- David Macleod, Research Fellow, Department of Medical Statistics

Search strategy

- Six medical literature databases searched between January 1960 and January 2016
- The following search terms were used as keywords:

1	developing country
2	developing or less* developed or under developed or underdeveloped or middle income or low* income
3	underserved or under served or deprived or poor*) adj (economy or economies).ti,ab.
4	developing or less* developed or under developed or underdeveloped or middle income or low* income
5	underserved or under served or deprived or poor*) adj (countr* or nation? or population? or world)).ti,ab.
6	(low* adj (gdp or gnp or gross domestic or gross national)).ti,ab.
7	(low adj3 middle adj3 countr*).ti,ab.
8	(lmic or lmics or third world or lami countr*).ti,ab.
9	Name of Country according to World Bank 2015 classification
10	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9
11	clubf??t
12	club-f??t
13	club ADJ1 f??t
14	talipes equinovarus
15	talipes ADJ2 equinovarus
16	talipes ADJ2 equino-varus
17	congenital talipes equinovarus
18	congenital ADJ2 talipes ADJ2 equinovarus
19	CTEV
20	11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19
21	birth defect or birth malformation or birth abnormality or congenital defect or congenital malformation or congenital abnormality
22	incidence or occurrence or frequency
23	21 and 22

24	20 or 23
25	10 and 25

Effort to include all available studies, including contact with authors

- The final list of included studies were discussed with experts in the field: Rosalind Owen, Executive Director of the Global Clubfoot Initiative and Nyengo Mkandawire, Professor of Orthopaedics, Malawi
- Studies of all languages were included and translated as required
- Primary authors were contacted for clarification of study period

Databases and registries searched

- EMBASE, Medline, Global Health, LLACS, Africa Wide Information and the Cumulative Index to Nursing, Allied Health Literature (CINAHL)

Use of hand searching

- The reference list of all included studies were examined for further relevant studies.

List of citations located and those excluded, including justification

- See web appendix 4 for full texts that were excluded
- Studies were first excluded by title and then by abstract
- The full text was obtained for any paper that was included at both title and abstract screening
- 72 full texts were read by 2 reviewers and included or excluded according to the criteria listed.

Inclusion Criteria	Exclusion criteria
(1) Original research that included congenital talipes equinovarus	(1) Full text unavailable
(2) Results reported, or allow calculation of, birth prevalence of clubfoot	(2) Unclear that all children were screened for clubfoot
(3) Undertaken in LMIC as defined by the World Bank country classification 2015	(3) Unclear source population that prevents clear definition of the population denominator
(4) All children were screened for clubfoot	(4)Duplicate reports from the same study

Method of addressing articles published in languages other than English

- Non-English articles were translated for each article obtained

Method of handling abstracts and unpublished studies

- Studies published only as abstracts where the case definition and denominator population were not defined were excluded, as were those that were unpublished.

Description of any contact with authors

- Four papers required additional information from authors to ascertain their precise study period. One author (Thong 2005) was contacted via the details

provided in the published study. The study period was clarified as 7th January 2002 to 28th February 2003.

Reporting of methods

Description of relevance or appropriateness of studies assembled for assessing the hypothesis to be tested

- All studies that provide observational epidemiological data on children born with confirmed CTEV
- Studies in LMICs in all WHO regions are included
- The studied period spans 55 years from 1960 to 2015

Rationale for selecting and coding of data

- Studies were included and excluded as per the criteria outlined above
- 10% of the abstracts were reviewed for agreement
- All full texts were reviewed independently by 2 reviewers (TS and either HK, CL or AF) and differences agreed by discussion
- Data were extracted according to The Centre for Reviews and Dissemination (CRD) guidelines
- Birth prevalence were recorded per 1,000 live births
- Wilson score confidence intervals were calculated

Documentation of how data were classified and coded

- Details are outlined within the methods section of the text

Assessment of confounding

- Not applicable with Wilcoxon rank-sum test due to small numbers of studies per region before 1985, two date ranges were assessed globally.

Assessment of study quality

- Inclusion/exclusion criteria were strict to ensure adequately defined study boundaries and case ascertainment

Assessment of heterogeneity

- Heterogeneity was assessed geographically, and statistically to identify differences between studies

Description of statistical methods

- Birth prevalence rates and Wilson score confidence intervals were calculated (using Stata 14.0) and plotted using forest plots
- The pooled estimate of birth prevalence was calculated per WHO region
- Wilcoxon rank sum test was used to examine change in birth prevalence between two time periods

Reporting of results

Graphic of individual summary estimates and pooled estimate

- Forest plots are drawn to demonstrate birth prevalence and confidence intervals
- A weighted pooled estimate of birth prevalence of clubfoot was calculated according to WHO region

Table giving descriptive information of all studies included and excluded

- Table of studies included is available within the body of text
- Web appendix 4 lists all excluded papers with reasons for exclusion

Indication of statistical uncertainty of findings

- 95% Wilson score confidence intervals for all calculated birth prevalence are given

Reporting of discussion

Quantification of bias

- Quantification of bias is not possible with this study design.

Justification of exclusion

- Papers were excluded based upon exclusion criteria and justification is outlined within text. Excluded papers and reasons for exclusion are listed in Web Appendix 4.

Assessment of quality of included studies

- The inclusion/exclusion criteria maximised the quality of included studies and the uncertainty is illustrated by 95% confidence

Reporting of conclusions

Consideration of alternative explanations for observed results

- Outlined in the discussion section of the main text

Generalisation of these conclusions

- Projected clubfoot cases per million population were estimated

Guidelines for further research

- Outlined in the discussion section of the main text

Disclosure of funding source

- Tracey Smythe received funding from the Beit Trust and Christian Blind Mission (CBM)

Web Appendix 2. Search terms for clubfoot and birth defects and LMIC

1	developing country
2	developing or less* developed or under developed or underdeveloped or middle income or low* income
3	underserved or under served or deprived or poor*) adj (economy or economies).ti,ab.
4	developing or less* developed or under developed or underdeveloped or middle income or low* income
5	underserved or under served or deprived or poor*) adj (countr* or nation? or population? or world)).ti,ab.
6	(low* adj (gdp or gnp or gross domestic or gross national)).ti,ab.
7	(low adj3 middle adj3 countr*).ti,ab.
8	(lmic or lmics or third world or lami countr*).ti,ab.
9	Name of Country according to World Bank 2015 classification
10	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9
11	clubf??t
12	club-f??t
13	club ADJ1 f??t
14	talipes equinovarus
15	talipes ADJ2 equinovarus
16	talipes ADJ2 equino-varus
17	congenital talipes equinovarus
18	congenital ADJ2 talipes ADJ2 equinovarus
19	CTEV
20	11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19
21	birth defect or birth malformation or birth abnormality or congenital defect or congenital malformation or congenital abnormality
22	incidence or occurrence or frequency
23	21 and 22
24	20 or 23
25	10 and 25

Web Appendix 3. Papers reporting CTEV birth prevalence published by year and WHO region prior to quality assessment

Years	Total number of Papers	African Region	Region of the Americas	South-East Asia Region	European Region	Eastern Mediterranean Region	Western Pacific Region
1960 - 1969	2	Uganda (1)[a], Nigeria (1)					
1970 - 1979	6	South Africa (1)		India (1), Indonesia (1), Pakistan (1)	Turkey (1)		Taiwan (1)
1980 - 1989	9	South Africa (1),		India (6), Thailand (1)		Tunisia (1)	
1990 - 1999	13	South Africa (2)	Brazil (1), Latin America (2)	India (3), Indonesia (1), Malaysia (1)	Turkey (1)	Lebanon (1), Pakistan (1)	
2000 - 2009	15	Malawi (2), Nigeria (2), Zimbabwe (1)	Brazil (1)	Malaysia (1)		Iran (4), Libya (1)	China (1), Philippines (1), Papua New Guinea (1)
2010 - 2015	26	Nigeria (3), Uganda (1)	Brazil (1), Columbia (2),	India (6), Thailand (1)		Egypt (1) , Iran (2), Iraq (1)	China (8), Viet Nam (1)

[a] Number of published papers

Web Appendix 4. Full text excluded studies

Primary Author and Reference	Year	Country	Study design	Data source	Time	Population	Population N	Clubfoot N	Birth prevalence /1000	Reason
Dash Sharma (78)	1970	India	Data review	Records of birth	3 years	live births	5,554	2	0.36	Retrospective data review, unclear if all children screened
Kromberg and Jenkins (76)	1982	South Africa	Data review	Data from register of births, nursery ward, paediatric ward and mortuary records	2 years	births	29, 633	46	1.55	Retrospective data review, unclear if all children screened
Choudhury (79)	1984	India	Data review	Hospital records of birth registers	4 years	births	21, 016	6	0.29	Retrospective data review, unclear if all children screened
Limpaphayom (80)	1985	Thailand	Data review	Medical records	2 years	births	not specified	104	1.30	No specified population
Roychoudhury (81)	1988	India	Data review	Maternity records	not specified	births	72, 617	not specified	varied from 0.02 to 0.11	Retrospective data review, unclear if all children screened
Castilla (82)	1990	Latin America	large database review	ECLAMC (Latin American Collaborative	4 years	births	Tropical: 287,165	tropical: 442	tropical: 1.54	ECLAMC data used in Lopez - Camelo paper
								non: 615	non: 1.06	

				Study of Congenital Malformations) data			Non tropical: 582, 585			
Masloman (83)	1991	Indonesia	Data review	Medical records of department of child health	5 years	births	13, 354	11	0.82	Retrospective data review, unclear if all children screened
Bhat (84)	1998	India	Prospective, physical examination	Physical examination within 24 hours	3 years 3 months	births	12, 797	40 (36 live, 4 still)	3.13	All foot deformities included
Najmi (85)	1998	Pakistan	Prospective, physical examination	Physical examination by Paediatrician	2 years 8 months	live births	11,148	2 TEV and 2 TE	0.18	Unclear definition
Singh (86)	2000	Libya	Data review	Maternal records, NICU registry and stillborn / death certificates	1 year	Births	16, 186	2: clubfoot or 4 talipes	0.37	Retrospective data review, unclear if all children screened
Madzivire (87)	2002	Zimbabwe	Data review	Children attending clubfoot clinic, <4yrs	3 years	Hospital catchment area	96,942	82	0.85	Retrospective data review, unclear if all children screened
Mkandawire (88)	2002	Malawi	Prospective physical examination	Research nurse identified and photographed	13 months	Live births	9,838	11	1.12	Data included in paper published in 2004
Padilla (89)	2003	Philippines	Large database	Birth defects registry	12 months	Births	191, 567	73	0.38	Includes all congenital

										deformities of the feet
Abdi-Rad (90)	2008	Iran	Data review	Chart review	4 years 6 months	Births	14, 121	27	1.90	Retrospective data review, unclear if all children screened
Ekanem (91)	2008	Nigeria	Data review	Data extracted from birth registries	23 years	Birth registry in 2 states of Nigeria	127, 929	31	0.24	Retrospective data review, unclear if all children screened
Culverwell (77)	2009	Papua New Guinea	Data review	Clubfoot clinic notes	2 years	Live births and hospital visits	11, 215	60	2.67	Retrospective data review, unclear if all children screened
Bakare (92)	2009	Nigeria	Prospective, physical examination	Physical examination	1 year	Live births	624	5	8.00	All foot deformities
Zarante (93)	2010	Columbia	Large database	ECLAMC	6 years 9 months	Births	52, 744	132	2.50	Foot deformities include calcaneovalgus
Ukoha (94)	2011	Nigeria	Data review	Hospital records	6 years	Children attending hospital (1 day - 2 yrs)	12,464	43	3.00	Data review for children attending hospital - not birth prevalence
Ekanem (95)	2011	Nigeria	Data review	Maternity records	13 years	Births	19,572	8	0.41	Retrospective data review, unclear if all children screened

Zhu (96)	2012	China	Data review	Data of neonates with congenital malformations	1 year	Live births	6,725	2	0.30	Retrospective data review
Vakilian (97)	2013	Iran	Data review	Review of maternal files	7 years	Live births	20,751	not specified	2.98	Retrospective data review, unclear if all children screened
Nhoncanse (98)	2014	Brazil	Data review	Review of birth certificates	5 years	Live births	12,199	4	0.32	Retrospective data review, unclear if all children screened
Ghorpade (99)	2015	India	Data review	Medical chart review	10 years	Live births	10, 674	60	5.62	Retrospective data review, unclear if all children screened

[a] ordered by year

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Epilogue (Main findings and recommendations)

The main finding of the systematic review and meta-analysis is the similarity of pooled estimates of birth prevalence of clubfoot in Africa, Eastern Mediterranean region, India and South East Asia (between 1.11 (95%CI 0.96 – 1.26)/1,000 live births and 1.21 (95%CI 0.73 -1.68)/1,000 live births).

There is no evidence for a large variation in birth prevalence of clubfoot between WHO regions.

The variation reported in the literature is likely due to inconsistent case definition and population denominators. Data heterogeneity suggests that the reported variation in clubfoot birth prevalence in LMIC is influenced by study design and data collection methods and possibly by region.

Overall the main limitation of the systematic review and meta-analysis is the low quality and narrow representation of available data, which is characterised by single tertiary hospital studies and large database reviews.

The birth prevalence of clubfoot in China is strongly influenced by two large database reviews that decrease the pooled estimate.

Only two papers contribute to the estimates of Europe (Turkey) and the South East Asia region with combined screened populations of 31,854 and 20,637 children respectively.

Additional Information: Erratum. The meta-analyses specified in this published paper uses a fixed-effects model. The I-squared statistic is not calculated (and is visualised by (I^2 = 0.00%, P = .) in all graphs) as the model assumes no heterogeneity between studies and allows weighting by study size. The birth prevalence data of clubfoot were visualised and there was no apparent trend over time.

Chapter 5. Assessment of success of the Ponseti method of clubfoot management in sub-Saharan Africa: a systematic review

Child wearing a foot abduction brace

Preamble

There is no agreed definition of successful outcome following both the correction and the bracing phases of the Ponseti method in clubfoot programmes in African countries.

There is a need for a standardised approach to report clubfoot treatment outcomes in order to establish normative standards to monitor, evaluate and improve clubfoot service delivery in Zimbabwe. This includes data on the expected results of clubfoot treatment and factors that influence these outcomes.

Although the Ponseti method is promoted as an effective and low cost treatment for clubfoot in Africa, there is no published review of the results of the Ponseti method in sub-Saharan Africa.

This chapter is a systematic review of the literature between 2000 and 2017 to determine the results of the Ponseti method in sub-Saharan Africa. The different ways that results are reported are evaluated with a view to developing a reference standard for the monitoring and evaluation of clubfoot treatment in Zimbabwe.

Twenty-two papers met the inclusion criteria and data were from ten countries: Ethiopia, Ghana, Kenya, Malawi, Mali, Nigeria, South Africa, Togo, Uganda and Zimbabwe.

This systematic review was published in the journal BMC Musculoskeletal Disorders in November 2017 after peer review.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?	BMC Musculoskeletal Disorders		
When was the work published?	15th November 2017		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion	n/a		
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Stage of publication	

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I designed the study, did the literature search, extracted the data, completed data analysis, interpreted the data, drafted the manuscript, prepared the subsequent revisions with consideration of comments from co-authors

Student Signature: [redacted] **Date:** 9/5/18

Supervisor Signature: [redacted] **Date:** 9/5/18

Smythe et al. BMC Musculoskeletal Disorders (2017) 18:453
DOI 10.1186/s12891-017-1814-8

BMC Musculoskeletal Disorders

RESEARCH ARTICLE Open Access

Assessment of success of the Ponseti method of clubfoot management in sub-Saharan Africa: a systematic review

Tracey Smythe[1*], Debra Mudariki[2], Hannah Kuper[1], Christopher Lavy[3] and Allen Foster[1]

Abstract

Background: Clubfoot is one of the most common congenital deformities affecting mobility. It leads to pain and disability if untreated. The Ponseti method is widely used for the correction of clubfoot. There is variation in how the result of clubfoot management is measured and reported. This review aims to determine and evaluate how success with the Ponseti method is reported in sub-Saharan Africa.

Methods: Five databases were examined in August 2017 for studies that met the inclusion criteria of: (1) evaluation of the effect of clubfoot management; (2) use of the Ponseti method; (3) original study undertaken in sub-Saharan Africa; (4) published between 2000 and 2017. We used the PRISMA statement to report the scope of studies. The included studies were categorised according to a hierarchy of study methodologies and a 27-item quality measure identified methodological strengths and weaknesses. The definition of success was based on the primary outcome reported.

Results: Seventy-seven articles were identified by the search. Twenty-two articles met the inclusion criteria, of which 14 (64%) reported a primary outcome. Outcomes were predominantly reported though case series and the quality of evidence was low. Clinical assessment was the most commonly reported outcome measure and few studies reported long-term outcome. The literature available to assess success of clubfoot management is characterised by a lack of standardisation of outcomes, with different measures reporting success in 68% to 98% of cases.

Conclusion: We found variation in the criteria used to define success resulting in a wide range of results. There is need for an agreed definition of good outcome (successful management) following both the correction and the bracing phases of the Ponseti method to establish standards to monitor and evaluate service delivery.

Keywords: Clubfoot, Congenital talipes equinovarus, Ponseti, Outcome, Evaluation, Treatment, Success, Africa, Sub-Sahara

Background

Clubfoot, or congenital talipes equinovarus (CTEV), is one of the most common congenital musculoskeletal deformities. Within the Africa region, clubfoot birth prevalence is estimated as 1.11 (95%CI 0.96–1.26) per 1000 live births [1]. Untreated clubfoot results in pain, physical impairment and can ultimately cause disability [2]. The Ponseti method is widely used for the management of clubfoot [3]. It consists of two distinct phases, the correction phase and the maintenance phase [4]. The correction phase involves precise manipulation of the foot around the talus to correct the cavus, adductus and varus of the deformity. The manipulation position is held in a long leg plaster of paris cast and the cast is typically changed weekly. A percutaneous tenotomy of the Achilles tendon is usually performed to correct the residual equinus. The maintenance phase involves the use of a foot abduction brace (FAB) for 23 h a day for three months, followed by nightly use until four to five years of age [5].

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Many classification systems have been proposed to assess the severity of the clubfoot deformity and to measure the impact of treatment [6]. Ponseti and Smoley [4] based their classification on clinical assessment of ankle dorsiflexion, heel varus, forefoot supination and tibial torsion after treatment. Feet were classified as good, acceptable or poor. Harrold and Walker [7] considered the extent of deformity correction. The Pirani score [8] and the Dimeglio score [9] are two of the most widely used classification systems for clubfoot deformity [10]. The Pirani score is from 0 to 6 where zero is a normal foot and six is the most severe deformity. It is reliable when used by non-specialist health workers [11]. The Dimeglio score has a maximum of 20 points and the deformity is graded as benign, moderate, severe or very severe.

Tools that have been developed to assess function include: assessment of patient satisfaction and pain, gait, heel position and range of motion [12, 13]; a questionnaire designed to measure overall satisfaction, foot appearance, pain and physical limitations [14]; and a detailed assessment of movement quality that requires mobility testing with a goniometer and muscle testing [15], but does not include parent reported outcomes.

There is a need for a standardised approach to report clubfoot treatment outcomes [16–18]. To address this gap, this review aims to investigate the literature and to determine and evaluate how success with the Ponseti method is reported in sub-Saharan Africa.

Methods

Search strategy

A systematic literature search was conducted in August 2017 for peer-reviewed articles presenting original research findings on the effect of treatment of clubfoot in children in sub-Saharan Africa. Studies were limited to outcomes of the Ponseti method as this technique is widely accepted as best practice [18]. There was no language restriction. Results are presented according to the PRISMA guidelines [19].

Excerpta Medica Database (EMBASE), Global Health, Medline, Africa Wide Information and African Journals Online were examined for studies meeting the following inclusion criteria: [1] evaluation of the effect of clubfoot management, [2] use of the Ponseti method, [3] original study undertaken in sub-Saharan Africa, and [4] published between 1st January 2000 and 1st August 2017. Concepts were expanded to include related terms and synonyms. A study was excluded if there was no evaluation of treatment, however there was no restriction on type of study to allow a quality assessment review. There was no limitation on age of children and the search was restricted by date (2000–2017) to capture current best practice. Full search terms are presented in Table 1 and the search terms for the country names are outlined in detail in Additional file 1.

Table 1 Search terms for treatment of clubfoot with the Ponseti method in sub-Saharan Africa

1	clubf??t or club-f??t or (club ADJ1 foot) or (talipes ADJ1 equinovarus) or (talipes ADJ1 equino-varus) or (congenital ADJ1 talipes ADJ1 equinovarus) or (congenital ADJ1 talipes ADJ1 equino-varus) or CTEV
2	Ponseti
3	Country name in sub-Saharan Africa[a]
4	1 AND 2 AND 3

[a]Outlined in detail in Additional file 1

All titles and abstracts were screened independently by two authors (TS and DM). The full paper was reviewed if selected by either author or if the abstract was absent. In addition, the reference lists of the included articles were screened. Consensus was reached through discussion where there was disagreement on eligibility.

Data extraction

A pilot-tested spread-sheet was used for data extraction from articles that met the inclusion criteria. All characteristics recorded by one author (TS) were reviewed for accuracy by another author (DM). Data extracted included authors, year of publication, type of study, sample size, age of participants, duration of follow up and reported measurement of treatment outcome. Two authors [20, 21] were contacted to provide missing information. Where other forms of treatment were detailed or where a paper included a country outside of sub-Saharan Africa, only data regarding the Ponseti method and from the sub-Saharan African country were extracted.

Assessment of study quality

Full articles that met the eligibility criteria were categorised according to a hierarchy of study methodologies [22] developed to assess intervention strategies used with children with developmental disabilities. Quality of evidence was ranked as:

I. Systematic review of randomised controlled trials (RCTs); RCT with $N > 100$
II. RCT with $N < 100$; Systematic review of cohort studies
III. Cohort studies with concurrent control group; Systematic reviews of case control studies
IV. Case series; Cohort study without concurrent control group; Case-control study
V. Expert opinion; Case study or report; Anecdotal Evidence.

In addition to the levels of evidence, we used a quality measure proposed by Downs and Black [23] to identify methodological strengths and weaknesses of the included studies as there was no limitation on type of study. The quality index is a 27-item checklist designed for use with both observational studies and randomised controlled trials. The index is comprised of five subscales: reporting (ten questions), external validity (three questions), internal validity (bias and confounding) (13 questions), and power (one question). Items are checked as 'yes', 'partially', 'no' or 'unable to determine' depending on the subscale and higher scores indicating higher quality. The maximum score is 32.

Data analysis

The definition of success was determined by the primary outcome reported in the studies or if explicitly stated. There were no studies that were sufficiently homogenous in terms of participants and outcomes to include in a meta-analysis and data were not combined due to methodological and clinical heterogeneity. An integrative review method [15] that included problem identification, data presentation and analysis was used to incorporate results. Summary statistics for the quality measure were calculated and include the mean and range (minimum and maximum).

Results

Search results

A total of seventy-seven articles were identified. Twenty-two studies met the inclusion criteria. The search strategy and reasons for excluding articles are presented in Fig. 1.

Study characteristics

Characteristics of the eligible studies are presented in Table 2 and include children from one day old [21] to 10 years [24].

The quality of evidence that reported outcomes of the Ponseti method in sub-Saharan Africa was low. Studies were included from ten countries in sub-Saharan Africa; studies undertaken in Nigeria and Malawi contributed five papers each. There were three RCTs, all with small sample sizes of less than 100 children. The majority of studies were classed as level IV [22] due to their observational nature.

Definition of success – Primary outcome

All authors described a form of clinical assessment to assess outcome of treatment. Only 14 studies (64%) gave a clear definition of success. The Pirani score was defined

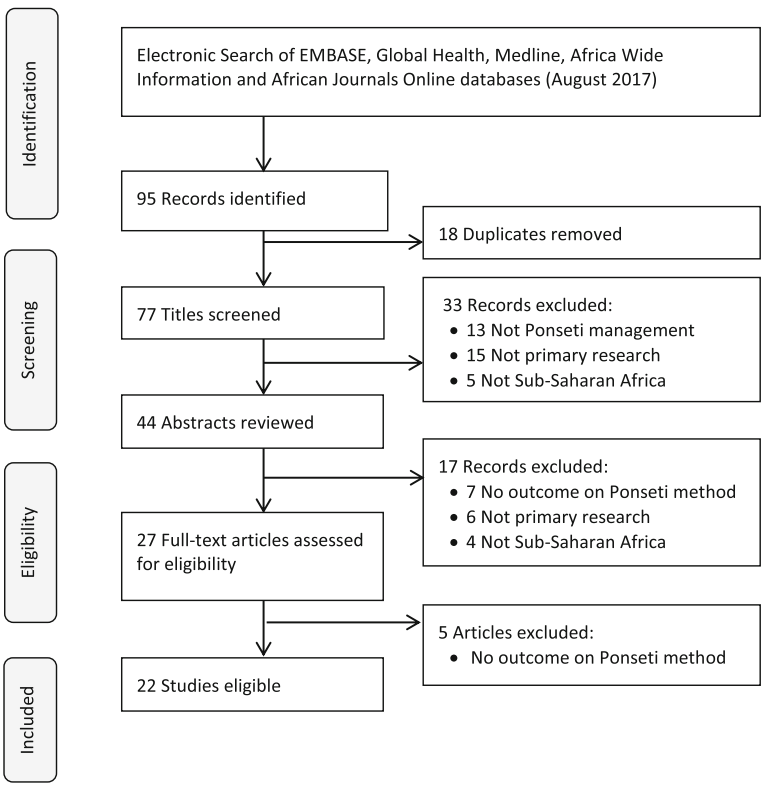

Fig. 1 Search Strategy with PRISMA flow diagram

Table 2 Characteristics of studies that report outcomes of the Ponseti method in sub-Saharan Africa[a]

Primary Author Year Country	Number of children and (feet) treated	Age Range	Type of study (Level of Evidence)	Comparator Group	Duration of Follow up
Ibraheem 2017 [21], Nigeria	23 (14)	<3 months	Randomised controlled trial (II)	Children managed by accelerated Ponseti treatment	32–77 days.
Malagelada 2016 [32], South Africa	65 (91)	4–63 months	Cross sectional survey (IV)	Cases in a UK urban clinic	Not applicable
Smythe 2016 [35], Zimbabwe	173 (268)	17 days – 5 years 7 months	Case series, retrospective (IV)	Pre-treatment status of cases	10.2 weeks (9.5–10.9)
Boakye 2016 [38], Ghana	271 (430)	<6 months	Case series, Retrospective (IV)	Pre-treatment status of cases	Not reported
Adegbehingbe 2015 [39], Nigeria	4931 (7745)	Not reported	Case series (IV)	Pre-treatment status of cases	Not reported
Adewole 2014 [33], Nigeria	106 (158)	7 days – 4 years	Case series, prospective (IV)	Pre-treatment status of cases	Mean: 3 years (range 2–4)
Ayana 2014 [24], Ethiopia	22 (32)	2–10 years	Case series, prospective (IV)	Pre-treatment status of cases	Not reported
Kouamo 2014 [40], Togo	24 (41)	17 days - 7 years	Case series, prospective (IV)	Pre-treatment status of cases	Not applicable
Mang'oli 2014, Kenya	223 (361)	Mean 23 months	Cross sectional survey (IV)	Status of cases at previous appointment	One year
Kaseke 2013 [41], Zimbabwe	14 (20)	Mean 7.43 weeks	Non randomised, prospective (III)	Children managed with Kite technique	6 weeks
Adegbehingbe 2012 [42], Nigeria	493 (749)	Not reported	Case series, prospective (V)	Pre-treatment status of cases	Not reported
Cashman 2012 [20], Malawi	>2000	Not reported	Case series (IV)	No comparator	Not reported
Pirani 2012 [43], Uganda	370	Majority under 14 weeks	Case series, prospective (IV)	Pre-treatment status of cases	Not reported
Harnett 2011 [44], Malawi	21 (32)	<2 months	Randomised controlled trial (II)	Children managed by accelerated Ponseti treatment	Mean 258 days (70 to 348)
Adegbehingbe 2010 [25], Nigeria	55 (80)	<18 years	Randomised controlled trial (II)	Children treated by surgery	3–36 months post last cast
Radler 2010 [45], Mali	52	< 1 year	Case series (IV)	Pre-treatment status of cases	Not reported
Firth 2009 [30], South Africa	70 (106)	1 day – 40 months	Case series, retrospective (IV)	Pre-treatment status of cases	Mean: 2 years 5 months
Biruk 2007 [26], Ethiopia	55 (82)	< 6 months	Case series, prospective (IV)	Children in different age category	Not reported
Lavy 2007 [28], Malawi	307 (482)	<12 months	Case series, retrospective (IV)	Pre-treatment status of cases	Not reported
Khan 2005 [27], South Africa	(61)	Not reported	Case series (IV)	Pre-treatment status of cases	Not reported
Tindall 2005 [29], Malawi	75 (100)	Under 4 years	Case series, prospective (IV)	Pre-treatment status of cases	5 ft followed for 12-18 months
Mkandawire 2003 [36], Malawi	54	Under 2 years	Case series, Prospective (IV)	Pre-treatment status of cases	12 months

[a]Ordered by year of publication

as the primary outcome measure to assess the deformity correction in 14 studies. Change in the mean Dimeglio score was evaluated in one study [25] and frequency of initial severity was reported with the Harrold-Walker classification in two studies [26, 27]. Other definitions of primary outcome included: the number of days in casts [21], number of patients treated without extensive surgery [25], a plantigrade foot [24, 28, 29], no residual deformity [30], deformity status compared to previous visits [31] and parent reported outcomes on impact of treatment [32]. Limited definition terms included "complete correction" [26] and "satisfactory outcome" [25]. The approach to reporting severity scores varied (Table 3).

Process outcomes

There was wide variation in the measurement of process outcomes. The point in treatment when the number of casts was calculated was either before or after the final post tenotomy cast and was inconsistently described. Studies either reported frequency of tenotomy per child or per foot. Definition of relapse or recurrence of deformity differed in the included studies and technical details were only described in five studies (23%).

Six studies report on brace use [25, 28, 30–33] with the focus on non-compliance. Non-compliance was not well defined in the studies and varied from 2% to 44%.

One study assessed parent reported outcomes. The study aimed to determine the impact of the casting and bracing phases of the Ponseti method on the family. Each caregiver completed three questionnaires [32] in order to examine the level of impact that Ponseti treatment had on lives of caregivers and the coping strategies employed.

Reported process outcomes are presented in Table 4.

According to the quality assessment (Additional file 2 outlines the individual study results using the Downs and Black (1998) criteria), the mean quality score of the included studies was 14.8 (5–21).

Reporting

Reporting was the highest scoring category of the quality assessment. All studies included a clear study hypothesis and aim and the majority (17/22) clearly described the characteristics of the patients and the intervention. However, while some distributions of principle confounders were partially described, few studies accounted for confounding in the study design or analysis. Loss to follow up was only reported in half of the studies. Few studies demonstrated a comprehensive attempt to measure adverse effects.

External validity

Many children were recruited from University and tertiary hospitals or national centres and therefore external validity was limited as the interventions undertaken in a specialist centre are likely unrepresentative of the hospitals most of the source population would attend.

Internal validity – Bias and confounding

Randomisation is not possible in cohort studies and in the studies where randomisation was used, it was not possible to determine if the intervention assignment was concealed from both parents and staff until recruitment was complete and irrevocable. Characteristics of losses of patient follow up were inconsistently taken into account and reported in seven (32%) studies. Statistical tests used to assess the main outcomes and why they were chosen were inconsistently described; for example, median, mean and maximum of the number of casts used to achieve correction are reported in different papers. Power calculations were only outlined in three studies.

Discussion

This literature review comprises results from case series, prospective trials and cross-sectional surveys in sub-Saharan Africa. There were few comparative studies concerning the Ponseti method in the region and there were no agreed protocols for reporting the results and outcome of treatment. Due to ethical considerations, most trials investigating treatment of clubfoot are not randomised controlled trials (RCTs) but comparisons of treatments or a review of cohort outcomes. Potential sources of bias in observational studies are well documented [34] and whilst systematic reviews of health care interventions most often focus on RCTs, the inclusion of cohort studies in this review highlights the need for quality design and reporting of studies to increase the strength of evidence.

Principal findings and considerations

A definition of a primary outcome (success) was described in 14 of the 22 studies. Successful outcome ranged from 68% to 98% of cases using different definitions in the 14 studies. There was no consensus on how to define a successful outcome of treatment. There was selective reporting of positive results with little detail given to treatment failure [35]. A range of process measures was included in the studies. The mean number of casts required ranged from 4.6 to 8.7 and is likely affected by the point at which the last cast was measured (pre- or post-tenotomy) and the unlimited age range of the review criteria. The studies used different criteria for relapse recognition and management. Two studies reported patient attrition over 30% [28, 36] however the length of follow-up in the majority of studies was short and few data were available on characteristics of children lost to follow up.

Acknowledging the limitations of the available reported papers, this review suggests that the Ponseti method appears to give successful correction of clubfoot during the correction phase when measured by the Pirani score, Dimeglio classification or simple clinical assessment. However, the lack of a consistent measure of success and insufficient follow up of cases restricts the conclusions that can be made about what happens during the bracing phase, be it success, recurrence or loss to follow-up.

Main findings as related to other publications

The included studies report success in 68% to 98% of cases after the correction (casting) phase. In contrast,

Table 3 Reported Primary Outcome using the Ponseti method in sub-Saharan Africa

Primary Author Year Country*	Clubfoot severity assessment	Reported Success Measure	Recurrence / relapse	Additional surgical intervention
Ibraheem 2017 [21], Nigeria	Pirani score	Number of days in casts, number of casts applied	Not reported	Not reported
Malagelada 2016 [32], South Africa	Pirani score	Parent reported outcomes	12% (8 children)	Not reported
Smythe 2016 [35], Zimbabwe	Pirani Score	85% feet; Pirani score < 1	Not reported	Not reported
Boakye 2016 [38], Ghana	Pirani Score	Number of casts to correction. Correction not defined.	Not reported	Not reported
Adegbehingbe 2015 [39], Nigeria	Not reported	89.7% (4426 patients) satisfactory outcome. Criteria for satisfactory outcome not defined.	4% (253 feet, 194 patients)	3%
Adewole 2014 [33], Nigeria	Pirani score and photograph	100%; based on clinical judgement, Plantigrade functional foot	5.16% (8 feet)	6 feet
Ayana 2014 [24], Ethiopia	Pirani score	28/41 good results Good = correction of all deformities. 97.8% achieved score of <3	2 patients, 4 feet	8 children/ (11 feet)
Kouamo 2014 [40], Togo	Not reported	94% (179/190) compliant with brace wear 93.5% no visible discomfort	12.2% (5 cases)	Not reported
Mang'oli 2014 , Kenya	Pirani score	Initial correction: 96.2% (152 feet) Initial correction not defined.	Not reported	Not reported
Kaseke 2013 [41], Zimbabwe	Pirani score	Rate of correction: Pirani score at 3 weeks and 6 weeks	Not reported	Not reported
Adegbehingbe 2012 [42], Nigeria	Pirani Score	89.7% treated successfully. Criteria for success not defined.	Not reported	3.2% (16 patients)
Cashman 2012 [20], Malawi	Not reported	30 children failed treatment (required more extensive surgery)	Not reported	30 children
Pirani 2012 [43], Uganda	Pirani Score	Mean score 5.4 falls to <2 by cast 6. Primary outcome not defined.	Not reported	Not reported
Harnett 2011 [44], Malawi	Pirani Score	Pirani score change. Median start Pirani: 5 (4 to 6). Median at tenotomy /end treatment: 0.5 (0.5 to 1) Median at 6 months: 0.5 (0 to 0.5)	No episodes of recurrence after 6 months	3 patients not corrected (7%) with Pirani >1
Adegbehingbe 2010 [25], Nigeria	Dimeglio classification	96.4% (53/55 children) = satisfactory (No recurrence) 3.6% (2/55) = fair (recurrence corrected with casts/FAB) Nil = poor (recurrence with repeat surgery)	2 had recurrence between 4 and 6 months	None
Radler 2010 [45], Mali	Not reported	77% (40 children): good or average. 23% (12 children): poor. Primary outcome not defined.	Not reported	Not reported
Firth 2009 [30], South Africa	Pirani score	61% fully corrected without residual deformity	23% (re-plaster 24 feet) 39% (41 feet mild recurrence)	7% (7 feet)
Biruk 2007 [26], Ethiopia	Harrold-Walker classification	76.8% (63 feet) No definition of complete correction.	Not reported	Not reported for Ponseti cohort
Lavy 2007 [28], Malawi	Pirani score	68% (327/482) Plantigrade or better	Not reported	12 children referred for surgery
Khan 2005 [27], South Africa	Harrold-Walker classification	6 failures from 61 feet. Criteria for success not defined.	Not reported	Not reported
Tindall 2005 [29], Malawi	Pirani score	98% plantigrade foot with Pirani score	Not reported	2%
Mkandawire 2003 [36], Malawi	Pirani score	Correction of deformity. Success of correction defined as fitting brace. Mean Pirani score decreased from 3.6–0.86	4 children with untreated clubfoot, 5 with complex and 7 with teratologic	Not reported

*Ordered by year of publication

Table 4 Outcomes of the Ponseti Method reported in sub-Saharan Africa[a]

Primary Author (Year) Country	Process Outcomes						
	Average number of casts	Duration of casts	Percutaneous Achilles Tenotomy	Receipt of braces	Brace compliance	Loss to follow up	Complications
Ibraheem (2017 Nigeria	5.43	52 days (35–77)	1 child did not have tenotomy, not reported case or control	100%	Not reported	Nil	Reported no complications with swelling
Malagelada (2016) South Africa	8.7 (range 1–24)	Not reported	89% (58 children)	100% due to inclusion criteria	2% (1 child) non-compliant	Not applicable	Defined as relapse and non-compliance: 9 children
Smythe (2016) Zimbabwe	7.27 (6.7–7.9)	10.2 (9.5–10.9) weeks included tenotomy	78.9% (127/161 children)	Not reported	Not reported	8.9% (17 children)	Not reported
Boakye (2016) Ghana	4.93	Not reported	77%	Not reported	Not reported	Excluded from analysis	Not reported
Adegbehingbe (2015) Nigeria	Not reported	Not reported	77% (5626 children)	Not reported	Not reported	Not reported	Not reported
Adweole (2014) Nigeria	4.6 (range 3–9)	Weekly cast change, tenotomy 3 weeks	26.6% (42 feet)	56.8% (60 patients)	No child with relapse wore braces	Not reported	9 feet: cast complications, blisters, ulcers, skin rash
Ayana (2014) Ethiopia	8 (range 6–10)	Casts changed every 2 weeks	63.6% (14 children, 21 feet)	100%; < 4 yrs. = FAB >4 yrs. = ankle foot orthosis	Not reported	1 patient	No major complications
Kouamo (2014) Togo	Not reported	Not reported	82.9% (34/41 feet)	Not reported	Not reported	Not reported	Not reported
Mangoli (2014) Kenya	Not reported	Not reported	Not reported	100% of interviewed parents	15% (33/223) non-compliant Mean use 18 months (6–23)	Not applicable	5% (11/223) skin lesion
Kaseke (2013) Zimbabwe	Not reported	Not reported	Not reported	Not reported	Not reported	6 feet not reported at 6 weeks	Not reported
Adegbehingbe (2012) Nigeria	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported
Cashman (2012) Malawi	Not reported	Not reported	>80%	Not reported	Not reported	107 children	Not reported
Pirani (2012) Uganda	Not reported	Majority corrected by 6th treatment'	Not reported	Not reported	Not reported	83% adherence rate to end of casting	Plaster burns in 19/1000
Harnett (2011) Malawi	Median 5 (4–7)	42 days (35–84) in plaster prior to tenotomy.	52% (11 children)	Given FAB to wear until 3 years old	Not reported	2 after plaster. 1 patient died	Not reported
Adegbehingbe (2010) Nigeria	≤ 6 (76.4%; range 2–6) >6 (23.6% range 7–10)	2.3–13.7 +/−1.7 weeks	5.5% (3 children)	Not reported	Noted as 'generally good'	None, not explicitly mentioned	3.6% ugly scar, recurrence, blister, infection
Radler (2010) Mali	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported
Firth (2009) South Africa	6.5 (range 2–18)	Not reported	74% (78 feet)	Received FABs, % unspecified	16% (11 patients) non-compliant	Not reported	8% (9 feet) minor blistering from braces
Biruk (2007) Ethiopia	Maximum cast 17 times	Weekly cast change	Not reported	60%, average wait time 3-4 months	Not reported	Not reported	Not reported for Ponseti cohort
	Not reported	Not reported	37% had tenotomy				

Table 4 Outcomes of the Ponseti Method reported in sub-Saharan Africa[a] *(Continued)*

Primary Author (Year) Country	Process Outcomes						
	Average number of casts	Duration of casts	Percutaneous Achilles Tenotomy	Receipt of braces	Brace compliance	Loss to follow up	Complications
Lavy (2007) Malawi				44% given FABs	44% (145/327 feet)	32% (155 feet)	307 adequate records
Khan (2005) South Africa	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported
Tindall (2005) Malawi	5.3	Mean treatment 9.1 weeks	41%	All	Not reported	Not reported	2 minor complications
Mkandawire (2003) Malawi	Weekly cast change	Mean treatment: 7.4 weeks for idiopathic, 7.1 weeks for complex	Not reported	Not reported	Not reported	32 patients (35%)	Not reported

[a]Ordered by year of publication

global success rates after the correction phase are cited as approximately 90% [18, 37]. Comprehensive tools to assess function (e.g. as described by Laaveg and Ponseti (12), the Roye tool [14], the Bangla tool [13] or the Clubfoot Assessment Protocol (CAP) [15]) are not reported in the studies from sub-Saharan Africa.

Implications of findings

We found that the differences between study populations, methodology and the way that outcomes are described contribute to the variation in results reported for the Ponseti method in sub-Saharan Africa. Currently, different scores are used for the assessment of clubfoot severity. Standardisation is required to define successful outcome of clubfoot management so that risk factors for good and poor outcome can be determined and services can be monitored and evaluated.

The Pirani score was the most frequent clinical assessment used. It has been validated in younger children and demonstrates acceptable interrater reliability [8]. A short assessment time is required and it is easy to use, however to ensure consistency more guidance would be helpful on how to measure the individual components, as similarly provided by the diagrams and video produced to aid assessment with the Dimeglio score. The Pirani scoring system is the only assessment that has evidence for use by paramedics, and is in our opinion the easiest severity measure to use in young children before walking age.

Methodologic issues

To our knowledge, this is the first systematic review of outcomes to measure success of the Ponseti method in sub-Saharan Africa. The observation of explicit methodology and lack of language restriction are strengths of this study. The literature available to assess success of clubfoot treatment is characterised by a lack of standardisation of outcomes. Studies routinely use the term "success rates" but do not define a successful outcome. Given that Ponseti management involves both correction and maintenance, the definition of success should always reflect both of these important endpoints and we encourage researchers to measure and report both. Bias in internal validity arose from studies where differences in follow up were regularly ignored, however compliance with the corrective phase of the intervention was generally reported as being good. Studies must include follow-up or acknowledge the limitations of selecting one part of the treatment process.

The potential for confounding in the reviewed studies to obscure true effects is significant as the majority are observational. Randomisation may be considered unethical in certain circumstances and well designed controlled trials may provide more opportunities to analyse different outcomes. Studies intended to address comparative effectiveness of management for clubfoot should use a careful control for covariates such as unilateral or bilateral clubfoot as disproportionate weighting is given to bilateral cases [17].

Research gaps

Although a number of studies are available on initial treatment (correction phase) outcomes, very few studies are available on long term outcomes and follow up in the bracing phase, which are essential for measuring success of the entire Ponseti method.

No study compared different scoring systems. A study comparing multiple assessments in the same patient before and after treatment would be of value in assessing the equivalence or superiority of measurement techniques.

Studies need to control for the side of clubfoot and previous treatment, account for loss to follow up and adjust for confounding in methods or analysis in order

to avoid the shortfalls of the current observational literature.

Recommendations

Consensus is needed to standardise the reporting of outcomes and how success after Ponseti management is defined. For sub-Saharan Africa the definition needs to be appropriate for use by trained therapists who are managing children with clubfoot. This systematic review contributes to the knowledge about the importance of providing evidence to improve clubfoot services.

Conclusions

The lack of good quality studies, variation in definition of success and limited follow-up of patients means the success rate of clubfoot treatment using the Ponseti method in sub-Saharan Africa is uncertain. There is need for an agreed definition of good outcome following both the correction and the bracing phase to monitor and evaluate service delivery and identify reasons for poor outcome. It is very important that children who complete the correction phase are followed through the bracing phase and results on success, recurrence and loss to follow up are reported. Studies are also required to document the correlation between clinical outcome, functional outcome and patient/family reported satisfaction.

Additional files

Additional file 1: Expanded search terms for country name in sub-Saharan Africa. (DOCX 13 kb)

Additional file 2: Quality index assessment for included studies (studies 1–11 assessed on pages 1–3 and studies 12–22 assessed on pages 4–6). (DOCX 35 kb)

Abbreviations

CTEV: Congenital Talipes Equinovarus; FAB: Foot Abduction Brace; PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses

Acknowledgements

Professor Daniel Chandramohan for early concept considerations.

Funding

The Beit Trust and CBM are gratefully acknowledged for funding provided to TS.

Availability of data and materials

Data sharing not applicable to this article as no datasets were generated or analysed during the current study. Two web appendices are attached: detailed search terms and quality assessment scores.

Author's contributions

TS conceived the study. TS AF and CL designed the study protocol. TS and DM searched the literature and extracted data for analysis. TS analysed and interpreted the extracted information. AF CL HK and DM critically revised the manuscript for intellectual content. All authors read and approved the final manuscript.

Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

Publisher's Note

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Received: 8 August 2017 Accepted: 7 November 2017
Published online: 15 November 2017

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Appendix 1.Expanded search terms for country name in sub-Saharan Africa

1. "africa south of the sahara"/ or africa, central/ or africa, eastern/ or africa, southern/ or africa, western/

2. ("africa south of the sahara" or sub-saharan africa or central africa or eastern africa or southern africa or western africa).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. Benin/
4. (Benin or Dahomey).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. Burkina Faso/
6. (Burkina Faso or Burkina Fasso or Upper Volta).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. Burundi/
8. Burundi.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
9. Central African Republic/
10. (Central African Republic or Ubangi-Shari).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
11. Chad/
12. Chad.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
13. Comoros/
14. (Comoros or Comoro Islands or Mayotte or Iles Comores).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
15. "Democratic Republic of the Congo"/
16. ((democratic republic adj2 congo) or belgian congo or zaire).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
17. Eritrea/
18. Eritrea.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
19. Ethiopia/
20. Ethiopia.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
21. Gambia/
22. Gambia.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
23. Guinea/
24. (Guinea not (New Guinea or Guinea Pig* or Guinea Fowl)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
25. Guinea-Bissau/
26. (Guinea-Bissau or Portuguese Guinea).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
27. Kenya/
28. Kenya.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
29. Liberia/
30. Liberia.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
31. Madagascar/

32. (Madagascar or Malagasy Republic).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
33. Malawi/
34. (Malawi or Nyasaland).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
35. Mali/
36. Mali.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
37. Mozambique/
38. (Mozambique or Portuguese East Africa).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
39. Niger/
40. (Niger not (Aspergillus or Peptococcus or Schizothorax or Cruciferae or Gobius or Lasius or Agelastes or Melanosuchus or radish or Parastromateus or Orius or Apergillus or Parastromateus or Stomoxys)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
41. Rwanda/
42. (Rwanda or Ruanda).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
43. Sierra Leone/
44. Sierra Leone.mp.
45. Somalia/
46. Somalia.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
47. Tanzania/
48. (Tanzania or Zanzibar).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
49. Togo/
50. (Togo or Togolese Republic).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
51. Uganda/
52. Uganda.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
53. Zimbabwe/
54. (Zimbabwe or Rhodesia).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
55. or/3-54
56. Cameroon/
57. Cameroon.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
58. Cape Verde/
59. (Cape Verde or Cabo Verde).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
60. Congo/
61. (congo not ((democratic republic adj3 congo) or congo red or crimean-congo)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
62. Cote d'Ivoire/
63. (Cote d'Ivoire or Ivory Coast).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
64. Ghana/
65. (Ghana or Gold Coast).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]

66. Lesotho/
67. (Lesotho or Basutoland).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
68. Mauritania/
69. Mauritania.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
70. Nigeria/
71. Nigeria.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
72. Atlantic Islands/
73. (sao tome adj2 principe).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
74. Senegal/
75. Senegal.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
76. Sudan/
77. Sudan.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
78. South Sudan.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
79. Swaziland/
80. Swaziland.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
81. Zambia/
82. (Zambia or Northern Rhodesia).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
83. or/56-80
84. Angola/
85. Angola.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
86. Botswana/
87. (Botswana or Bechuanaland or Kalahari).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
88. Gabon/
89. (Gabon or Gabonese Republic).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
90. Mauritius/
91. (Mauritius or Agalega Islands).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
92. Namibia/
93. Namibia.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
94. Seychelles/
95. Seychelles.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
96. South Africa/
97. South Africa.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
98. or/84-97
99. Equatorial Guinea/

100. (Equatorial Guinea or spanish guinea).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
101. 99 or 100
102. 1 or 2 or 55 or 83 or 98 or 101

Appendix 2: Quality Index assessment for included studies (studies 1-11 assessed on pp 1-3 and studies 12-22 assessed on pp 4-6)

Quality Index (Downs & Black, 1998, pp. 382-383) (papers 1- 11)	Ibraheem 2017, Nigeria	Malagelada 2016, SouthAfrica	Smythe 2016, Zimbabwe	Boakye 2016, Ghana	Adegbehingbe 2015, Nigeria	Adewole 2014, Nigeria	Ayana 2014, Ethiopia	Kouamo 2014, Togo	Mang'oli 2014, Kenya	Kaseke 2013, Zimbabwe	Adegbehingbe 2012, Nigeria
Reporting: Were the following clearly described? (Y/N)											
1. Study hypothesis/aim/objective	1	1	1	1	1	1	1	1	1	1	1
2. Main outcomes	1	1	1	0	0	1	1	1	0	1	0
3. Characteristics of the participants	1	1	1	1	0	1	1	1	1	0	1
4. Interventions of interest	1	1	1	1	0	1	1	1	1	0	0
5. Distributions of principal confounders in each group	1	1	1	1	0	1	0	0	0	0	0
6. Main findings	1	1	1	1	1	1	1	1	1	1	1
7. Estimates of random variability for main outcomes	1	1	1	1	0	1	1	0	0	1	0
8. All the important adverse events that may be a consequence of intervention are reported	0	0	0	0	0	1	1	0	0	0	0
9. Characteristics of patients lost to follow-up	1	1	1	0	0	0	1	0	1	0	0
10. Actual probability values for main outcomes	1	1	1	1	0	0	0	0	0	1	1
External validity (Y/N/unable to determine)											
11. Were subjects who were asked to participate representative of the entire population from which they were recruited?	1	0	1	1	1	1	1	0	0	1	1
12. Were subjects who were prepared to participate representative of the entire	1	0	1	1	1	1	1	0	0	1	1

population from which they were recruited?											
13. Were the staff, places, and facilities representative of the treatment the majority of subjects received?	0	0	0	0	1	0	0	0	1	0	0
Internal validity – bias (Y/N/unable to determine)											
14. Was an attempt made to blind subjects to the intervention they received?	0	1	0	0	0	0	0	0	0	0	0
15. Was an attempt made to blind those measuring main outcomes of the intervention?	0	0	0	0	0	0	0	0	0	0	0
16. If any of the results of the study were based on “data dredging” was this made clear?	1	1	1	1	0	1	1	1	1	1	1
17. In trials and cohort studies, do analyses adjust for different lengths of follow-up? Or, in case-control studies, is the period between intervention and outcome the same for cases and controls?	1	1	1	1	0	1	1	1	1	1	1
18. Were appropriate statistical tests used to assess the main outcomes?	1	1	1	1	0	1	1	1	1	1	1
19. Was compliance with the intervention reliable?	1	1	1	1	0	1	1	1	1	0	1
20. Were main outcome measures reliable and valid?	1	1	1	1	0	1	1	1	1	0	1
Internal validity – confounding (selection bias) (Y/N/unable to determine)											
21. For trials and cohort studies, were patients in different intervention groups?	1	1	1	1	1	1	1	1	1	1	1

For case-control studies, were cases and controls recruited from the same population?											
22. Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?	1	0	1	1	0	1	1	1	1	0	1
23. Were subjects randomized to intervention groups?	1	0	0	0	0	0	0	0	0	0	0
24. Was the randomized intervention assignment concealed from both patients and staff until recruitment was complete and irrevocable?	0	0	0	0	0	0	0	0	0	0	0
25. Was there adequate adjustment for confounding in the analyses from which main findings were drawn?	0	0	1	0	0	0	0	0	0	0	0
26. Were losses of subjects to follow-up taken into account?	1	1	1	1	0	0	1	0	1	0	0
Power											
27. Did the study have sufficient power to detect a clinically important effect where the probability for a difference due to chance was less than 5%?	0	0	0	0	0	0	0	0	0	1	0

Quality Index (Downs & Black, 1998, pp. 382-383) (papers 12-22)	Cashman 2012, Malawi	Pirani 2012, Uganda	Harnett 2011, Malawi	Adegbehingbe 2010, Nigeria	Radler 2010, Mali	Firth 2009, South Africa	Biruk 2007, Ethiopia	Lavy 2007, Malawi	Khan 2005, South Africa	Tindall 2005, Malawi	Mkandawire 2003, Malawi
Reporting: Were the following clearly described? (Y/N)											
1. Study hypothesis/aim/objective	1	1	1	1	1	1	1	1	1	1	1
2. Main outcomes	1	0	1	1	0	1	0	1	0	1	1
3. Characteristics of the participants	0	1	1	1	0	1	1	1	0	1	1
4. Interventions of interest	0	1	1	1	1	1	1	1	1	1	1
5. Distributions of principal confounders in each group	0	1	1	1	0	1	0	1	0	1	1
6. Main findings	1	1	1	1	1	1	1	1	0	1	1
7. Estimates of random variability for main outcomes	0	0	1	1	0	1	1	1	0	1	1
8. All the important adverse events that may be a consequence of intervention are reported	0	1	1	1	0	1	1	0	0	1	0
9. Characteristics of patients lost to follow-up	0	0	1	0	0	0	0	0	0	1	0
10. Actual probability values for main outcomes	0	0	1	1	0	1	0	0	0	0	0
External validity (Y/N/unable to determine)											
11. Were subjects who were asked to participate representative of the entire population from which they were recruited?	1	1	1	1	0	0	1	1	0	1	1
12. Were subjects who were prepared to participate representative of the entire	1	1	1	1	0	0	1	0	0	1	1

population from which they were recruited?											
13. Were the staff, places, and facilities representative of the treatment the majority of subjects received?	1	1	0	0	0	0	0	1	0	0	0
Internal validity – bias (Y/N/unable to determine)											
14. Was an attempt made to blind subjects to the intervention they received?	0	0	0	1	0	0	0	0	0	0	0
15. Was an attempt made to blind those measuring main outcomes of the intervention?	0	0	1	1	0	0	0	0	0	0	0
16. If any of the results of the study were based on “data dredging” was this made clear?	1	1	1	1	1	1	1	1	1	1	1
17. In trials and cohort studies, do analyses adjust for different lengths of follow-up? Or, in case-control studies, is the period between intervention and outcome the same for cases and controls?	0	1	1	1	1	0	0	1	1	1	1
18. Were appropriate statistical tests used to assess the main outcomes?	0	1	1	1	0	1	1	1	1	1	1
19. Was compliance with the intervention reliable?	0	1	1	1	0	1	0	0	0	1	0
20. Were main outcome measures reliable and valid?	0	1	1	1	0	1	0	1	0	1	1
Internal validity – confounding (selection bias) (Y/N/unable to determine)											
21. For trials and cohort studies, were patients in different intervention groups? For case-control studies, were cases and	1	1	1	1	1	1	1	1	0	1	1

controls recruited from the same population?											
22. Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?	1	1	1	1	1	1	1	1	0	1	1
23. Were subjects randomized to intervention groups?	0	0	1	1	0	0	0	0	0	0	0
24. Was the randomized intervention assignment concealed from both patients and staff until recruitment was complete and irrevocable?	0	0	0	0	0	0	0	0	0	0	0
25. Was there adequate adjustment for confounding in the analyses from which main findings were drawn?	0	0	0	0	0	0	0	0	0	0	0
26. Were losses of subjects to follow-up taken into account?	0	0	1	0	0	0	0	0	0	1	1
Power											
27. Did the study have sufficient power to detect a clinically important effect where the probability for a difference due to chance was less than 5%?	0	0	1	0	0	0	1	0	0	0	0

Epilogue (Main findings and limitations)

This systematic review found that only 64% (14/22) of included studies described a primary outcome. Clinical assessment was the most commonly reported outcome measure, and few studies reported long-term outcome. The most frequently used measure to define outcome was the Pirani score, however there was no agreement on cut-off score for success.

Outcomes were predominantly reported though case series. These different outcome measures reported success in 68% to 98% of cases after the correction (manipulation and casting) phase.

The differences between study populations, methodology and the way that outcomes are described contribute to the variation in results reported for the Ponseti method in sub-Saharan Africa.

This review is limited by the lack of standardisation of outcomes in the available literature, and the insufficient follow up of cases, which restricts the conclusions that can be made about what happens during the bracing phase.

There is a high risk of internal validity bias in the included studies; differences in follow up were regularly ignored. The potential for confounding to obscure true effects of treatment in the studies is also high as the majority of included studies were observational.

Many children were recruited from University and tertiary hospitals or national centres, which limits external validity, as the interventions undertaken in a specialist centre are not likely to represent the hospitals that most of the population would attend.

Acknowledging these limitations, this review suggests that the Ponseti method gives successful correction of clubfoot when measured by the Pirani score, Dimeglio classification or simple clinical assessment during the correction phase.

Chapter 6. Results of clubfoot treatment after manipulation and casting using the Ponseti method: experience in Harare, Zimbabwe

Assessment of a child with clubfoot

Preamble

Parirenyatwa hospital in Harare, Zimbabwe, has provided clubfoot care as recommended by the Ponseti method since 22 March 2011. The outcomes of the Ponseti manipulation and casting method of clubfoot treatment have not been evaluated in any clinic in Zimbabwe, and predictors of these outcomes are unknown.

Evidence from case series, prospective trials and cross-sectional surveys in sub-Saharan Africa reported in the systematic review (Chapter 5) informed this study. The systematic review found that the Pirani score was the most frequent measure to define outcome. The Pirani score was therefore used as the standard reference to assess the outcome of Ponseti treatment for clubfoot in this case series.

The case series included 218 children being treated for idiopathic clubfoot at Parirenyatwa Hospital's clubfoot clinic in Harare over more than two years (22 March 2011 to 23 April 2013 (25 months)). Data were extracted retrospectively from clinic records and analysed.

This chapter reports the short-term (only after the manipulation and casting phase) outcomes of clubfoot treatment, and explores the predictors of these results. It comprises a published study that examines the outcomes of the corrective phase for clubfoot as recommended by the Ponseti method in one tertiary clinic in Harare.

This research paper was published in the journal Tropical Medicine & International Health in October 2016 after peer review (epublication: July 2016).

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?	Tropical Medicine and International Health		
When was the work published?	8th July 2016		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion	n/a		
Have you retained the copyright for the work?*	The publication is covered by a Creative Commons Attribution CCBY Creative Commons Licence. Anyone may copy, distribute, or reuse the content as long as the author and original source are properly cited	Was the work subject to academic peer review?	Yes

**If yes, please attach evidence of retention. If no, or if the work is being included in its published format, please attach evidence of permission from the copyright holder (publisher or other author) to include this work.*

SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	
Please list the paper's authors in the intended authorship order:	
Stage of publication	.

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I co-designed the study, extracted the data, completed data analysis, interpreted the data with co-authors, drafted the manuscript, prepared the subsequent revisions with consideration of comments from co-authors

Student Signature: [redacted] **Date:** 9/5/18

Supervisor Signature: [redacted] **Date:** 9/5/18

Tropical Medicine and International Health doi:10.1111/tmi.12750

VOLUME 21 NO 10 PP 1311–1318 OCTOBER 2016

Results of clubfoot treatment after manipulation and casting using the Ponseti method: experience in Harare, Zimbabwe

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3 *Oxford NIHR Musculoskeletal Biomedical Research Unit, University of Oxford, Headington, UK*

Abstract

OBJECTIVES The objective of this study was to evaluate the outcomes of the Ponseti manipulation and casting method for clubfoot in a tertiary hospital in Zimbabwe and explore predictors of these outcomes.

METHODS A cohort study included children with idiopathic clubfoot managed from 2011 to 2013 at Parirenyatwa Hospital. Demographic data, clinical features and treatment outcomes were extracted from clinic records. The primary outcome measure was the final Pirani score (clubfoot severity measure) after manipulation and casting. Secondary outcomes included change in Pirani score (pre-treatment to end of casting), number of casts for correction, proportion receiving tenotomy and proportion lost to follow up.

RESULTS A total of 218 children (337 feet) were eligible for inclusion. The median age at treatment was 8 months; 173 children (268 feet) completed casting treatment within the study period. The mean length of time for corrective treatment was 10.2 weeks (9.5–10.9 weeks). Of the 45 children who did not complete treatment, 28 were under treatment and 17 were lost to follow up. A Pirani score of 1 or less was achieved in 85% of feet. Mean Pirani score at presentation was 3.80 (SD 1.15) and post-treatment 0.80 (SD 0.56, *P*-value <0.0001). Severity of deformity and being male were associated with a higher (worse) final Pirani score. Severity and age over two were associated with an increase in the number of casts required to correct deformity.

CONCLUSION This case series demonstrates that the majority (80%+) of children with clubfoot can achieve a good outcome with the Ponseti manipulation and casting method.

keywords clubfoot, congenital talipes equinovarus, Zimbabwe, Ponseti, treatment, cohort analysis

Introduction

Congenital talipes equinovarus (CTEV), a deformity commonly called clubfoot, is a complex congenital condition affecting the foot. It presents with varying degrees of rigidity that is not passively correctable [1]. Untreated clubfoot results in pain and disability [2]. The incidence of clubfoot varies globally [3] and it is estimated that 80% of children born with clubfoot each year live in low- and middle-income countries (LMICs) [4]. Boys are affected 2.5 times as often as girls, and the condition is bilateral in half of the cases [5]. The causes of clubfoot are poorly understood, but may include genetic factors [6].

The Ponseti method of conservative clubfoot management is considered the best practice [7] and avoids corrective surgery in over 90% of cases [8]. Use of a primarily non-operative technique is beneficial in LMICs where there are limited resources and different cadres of health workers can be trained to treat clubfoot [9].

The Ponseti method is divided into two distinct phases: the corrective phase and the maintenance phase. The corrective phase involves manipulation of the foot followed by casting with plaster of paris. The cast holds the stretch achieved through manipulation of tight structures and allows time for soft tissue remodelling and correction of the position of the bones in the foot [10]. Sequential correction of the cavus, adductus and varus of clubfoot occurs around the talus [8]. A percutaneous Achilles tenotomy is usually required to correct the residual equinus (downward position of the foot), followed by 3 weeks in a cast to assist healing [11].

The maintenance phase consists of a bracing regime to prevent recurrence. The foot abduction brace is worn 23 h a day for the first 3 months and then at night during sleep until five years of age [11].

This study examines the outcomes of the corrective phase (manipulation and casting) for clubfoot as recommended by the Ponseti method in a cohort of children undergoing treatment at one referral hospital in Zimbabwe.

Methods

Ethical approval for this study was granted by the Joint Research Ethics Committee for the University of Zimbabwe, College of Health Sciences and Parirenyatwa Group of Hospitals, and the ethics committees at the Medical Research Council of Zimbabwe and the London School of Hygiene and Tropical Medicine.

Study design

A cohort included children being treated for idiopathic clubfoot at Parirenyatwa Hospital's clubfoot outpatient clinic in Harare from 22 March 2011 to 23 April 2013 (25 months). All children with a diagnosis of unilateral or bilateral idiopathic clubfoot corrected by the Ponseti method at the study hospital were eligible. There was no restriction on previous treatment for clubfoot. The only exclusion criterion was foot conditions other than idiopathic clubfoot, for example clubfoot associated with a syndrome or comorbidity, such as spina bifida.

Children aged under 2 years. The corrective phase of the Ponseti method followed the kinematic principles of correction [10] and involved weekly cast changes. The knee was fixed at 90 degrees in the long leg plaster of paris casts (toe to groin). Physiotherapists performed the manipulation and casting, and casts were removed by parents at the clinic in the morning of the appointment. Medical officers performed a percutaneous Achilles tenotomy with local anaesthetic when indicated by clinical assessment. The foot was held in a long leg plaster cast in abduction and dorsiflexion for 3 weeks post-tenotomy.

The maintenance phase used the Steenbeek foot abduction brace [12] to hold both feet in seventy degrees abduction. The brace was made locally and supply was regular. The brace regime was initiated on the day of removal of the final cast.

Children aged over 2 years. Children over 2 years were treated with the modified principles of the Ponseti method as recommended for older children [13]. Abduction of thirty to forty degrees was achieved with manipulation and long leg plaster casts with seventy degrees of knee flexion. The casts were changed weekly. A percutaneous Achilles tenotomy was performed for all children aged over 2 years with residual equinus and the cast post-tenotomy remained for 4–6 weeks. In the maintenance phase, the foot abduction brace held both feet in thirty degrees of abduction.

The key variables recorded on the clinic records by physiotherapists and extracted by the researchers, included child's date of birth, gender, diagnosis, order of birth, mother's date of birth, place of birth, family history of clubfoot, side of affected foot, history of previous treatment and weekly recording of Pirani score. The Pirani score is a classification system for assessing severity of clubfoot based on clinical examination [14] and was scored by the clinic physiotherapists. A decrease in score correlates to improvement in deformity. The Pirani score is from 0 to 6 where zero is a normal foot and six is the most severe deformity. Scoring is performed at half-point intervals.

Data management and analysis

Data were managed and analysed using Stata 14.1, StataCorp 4905, Lakeway Drive College Station, Texas 77845, USA. A hierarchical conceptual framework for analysis was developed [15]. The primary outcome measure was defined as Pirani score at the end of the manipulation and casting phase, with success determined as a score of 1 or less. The following secondary outcomes were defined: (i) the change in Pirani score from pre-treatment (first manipulation and casting appointment) to post-treatment (first brace fitting); (ii) number of casts required for correction, inclusive of the final cast after tenotomy; (iii) proportion having a tenotomy; (iv) proportion lost to follow up (defaulted appointment during casting and was not seen again).

Three factors that may influence the Ponseti manipulation and casting process were identified *a priori*: age at first treatment, history of previous treatment and pre-treatment severity of deformity. Age at first treatment and a combination of treatments may affect the extent of clubfoot correction [16]. Response to casting may be influenced by initial presenting severity [4, 17]. Gender was included as a fourth possible predictor according to the hierarchical conceptual framework.

Age at first treatment was defined as a binary variable ($\leq$2 or >2 years) to allow comparison to findings from the literature. An ordered categorical variable for pre-treatment severity of deformity was created based on Pirani score before treatment. An increase of 1 in the categorical variable represents an increase of 0.5 in the Pirani score.

Statistical methods

The associations between outcomes and gender, age at first treatment, history of previous treatment and severity

category were explored. Statistical significance for the case series was set at the 95% confidence level.

The outcomes of Pirani score (final and change in score) were analysed by feet. The outcomes of number of casts, tenotomy performed and loss to follow up were analysed by child.

Unilateral and bilateral clubfeet were included in analysis of the Pirani score. As right and left feet are correlated in the same child [18], a random effects model was used to assess the association between potential predictors and the Pirani score. The random effects model means that the right and left feet on the same child were not treated as independent. A linear regression model was fitted for the outcome of number of casts. This was possible on an individual level as children with bilateral clubfeet had the same number of casts applied per foot to enable both tenotomies to be performed on the same day. A logistic regression model was fitted to assess factors associated with the binary variables of tenotomy performed and loss to follow up. The models assumed no effect modification.

A second model was constructed with a backward elimination approach to identify a model of best fit and the joint predictive capability of the variables [19]. Any variable that affected the primary outcome by less than 10% was dropped and not considered as a confounder [19]. Further adjustment through backward elimination did not change the outcome of the first random effects model and is not included in the results.

Results

Characteristics

A total of 218 children (337 feet) with a diagnosis of idiopathic clubfoot presented for treatment (Figure 1); 28 children were excluded from analysis as they had yet to complete the manipulation and casting treatment, and of the remaining 190 children, 17 (8.9%) were lost to follow up during the casting period. Loss to follow up was not associated with gender, age at first treatment, previous treatment or severity; 173 children completed treatment within the study time period and continued to the bracing phase (Table 1). Mean length of time for the corrective phase was 10.2 weeks (9.5–10.9 weeks) and is inclusive of the final cast post-tenotomy.

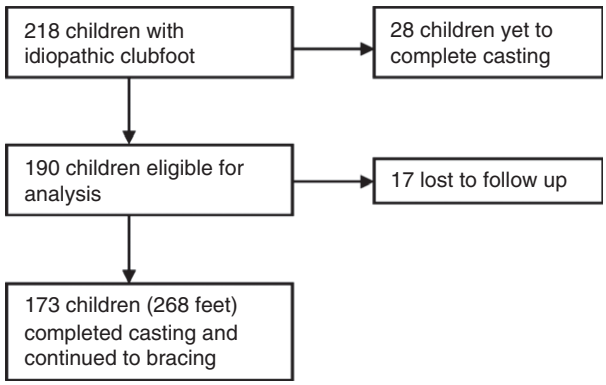

Figure 1 Flow chart of clubfoot treatment pathway.

Bilateral clubfoot was seen in 90 children (52%); 83 (48%) had unilateral clubfoot (37 left and 46 right). The sample included more males than females. The median age at first treatment was 8 months (interquartile range 2.8 months to 20.1 months old); 169 (97.7%) children

Table 1 Characteristics of the children who completed manipulation and casting (N = 173)

Characteristics	N (%)*
Gender	
Male	119 (68.8)
Female	54 (31.2)
Previous treatment†	
None	40 (23.7)
Kite Method	118 (69.8)
Long leg cast	8 (4.7)
Short leg cast and surgery	2 (1.2)
Strapping	1 (0.6)
Presentation age	
≤2 years	143 (82.7)
>2 years	30 (17.3)
Birthplace‡	
Hospital	73 (45.3)
Clinic	73 (45.3)
Home	15 (9.2)
Side of clubfoot	
Bilateral	90 (52.0)
Unilateral	83 (48.0)
Birth order†	
First born	61 (36.1)
Second born	47 (27.8)
Third born	37 (21.9)
≥Fourth born	24 (14.2)
Previous family history§	
Yes	18 (10.5)
No	153 (89.5)
Age of mother at first treatment‡	
≤20	23 (14.3)
20.1–30	89 (55.3)
30.1–40	42 (26.1)
≥40.1	7 (4.3)

*Percentage excluding missing values.
†Missing for 4 children (2.3%).
‡Missing for 12 children (6.9%).
§Missing for 2 children (1.2%).

were under the age of 5 years, with four children between 5 and 6 years of age. At presentation, 67 (38.7%) children were under 6 months, and 40 (23.7%) children were new referrals with no previous treatment; 118 of 129 (91.5%) children with a history of previous treatment were treated with below-knee casts using the Kite method [20]. The mean number of casts reported with the Kite method was 16.03 (95% CI: 13.94–18.12). Less than 10% of children were born at home. A family history of clubfoot was reported by 18 (10.5%) parents.

Pirani score

Of the 173 children (268 feet) who completed casting, 161 children (246 feet) had a full record of pre- and post-treatment Pirani scores (Table 2).

At the start of treatment, 221 of 246 feet (89.8% – boys 90.9% and girls 89.2%) had a Pirani score of 2.0 or more. After completion of the corrective phase, 209 of 246 feet (85.0%) achieved a Pirani score of 1 or less, of which 37 feet had a Pirani score of 0; 99 achieved 0.5; and 73 had a Pirani score of 1. A further 27 (11.0%) feet had a Pirani score of 1.5 and 10 (4.0%) failed to achieve a score of 1.5 or less.

Table 2 Pre- and post-treatment Pirani scores of feet that completed casting

Pirani score	Pre-treatment foot *N** (%)†	Post-treatment foot *N** (%)†
0–1.0	13 (5.3)	209 (85.0)
1.5	12 (4.9)	27 (11.0)
2.0–6.0	221 (89.8)	10 (4.0)
Total	246 (100.0)	246 (100.0)

*Missing for 22 children.
†Percentage excluding missing values.

A Pirani score of 1 or less was achieved more frequently in girls with clubfoot than boys (94.0% *vs.* 81.5%). The multivariable adjusted regression model showed that a girl's foot was 7.49 (95% CI: 1.24–45.45) times more likely to achieve a score of 1 or less after manipulation and casting than that of a boy. A more severe clubfoot was less likely to achieve an end Pirani score of 1 or less (AOR = 0.75, 95% CI: 0.56–0.99). Age at first treatment and previous treatment were not associated with final Pirani score (Table 3).

The mean Pirani score on presentation was 3.80 (SD 1.15), and mean Pirani score on final cast removal was 0.80 (SD 0.56) with $P < 0.001$ on paired t-test for change in Pirani score. Change in Pirani score is demonstrated in Figure 2.

Boys with clubfoot experienced less change in score than girls [−0.25 (95% CI: −0.43 to −0.07) (Table 4). A more severe clubfoot was likely to have a greater change in score with treatment. There is no evidence that age at first treatment or history of previous treatment is associated with change in Pirani score after multivariable adjustment.

The mean initial Pirani score for a girl's foot was 3.40 (95% CI: 3.05–3.74) and for a boy's foot, 3.76 (95% CI: 3.60–3.93). While there was overlap in the confidence intervals, there is a trend for girls having less severe deformity at baseline, which may explain the better (lower) final Pirani score outcome.

Number of casts

The mean number of casts required in the case series to completion of corrective phase was 7.27 (95% CI 6.7–7.9) (Table 5). Children under the age of 2 years required fewer casts (−1.72, 95% CI −3.27 to −0.18), but the more severe the initial deformity, the more casts were required (0.75, 95% CI: 0.44–1.06). There was no

Table 3 Factors associated with a Pirani score of 1 or less (Number of feet = 246)

Predictor Variable	Category	Score ≤1 Feet *N* (%)	Score >1 Feet *N* (%)	Crude OR (95%, CI)	AOR for all variables* (95%, CI)
Gender	Male	146 (81.6)	33 (18.4)	1	1
	Female	63 (94.0)	4 (6.0)	5.13 (1.07, 24.70)	7.49 (1.24, 45.45)
Age at first treatment	>2 years	31 (70.5)	13 (29.5)	1	1
	≤2 years	178 (88.1)	24 (11.9)	4.89 (1.23, 19.49)	1.13 (0.31, 4.10)
Previous treatment	No	52 (91.2)	5 (8.8)	1	1
	Yes	154 (83.7)	30 (16.3)	0.43 (0.11, 1.69)	0.46 (0.11, 1.95)
Pre-treatment severity category	Unit increase			0.82 (0.63, 1.06)	0.75 (0.56, 0.99)

*Adjusted for place of birth, family history, birth order, affected foot, mother's age and remaining three potential predictors using a random effects model.

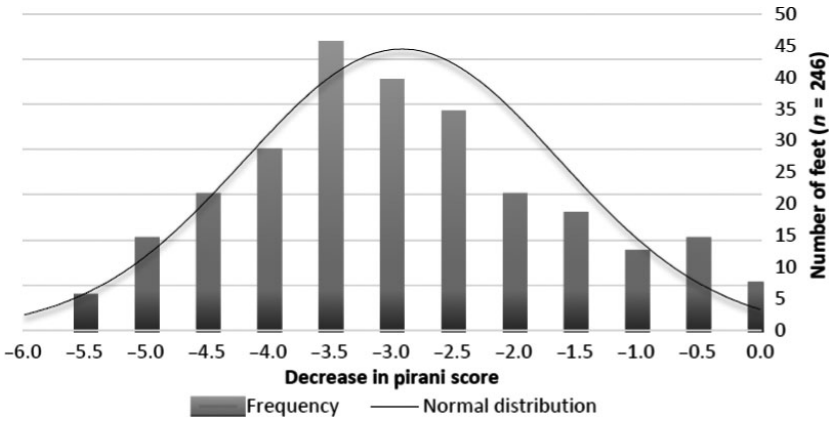

Figure 2 Distribution of change in Pirani score from pre- to post-treatment.

Table 4 Predictors of change in pre- to post-treatment Pirani scores (*N* feet = 246)

Predictor variable	Category	Mean score change (95%, CI)	Crude coefficient (95%, CI)	Adjusted coefficient* (95%, CI)
Gender	Female	2.87 (2.52, 3.23)	Reference	Reference
	Male	2.93 (2.75, 3.11)	0.07 (−0.35, 0.50)	−0.25 (−0.43, −0.07)
Age at first treatment	>2 years	2.59 (2.23, 2.96)	Reference	Reference
	≤2 years	2.99 (2.81, 3.17)	0.50 (−0.01, 1.01)	−0.01 (−0.23, 0.21)
Previous treatment	No	3.57 (3.25, 3.89)	Reference	Reference
	Yes	2.70 (2.52, 2.88)	−0.91 (−1.35, −0.47)	0.06 (−0.13, 0.26)
Pre-treatment severity category	Unit increase		0.47 (0.45, 0.50)	0.47 (0.44, 0.50)

*Adjusted for place of birth, family history, birth order, affected foot, mother's age and remaining three potential predictors using a random effects model.

Table 5 Factors associated with number of casts (*N* = 164)

Predictor Variable	Category	Mean number of casts (95%, CI)	Crude coefficient (95%, CI)	Adjusted coefficient* (95%, CI)
Gender	Female	6.53 (5.47, 7.58)	Reference	Reference
	Male	7.61 (6.80, 8.42)	0.65 (−0.69, 2.00)	0.53 (−0.87, 1.94)
Age at first treatment	>2 years	9.04 (7.25, 10.83)	Reference	Reference
	≤2 years	6.90 (6.22, 7.57)	−1.72 (−3.27, −0.18)	−2.49 (−4.10, −0.88)
Previous Treatment	No	7.55 (6.26, 8.84)	Reference	Reference
	Yes	7.24 (6.46, 8.01)	−0.62 (−2.10, 0.85)	−0.37 (−1.16, 1.90)
Pre-treatment severity category	Unit increase		0.75 (0.48, 1.01)	0.75 (0.44, 1.06)

*Adjusted for place of birth, family history, birth order, affected foot, mother's age and remaining three potential predictors.

association between gender or previous treatment and the number of casts required to achieve correction.

Achilles tenotomy

A percutaneous Achilles tenotomy was performed on 127 (78.9%) of 161 children who completed treatment to bracing phase; 69 tenotomies were on bilateral feet and 58 on unilateral (Table 6).

A child with a history of previous treatment was 8.15 (95% CI: 2.00–33.20) times more likely to have a tenotomy performed than a child with no prior treatment. An increase of pre-treatment Pirani score increased the likelihood that a tenotomy was performed (AOR = 1.72, 95%

Table 6 Factors associated with tenotomy (N = 173)

Predictor variable	Category	Tenotomy *N* (%)	No Tenotomy *N* (%)	Crude OR (95%, CI)	Adjusted OR* (95%, CI)
Gender	Female	36 (72.0)	14 (28.0)	1	
	Male	91 (81.3)	21 (18.7)	1.69 (0.77, 3.67)	1.72 (0.54, 5.48)
Age at first treatment	>2 years	23 (79.3)	6 (20.7)	1	
	≤2 years	104 (78.2)	29 (21.8)	0.94 (0.35, 2.51)	0.99 (0.20, 4.99)
Previous treatment	No	27 (69.2)	12 (30.8)	1	
	Yes	97 (80.2)	24 (19.8)	1.64 (0.72, 3.78)	8.15 (2.00, 33.20)
Pre-treatment severity category	Unit increase			1.51 (1.26, 1.81)	1.72 (1.33, 2.23)

*Adjusted for place of birth, family history, birth order, affected foot, mother's age and remaining three potential predictors.

CI: 1.33–2.23). There was no evidence for an association between gender or age at first treatment and whether a tenotomy was performed.

Discussion

The gender ratio and bilateral to unilateral clubfoot ratio are similar to global figures, with more males than females (2.2:1) and slightly more bilateral than unilateral cases [21]. The most common presenting Pirani (severity) score was 4.5, suggestive of a moderately severe clubfoot presentation, although less severe than other clubfoot cohort reports in the region [17, 22, 23]. Children attended Parirenyatwa clubfoot clinic prior to the introduction of the Ponseti method when treatment was with the Kite method [20]. The reduced initial Pirani scores are likely to be due to the inclusion of 118 children who had previous treatment with the Kite method. The mean age at first treatment was 13.5 months and less than 39% of children presented under 6 months of age.

This large cohort in Zimbabwe found that the Ponseti manipulation and casting method was successful in 85% of feet, when success is defined as a Pirani score of 1 or less at the end of the correction phase. However, further follow-up of this cohort is required to evaluate the longer-term outcome after bracing up to the age of 5 years. It will be important to see whether the good results obtained in the manipulation and casting phase can be maintained through the bracing phase when compliance with treatment is likely to be a greater problem. Also, it will be necessary to evaluate the patient or carer reported functional outcome, as well as the anatomical structural outcomes [24].

The only predictor of low Pirani score at the end of casting was female gender. Severity of deformity was associated with a larger change in Pirani score (pre- to post-treatment), which is in line with current literature that suggests the Ponseti method is also effective in severe cases [25]; however, a higher Pirani score at baseline was less likely to achieve a satisfactory outcome (Pirani score of 1 or less) on completion of manipulation and casting.

Children with high Pirani scores at presentation (severity of clubfoot) were more likely to require additional casts as expected [17, 25, 26]. In addition, more casts were required for the correction of clubfoot for children over 2 years.

The proportion of children having tenotomy in the current series (78.9% of children/79.7% of clubfeet) is higher than reported in most studies from sub-Saharan Africa [27]. The Parirenyatwa clubfoot clinic is a physiotherapist-led clinic and communication with the orthopaedic department occurred regularly regarding the progress of correction and timing of tenotomy. Medical officers and equipment were available to complete the required tenotomies upon request.

Children with high Pirani scores at presentation were more likely to require tenotomy, as shown previously [28]. This is likely due to tighter connective tissues which are evident in severe deformity and could relate to a biomechanical difference in the collagen itself [29]. In addition, children with a history of previous treatment were over eight times more likely to have a tenotomy performed than if there was no history of prior treatment. This may be due to previous inadequate management with the Kite procedure [30], or because children referred to Parirenyatwa with a history of previous treatment may have had more severe deformity, as children treated successfully elsewhere would be less likely to present to the clinic.

Seventeen children were lost to follow up during casting. No attempt was made to contact those lost to follow up. While services are provided free of charge for children under the age of five at Parirenyatwa, further cultural, financial and structural barriers pertinent to surgery [31] may contribute to the ability to adhere to treatment.

Results of the current cohort of children treated at Parirenyatwa hospital may not be representative of other settings in Zimbabwe. This tertiary referral clubfoot clinic had a regular supply of plaster of paris, Steenbeek foot abduction braces and access to Medical Officers during the study time period. It is difficult to generalise the results to clinics that experience shortages of orthopaedic, surgical and material resources.

Well-maintained medical records for a large cohort of children in a low-income country are strengths of this study and the 173 cases provide one of the largest case series reported in sub-Saharan Africa.

The study has a number of limitations. There was no comparator (control or other treatment group) within the case series. Although attribution of the change in Pirani score to the intervention is plausible, it is not proven. The study participants were self-selected and therefore selection bias cannot be ruled out. In addition, the Pirani score has been reported to have good intra- and inter-rater reliability and validity in young children [14] and is recommended for use in LMICs [32]; however, it is not validated for older children. No information was available from the medical records as to whether further treatment including surgery was performed on the 37 feet that failed to achieve a Pirani score of 1.

Plausible evidence from case studies, before-and-after treatment studies and a small number of RCTs suggests the minimally invasive Ponseti method as the best practice. Continued evaluation of treatment results is required, and in the light of the evidence, randomised controlled trials with historical techniques are unethical. A standardised method of reporting service delivery is necessary for the evaluation of outcomes between clinics and regions. In addition, clearly outlined protocols will assist further exploration of the Ponseti method outcomes and predictors of success for clubfoot management.

In conclusion, a consensus is required on a simple user-friendly definition of success for the Ponseti method to compare one series with another and to assist field therapists monitor the results. Success for this case series on the completion of the corrective phase of treatment was defined as a Pirani score of 1 or less. The case series demonstrates that the majority (80%+) of children with clubfoot can achieve a Pirani score of 1 or less, with a relatively small loss to follow up (8.9%) to completion of corrective phase. Age at presentation and previous history of treatment did not greatly affect the final Pirani score. Follow-up is required to assess maintenance of a good result and if a low dropout rate can be maintained through bracing.

Acknowledgements

We thank the Zimbabwe Ministry of Health and Child Care, the Parirenyatwa Clubfoot Clinic Staff, Dr Max Gova and his orthopaedic team, Mrs R Muzarurwi, Ms C Ncube and the physiotherapy team, Ryan Bathurst and Debra Mudariki of The Zimbabwe Sustainable Clubfoot Programme, for their diligence in the treatment of clubfoot and support of this project. The Beit Trust is gratefully acknowledged for the scholarship awarded to TS.

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Epilogue (Main findings and limitations)

173 children (268 feet) completed manipulation and casting within the study period. The mean length of time for the corrective phase was 10.2 weeks. Seventeen (8.9%) of the initial 218 children were lost to follow up.

A Pirani score of 1 or less (defined as success) was achieved in 85% of feet. The only predictor of success (Pirani score ≤ 1) at the end of casting was female sex.

Children with severe clubfoot deformity (high Pirani scores at presentation), and those over two years old were more likely to require additional casts for correction. The proportion of children that had a tenotomy was 79%. Children with severe clubfoot deformity were more likely to receive a tenotomy.

The limitations of this retrospective case series are: (a) the clubfoot clinic records did not include all the details for risk factor analysis and therefore limit the analysis; (b) there is no comparator within the case series and although attribution of success to the intervention is very likely, it is not proven; (c) the cut-off score for success (Pirani score of 1 or less) was decided in consultation with expert Ponseti method trainers and was based on the acceptability of outcomes rather than a 'perfect' score; and (d) there is a risk of selection bias as the children are self-selected and attended a tertiary clinic that may not be representative of the clinics that other children in Zimbabwe attend.

Additional Information: Erratum. The odds of achieving a Pirani score of 1 or less are 25% less in a child with a more severe clubfoot compared to a child with a less severe clubfoot (AOR = 0.75, 95% CI: 0.56–0.99). A child with a history of previous treatment had eight times the odds (AOR= 8.15 (95% CI: 2.00–33.20) of having a tenotomy performed than a child with no prior treatment. The odds that a tenotomy would be performed increased with an increase of pre-treatment Pirani score (AOR = 1.72, 95% CI: 1.33–2.23). Children with a history of previous treatment had over eight times the odds of receiving a tenotomy compared to those without any previous treatment. This may be due to inadequate management with the Kite procedure (previous treatment), which relates to the formation of a complex clubfoot.

Chapter 7. What is a good result after clubfoot treatment? A Delphi-based consensus on success by regional clubfoot trainers from across Africa

Family waiting for first review in clubfoot clinic

Preamble

Considerable variation exists in the evaluation of outcomes of clubfoot treatment. Whilst the Pirani score is frequently used to assess success during the corrective phase of treatment (Chapter 5), it is not validated for use in children of walking age because it does not reflect the extent of the deformity. Therefore, a different measure for use during the bracing (maintenance) phase is required to assess long-term outcome (Chapter 6).

In addition, non-specialist health workers deliver clubfoot services in clinics in Africa and these ‘clubfoot therapists’ assess, treat and manage children born with clubfoot. They require valid and repeatable outcome measures that are easy to use to determine their results of clubfoot treatment, and to know when to refer a child for specialist treatment.

As there is no agreed measure recommended for use by clubfoot therapists in Africa to document long-term outcome of treatment, the opportunistic meeting of 35 expert Ponseti method trainers in Ethiopia for the Africa Clubfoot Training project provided an environment to establish consensus on criteria for successful clubfoot correction during the bracing phase (long-term outcome).

A Delphi process with the regional trainers and experts in Africa was used to reach consensus on the most important criteria for assessing success in children of walking age. Although this sequential process does not ensure validity, the agreement provides a basis for establishing criteria that are likely to have clinical sensibility.

This chapter comprises a published research paper that outlines the Delphi method used, and the criteria identified for successful clubfoot management in Africa.

This research paper was published in the journal PLoS ONE in December 2017 after peer review.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?	PLOS ONE		
When was the work published?	21st December 2017		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion	n/a		
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For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I co-designed the study, designed the data collection tools, extracted the data, completed data analysis, drafted the manuscript, prepared the subsequent revisions with consideration of comments from co-authors

Student Signature: _____ **Date:** 9/5/18

Supervisor Signature: _____ **Date:** 9/5/18

PLOS ONE

RESEARCH ARTICLE

What is a good result after clubfoot treatment? A Delphi-based consensus on success by regional clubfoot trainers from across Africa

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Citation: Smythe T, Wainwright A, Foster A, Lavy C (2017) What is a good result after clubfoot treatment? A Delphi-based consensus on success by regional clubfoot trainers from across Africa. PLoS ONE 12(12): e0190056. https://doi.org/10.1371/journal.pone.0190056

Editor: Hiroyuki Tsuchiya, Kanazawa University, JAPAN

Received: December 23, 2016

Accepted: December 7, 2017

Published: December 21, 2017

Data Availability Statement: All relevant data are within the paper and its Supporting Information files.

Funding: This work was supported by Beit Trust (TS) and CBM (TS).

Competing interests: The authors have declared that no competing interests exist.

Abstract

Background

Congenital talipes equino-varus (CTEV), also known as clubfoot, is one of the most common congenital musculoskeletal malformations. Despite this, considerable variation exists in the measurement of deformity correction and outcome evaluation. This study aims to determine the criteria for successful clubfoot correction using the Ponseti technique in low resource settings through Africa.

Methods

Using the Delphi method, 18 experienced clubfoot practitioners and trainers from ten countries in Africa ranked the importance of 22 criteria to define an 'acceptable or good clubfoot correction' at the end of bracing with the Ponseti technique. A 10cm visual analogue scale was used. They repeated the rating with the results of the mean scores and standard deviation of the first test provided. The consistency among trainers was determined with the intraclass correlation coefficient (ICC). From the original 22 criteria, ten criteria with a mean score >7 and SD <2 were identified and were rated through a second Delphi round by 17 different clubfoot treatment trainers from 11 countries in Africa. The final definition consisted of all statements that achieved strong agreement, a mean score of >9 and SD<1.5.

Results

The consensus definition of a successfully treated clubfoot includes: (1) a plantigrade foot, (2) the ability to wear a normal shoe, (3) no pain, and (4) the parent is satisfied. Participants demonstrated good consistency in rating these final criteria (ICC 0.88; 0.74,0.97).

Conclusions

The consistency of Ponseti technique trainers from Africa in rating criteria for a successful outcome of clubfoot management was good. The consensus definition includes basic physical assessment, footwear use, pain and parent satisfaction.

Introduction

Congenital talipes equino-varus (CTEV), or clubfoot, is one of the most prevalent congenital musculoskeletal malformations that affects mobility [1]. The most common method of treatment worldwide is now the Ponseti technique [2]. This primarily non-operative technique is beneficial in low- and middle-income countries (LMICs) where there are limited resources and different cadres of health workers can be trained to treat clubfoot [3]. Despite this, considerable variation exists in the assessment of deformity correction and outcome. Goals of clubfoot treatment include improvement in foot function, the creation of a pain free, shoeable foot, parent satisfaction [4,5] and avoidance of corrective surgery [6]. As the treatment can be delivered by trained health workers [7], the non-specialist health workers require valid, repeatable and easy to measure outcome measures to determine their results of clubfoot treatment in low-resource settings. There is no consensus regarding the definition of success of clubfoot management and diverse criteria [8–10] have been proposed. The concept of success after the bracing phase requires further investigation in environments that share a context of public health systems with overcrowded clinics and limited access to equipment, such as goniometers.

The Delphi method is a structured consensus technique that may be used to reach agreement about outcomes [11]. It is a sequential process through which the anonymous opinions of participants are sought [12] and this allows equal weight to be given to all participants [13,14]. After the completion of each round of questionnaires, the collated group responses are fed back to participants. Establishing consensus does not ensure validity, however agreement provides a basis for establishing criteria that are likely to have clinical sensibility [15].

This study aims to determine criteria for successful clubfoot correction at the conclusion of the bracing phase in a low resource setting, by establishing consensus amongst expert Ponseti trainers in the Africa region.

Materials and methods

The study was performed and reported following the recommended guidelines [11] for selection of healthcare quality indicators. Eighteen trainers from ten national clubfoot programmes in Africa attended a workshop in January 2016. The trainers were regional experts in clubfoot management, and they deliver training in the Ponseti method in their respective countries. The participants were chosen based on willingness to participate and knowledge of the topic [16] and included orthopaedic surgeons, physiotherapists and orthopaedic technicians. The mean length of time that the trainers had used the Ponseti method for was 7.7 years (95%CI 6.0–9.3) and the average number of trainings delivered was 4.7 (95%CI 2.2–7.1)

The Delphi method employed was a two-round self-administered questionnaire. To identify outcomes that are important, the questionnaire was developed through a regional workshop of Ponseti experts [17]. Potential criteria for assessment of good clubfoot correction were discussed. A systematic literature review of outcomes reported for clubfoot treatment through

Africa found different definitions of success at various points in clubfoot treatment [18–23], all of which were discussed by the experts. Twenty-two potentially relevant criteria for good clubfoot correction were identified in the workshop by the Ponseti technique trainers. The questionnaire was pilot tested for suitability.

The 18 regional trainers were invited to participate, and all completed a questionnaire that included the criteria of a successful outcome, previously generated in the workshop. The questionnaire asked the respondents to rate each of the items for their relative importance using a 10cm visual analogue scale (VAS) with the anchors 'completely unimportant' and 'extremely important' at each extreme. Respondents were asked to consider all the listed criteria as independent; the paper questionnaires were completed by hand.

The VAS means and standard deviations (SD) were calculated based on the responses of all the trainers. After two days, a second questionnaire was delivered to the same 18 trainers with the results of the previous questionnaire (VAS mean and SD). No criteria were excluded and there was no discussion among the participants.

The consistency among the 18 trainers was determined with the intra-class correlation coefficient (ICC). The conventional interpretation of the ICC is as follows: $\leq$0.40, poor consistency or large variation in opinion; 0.41 to 0.74, acceptable consistency; and $\geq$0.75 good consistency [24]. All data were managed and analysed using Stata 14.2, StataCorp 4905, Lakeway Drive College Station, Texas 77845, USA.

In July 2016, 6 months after the first workshop, a second, two-round Delphi method was used to reach a consensus definition of good or acceptable clubfoot correction after bracing. Ten "successful outcome" criteria generated in January 2016 were included which met two predefined criteria: (1) a mean VAS higher than 7 on the ten-point scale, and SD $<$2; and (2) applicable to outcome at the conclusion of the bracing phase. As there is variability in the measurement of distribution of scores in studies that use the Delphi method [25], the thresholds for the VAS mean and SD were decided *a-priori*. The aim was to generate a list of items that participants considered important to assess for acceptable clubfoot correction after bracing from the original list of questions that related to outcome in general.

Seventeen different regional clubfoot trainers (from eleven countries) who attended a workshop in July 2016 were invited to participate. The mean length of time that the trainers had used the Ponseti method for was 7.9 years (95%CI 6.9–9.9) and the average number of trainings delivered was 8.3 (95%CI 5.1–11.5). The Delphi process was similar to that undertaken in January using a 10cm visual analogue scale with the anchors 'completely unimportant' and 'extremely important.' The paper questionnaires were completed by hand. The trainers repeated the rating with the results of the mean scores and standard deviation of the first test visible two days later. The questionnaires used in the second rounds of the Delphi process are included in Supplementary Information files (S1 and S2 Files).

Criteria with a mean VAS rated $>$ 9 and with a SD $<$ 1.5 were considered to have high agreement. Where two criteria described the same indicator with the difference being only the language used, (e.g. foot is flat on the floor and plantigrade foot) the criterion with the highest VAS mean and lowest SD was selected.

The study methodology and course of action for the management of responses is outlined in Fig 1.

Ethics statement

Ethical approval for this study was granted by the London School of Hygiene & Tropical Medicine Ethics Committee (approval number 10412). Written consent was obtained at the beginning of the workshop and data were analysed anonymously.

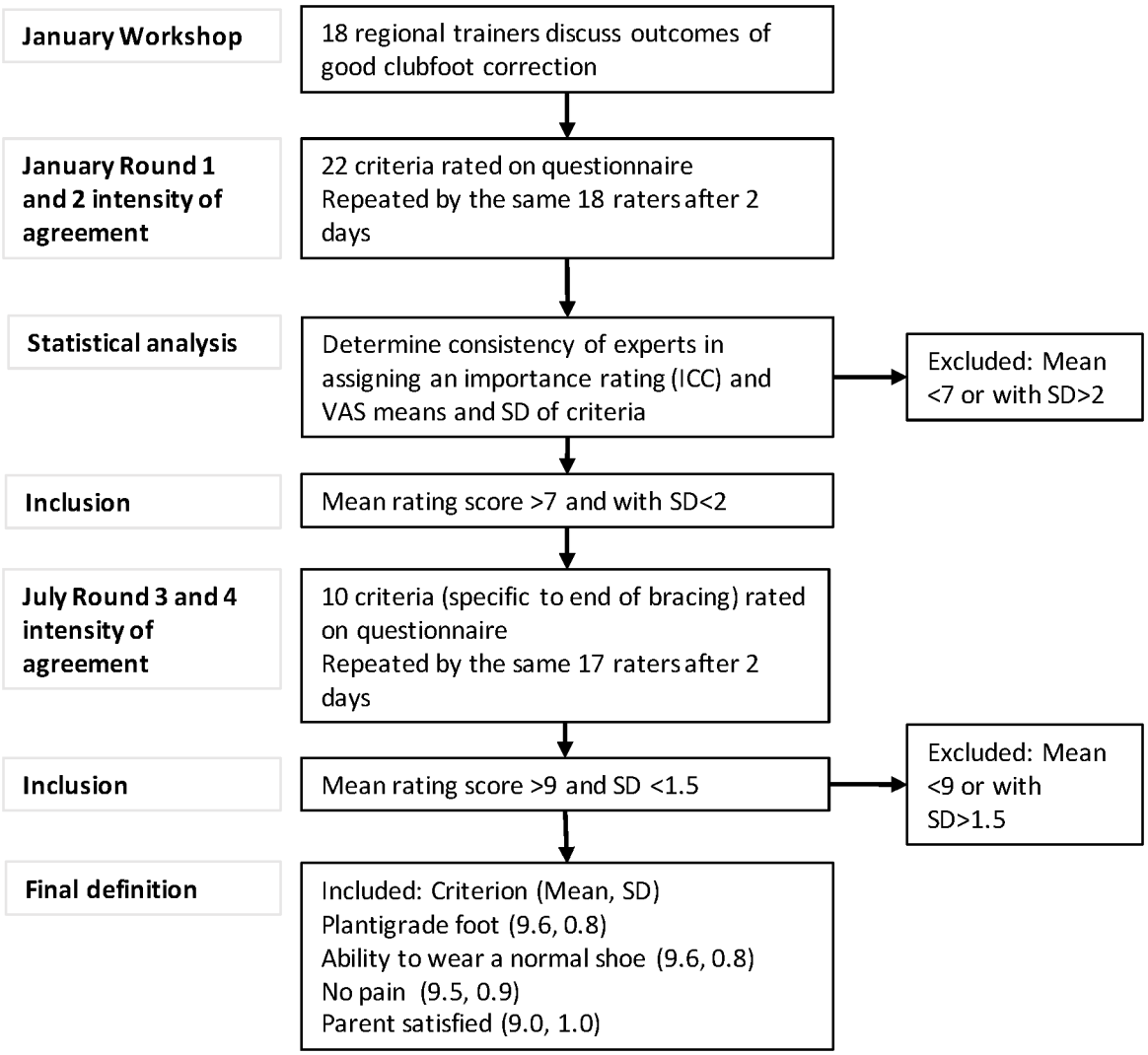

Fig 1. Flow chart of criteria selection. Definitions of abbreviations: SD = standard deviation, ICC = intra-class correlation, VAS = visual analogue scale).

https://doi.org/10.1371/journal.pone.0190056.g001

Results

The response rate of trainers to the questionnaires was 100% on the first round and 94% on the second round in both January and July 2016. The consistency of Ponseti trainers in Africa in rating criteria for successful outcome of clubfoot management was good. The first Delphi ICC had external consistency of 0.83 (0.71–0.92) and the second Delphi ICC had external consistency of 0.88 (0.74–0.97). From the initial 22 criteria, 10 met the inclusion criteria for the second two rounds of Delphi. Details for the ranking of each criterion by trainers in Africa are shown in Tables 1 and 2.

The distribution of the data for the final ten criteria is displayed in Fig 2.

Outcomes that had >9 VAS mean with <1.5 SD were criterion (4) plantigrade foot, (6) ability to wear a normal shoe, (10) no pain and (1) carer is satisfied.

Table 1. Final rating of criteria in the first Delphi round (ordered by visual analogue scale mean and standard deviation).

First Delphi round, January 2016		
Criterion	**Mean**	**SD**
The foot fits comfortably into a Foot Abduction Brace	8.90	0.71
The foot is plantigrade	8.78	1.26
The foot has 15 degrees of dorsiflexion or more	8.57	1.95
The heel is in a neutral position (no longer in varus)	8.23	1.53
The child can wear a normal shoe	8.16	1.76
The child reports no pain	8.13	1.83
The child demonstrates heel strike when walking	8.11	1.64
The forefoot adductus is corrected	8.09	1.15
The carer is satisfied	7.96	1.43
The foot does not supinate in swing phase when walking	7.92	1.33
The foot does not have less than 60 degrees of abduction	7.74	1.86
The Pirani score is 0.5 or less	7.67	3.04
The child keeps up with peers when walking and running	7.58	2.24
The foot has 10 degrees of dorsiflexion or more	7.52	1.81
The Pirani score is 0.5 or less	6.90	2.73
The foot is corrected within 6 casts	6.76	2.54
The Pirani score is 1 or less	6.66	2.72
The wear on the shoes are symmetrical (in unilateral clubfoot)	6.10	2.67
The child had a tenotomy	5.80	3.02
The foot has more than 30 degrees of abduction	5.44	1.74
The Pirani score is 1.5 or less	5.36	2.34
The Pirani score is 2 or less	4.94	2.74

https://doi.org/10.1371/journal.pone.0190056.t001

Discussion

Non-specialist health workers require valid, repeatable and easy to measure outcome measures to determine their results of clubfoot treatment in clinics through Africa. This study determined the opinions of experts from eleven countries in Africa about the criteria for success following clubfoot treatment after the manipulation and bracing phases with the Ponseti technique. The aim of the Delphi method was to define criteria that could be used by any

Table 2. Final rating of criteria in the second Delphi round (ordered by visual analogue scale mean and standard deviation).

Second Delphi round, July 2016		
Criterion	**Mean**	**SD**
The foot is plantigrade	9.56	0.79
The child can wear a normal shoe	9.56	0.79
The child reports no pain	9.47	0.88
The carer is satisfied	9.01	1.13
The foot has 15 degrees of dorsiflexion or more	8.99	1.01
The heel is in a neutral position (no longer in varus)	8.79	1.10
The forefoot adductus is corrected	8.65	1.22
The child demonstrates heel strike when walking	8.51	1.01
The foot does not supinate in swing phase when walking	8.44	1.18
The foot does not have less than 60 degrees of abduction	7.44	2.04

https://doi.org/10.1371/journal.pone.0190056.t002

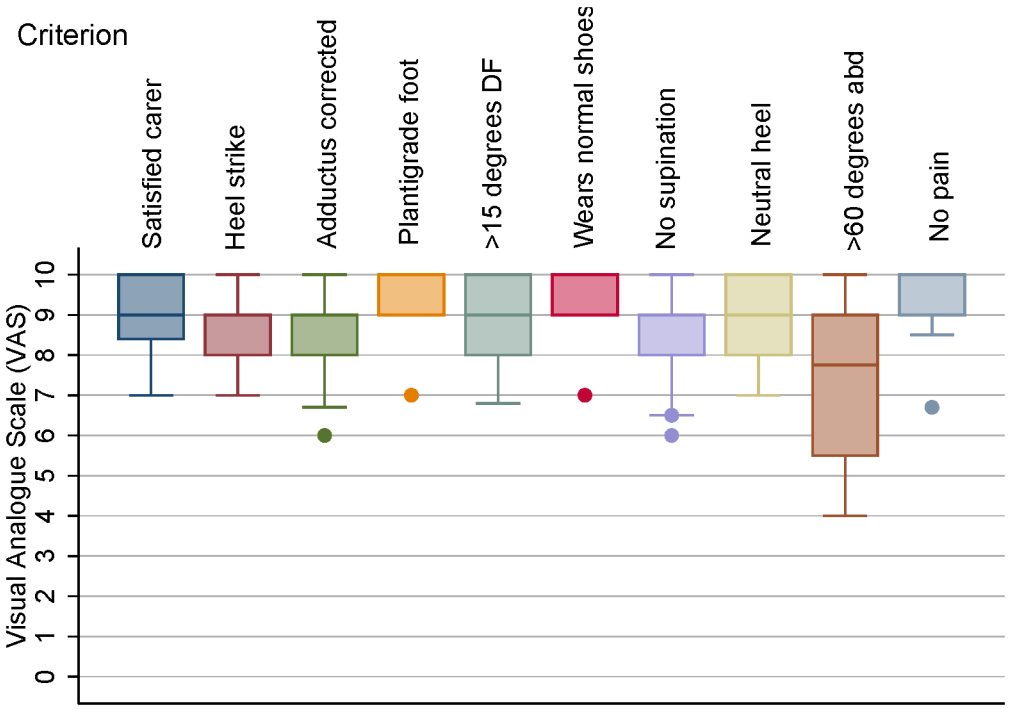

Fig 2. Ratings for successful clubfoot correction after bracing (10 criteria in the order asked on the questionnaire). Box and whisker plot of the final ten criteria. The middle 50% of the VAS ratings are shown as the box. The horizontal line in the box represents the median value. The upper and lower quartiles are indicated by the whiskers and outliers are indicated by a circle. (Definitions of abbreviations: DF = dorsiflexion, abd = abduction).

https://doi.org/10.1371/journal.pone.0190056.g002

clubfoot practitioner working in busy clinics with limited resources through Africa. Regional trainers were therefore deemed the most appropriate experts to interview in this context.

This study found that the highest rated outcomes were a plantigrade foot, ability to wear a normal shoe, parent satisfaction and absence of pain. These criteria are included in other published assessment tools. Laaveg and Ponseti described a detailed functional rating system [8] that requires the use of a goniometer to evaluate outcomes of treatment and incorporates patient satisfaction, pain, gait, heel position and range of motion. The Roye tool [4] consists of ten questions designed to measure treatment outcomes through overall satisfaction, appearance, pain and physical limitations in a high income setting [26]. The Bangla tool [9] was developed to evaluate results of clubfoot management in Bangladesh, where clinics required a tool that was quick, relevant and reliable for use in children of walking age. The Clubfoot Assessment Protocol (CAP) includes a detailed assessment of movement quality and requires accurate passive mobility testing with a goniometer and awareness of muscle testing [10], but it does not include parent reported outcomes. These four tools have been developed in local contexts by individual institutions.

This study used a Delphi process with many experts, in the context of Africa, to develop and then rank criteria that are viewed to be important in the assessment of a successful outcome for clubfoot management in low resource settings. The finding that these four criteria are included in the criteria of the other published assessment tools contributes to evidence of their validity.

There are limitations of this study. Previous research has shown that panel composition influences ratings [27]. The panel in this study was selected for their expertise, but may not be

representative of all Ponseti treatment practitioners. There may be some criteria that were not considered which may also be important.

The expert trainers showed good consistency in rating satisfactory outcomes for clubfoot management (ICC 0.88; 0.74,0.97) and a strength of this study includes the high response rate of the survey (94%).

The consensus definition includes four criteria—a simple physical assessment, footwear use, patient pain and parent reported outcome measures. It is likely that these four criteria will provide a good overall assessment of successful treatment of a child with clubfoot in Africa, and may be useful in other geographic contexts after further investigation. The use of these four criteria should allow the development a simple assessment tool that can be used by non-specialist health workers. The other aspects of utility and reliability of this tool will then need to be studied in future research.

Conclusion

Appropriate measures are required to determine the successful outcome of clubfoot treatment and to compare different treatment techniques in low resource settings. Using a Delphi process with experts from across Africa, we were able to find consensus for the four most important criteria of a successful clubfoot treatment using the Ponseti method.

Supporting information

S1 File. Questionnaire for Delphi 1 round 2.
(DOCX)

S2 File. Questionnaire for Delphi 2 round 2.
(DOCX)

Acknowledgments

We thank the experts who participated in this study.

Author Contributions

Conceptualization: TS AW AF CL.

Data curation: TS.

Formal analysis: TS.

Funding acquisition: TS AF CL.

Investigation: TS AW CL.

Methodology: TS AW AF CL.

Project administration: TS CL.

Resources: TS AW AF CL.

Supervision: AW AF CL.

Validation: TS CL AW AF.

Visualization: TS.

Writing – original draft: TS.

Writing – review & editing: TS AW AF CL.

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S1: Appendix 1. Delphi 1 round 2

How important are the following in determining a '**good'** clubfoot correction?

1. The carer/family is happy with the results (average 8.0)

0 1 2 3 4 5 6 7 8 9 10

2. The child demonstrates heel strike when walking (average 8.1)

0 1 2 3 4 5 6 7 8 9 10

3. The forefoot adductus is corrected (average 8.2)

0 1 2 3 4 5 6 7 8 9 10

4. The wear on the shoes are symmetrical (unilateral clubfoot) (average 6.1)

0 1 2 3 4 5 6 7 8 9 10

1. The child had a tenotomy (average 4.1)

0 1 2 3 4 5 6 7 8 9 10

6. The foot is plantigrade (average 8.7)

0 1 2 3 4 5 6 7 8 9 10

7. The Pirani score is 2 or less (average 4.1)

0 1 2 3 4 5 6 7 8 9 10

8. The foot has 15 degrees of dorsiflexion or more (average 8)

0 1 2 3 4 5 6 7 8 9 10

9. The Pirani score is 1 or less (average 6.3)

0 1 2 3 4 5 6 7 8 9 10

10. The child can wear a normal shoe (average 8.1)

0 1 2 3 4 5 6 7 8 9 10

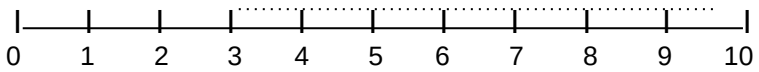
11. The Pirani score is 1.5 or less (average 5.5)

0 1 2 3 4 5 6 7 8 9 10

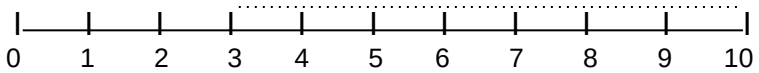
12. The foot does not supinate in swing phase when walking (average 7.4)

0 1 2 3 4 5 6 7 8 9 10

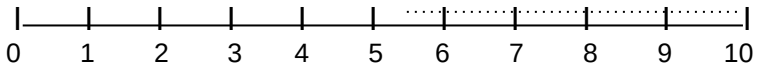
13. The Pirani score is 0.5 or less (average 6.5)



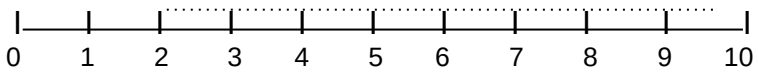
14. The Pirani score is 0 (average 6.7)



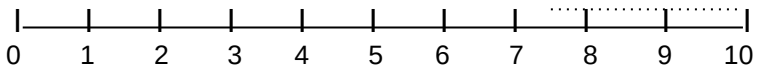
15. The heel is in a neutral position (no longer in varus) (average 7.7)



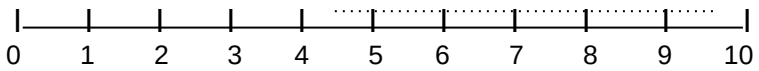
16. The foot is corrected within 6 casts (average 5.6)



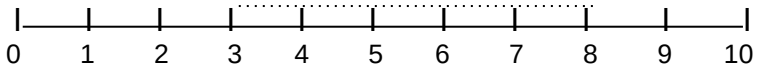
17. The foot fits comfortably into a foot abduction brace (average 8.8)



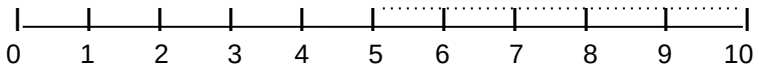
18. The foot does not have less than 60degrees of abduction (average 7.1)



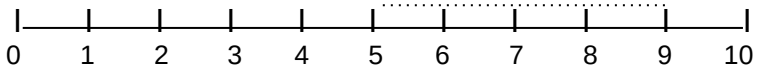
19. The foot has more than 30 degrees of abduction (average 5.7)



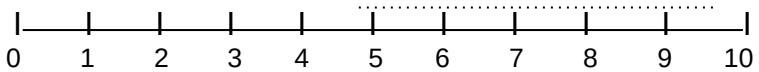
20. The foot has 15 degrees of dorsiflexion or more (average 7.8)



21. The child keeps up with peers when walking and running (average 7.3)



22. The child reports no pain (average 7.7)



S2: Appendix 2. Delphi 2 round 2

How important are the following to determine an acceptable or '**good'** clubfoot correction at completion of bracing?

1. The carer/family is happy with the results (average 8.9)

0 1 2 3 4 5 6 7 8 9 10

2. The child demonstrates heel strike when walking (average 8.1)

0 1 2 3 4 5 6 7 8 9 10

3. The forefoot adductus is corrected (average 8.3)

0 1 2 3 4 5 6 7 8 9 10

4. The foot is plantigrade (average 9.3)

0 1 2 3 4 5 6 7 8 9 10

5. The foot has 15 degrees of dorsiflexion or more (average 8.8)

0 1 2 3 4 5 6 7 8 9 10

6. The child can wear a normal shoe (average 9.6)

0 1 2 3 4 5 6 7 8 9 10

7. The foot does not supinate in swing phase when walking (average 8.4)

0 1 2 3 4 5 6 7 8 9 10

8. The heel is in a neutral position (no longer in varus) (average 8.7)

0 1 2 3 4 5 6 7 8 9 10

9. The foot has 60 degrees of abduction or more (average 7.0)

0 1 2 3 4 5 6 7 8 9 10

10. The child reports no pain (average 9.3)

0 1 2 3 4 5 6 7 8 9 10

Epilogue (Main findings and limitations)

This study found that the highest rated outcomes of clubfoot treatment were a plantigrade foot, ability to wear a normal shoe, parent satisfaction and absence of pain. These individual criteria are included in other published assessment tools.

It is likely that these four criteria taken together will provide a good overall assessment of successful treatment of a child of walking age with clubfoot in Africa.

The main limitations of this study include (a) the ratings of the criteria may have been influenced by the expert panel composition; (b) the panel was selected for their expertise and may not be representative of all Ponseti treatment practitioners; and (c) there may be some criteria that were not considered which may also be important.

Finally, whilst this Delphi study determined opinions of experts in Africa and was used to reach agreement, this process does not ensure validity.

Additional Information: Erratum. Table 1 line 15 of the published paper should read, “The Pirani Score is 0” (with a mean of 6.9 and a SD of 2.73).

Chapter 8. Evaluation of a simple tool to assess the results of Ponseti treatment for use by clubfoot therapists

Functional assessment in the clubfoot clinic

Preamble

There is need for a user-friendly, comprehensive tool to assess children of walking age who have undergone Ponseti treatment for clubfoot in Africa.

The consensus definition of a ‘good’ long-term outcome of clubfoot treatment (Chapter 7) included four criteria - one simple physical assessment and three questions for the caregiver on the child’s use of footwear, on-going pain experienced by the child, and carer satisfaction.

We developed a tool with four possible answers for each of the four identified criteria. Each answer is scored from 0 to 3, with 3 being the maximum and a total of 12 being the best result.

The submitted research paper in this chapter uses these four criteria to define and evaluate a simple assessment tool, called the ‘Assessing Clubfoot Treatment (ACT) score’, to assess the long-term success of Ponseti treatment in children of walking age.

A prospective cohort study of 218 children with clubfoot, treated in one clinic in Zimbabwe, was used. Two experts documented the long-term outcome in 68 children attending follow-up using the ACT tool against a gold standard of full clinical assessment.

The study evaluates how effective the use of the ACT score was in identifying children who needed referral for further specialised management. The study also identified factors that influence long-term successful outcome.

This paper was submitted to the journal Medicine in January 2018 and is undergoing peer review.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?			
When was the work published?			
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion	n/a		
Have you retained the copyright for the work?*		Was the work subject to academic peer review?	

**If yes, please attach evidence of retention. If no, or if the work is being included in its published format, please attach evidence of permission from the copyright holder (publisher or other author) to include this work.*

SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	Medicine
Please list the paper's authors in the intended authorship order:	Smythe T, Mudariki D, Gova M, Foster A, Lavy C
Stage of publication	Submitted

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I co-designed the study and designed the data collection protocol. I developed the tool algorithm and performed the statistical analysis. I wrote the first draft of the manuscript and prepared the revisions with comments from co-authors

Student Signature: _____ **Date:** 9/5/18

Supervisor Signature: _____ **Date:** 9/5/18

Evaluation of a simple tool to assess the results of Ponseti treatment for use by clubfoot therapists: a diagnostic accuracy study

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Funding awarded to TS: The Beit Trust, MiracleFeet, ZANE

List of abbreviations:

ACT: assessing clubfoot treatment

FAB: foot abduction brace

ICC: interclass correlation coefficient

QoL: quality of life

ROC: Receiver Operating Characteristic

STARD: Standards for Reporting of Diagnostic Accuracy Studies

Abstract

Background:
We aimed to develop and evaluate a tool for clubfoot therapists in low resource settings to assess the results of Ponseti treatment of congenital talipes equinovarus (CTEV) in children of walking age.

Method:
A literature review and a Delphi process were used to develop the Assessing Clubfoot Treatment (ACT) tool and score. We followed up 68 children with CTEV, treated between 2011 and 2013, in 2017. A full clinical assessment was conducted to decide if treatment was successful or if further treatment was required. The ACT score was then calculated for each child. Inter-observer variation for the ACT tool was assessed. Sensitivity, specificity, positive and negative predictive values were calculated for the ACT score compared to full clinical assessment (gold standard). Predictors of a successful outcome were explored.

Results:
The ACT tool consists of 4 questions; each scored from 0 to 3, giving a total from 0 to 12 where 12 is the ideal result. The 4 questions consist of one physical assessment and three parent reported outcome measures. It takes 5 minutes to administer and had excellent inter-observer agreement.

An ACT score of 8 or less demonstrated 79% sensitivity and 100% specificity in identifying children that required further intervention, with a positive predictive value of 100% and negative predictive value of 90%. Children who completed two or more years of bracing were four times more likely to achieve an ACT score of 9 or more compared to those who did not (OR: 4.08, 95%CI: 1.31 – 12.65, p = 0.02).

Conclusions:
The ACT tool is simple to administer, had excellent observer agreement, and good sensitivity and specificity in identifying children who need further intervention. The score can be used to identify those children who definitely need referral and further treatment (score 8 or less) and those with a definite successful outcome (score 11 or more), however further discrimination is needed to decide how to manage children with a borderline ACT score of 9 or 10.

Level of Evidence: Level II, Diagnostic Study

Introduction

Clubfoot, or congenital talipes equinovarus (CTEV), is a common deformity where the affected foot is fixed downward and inward. The birth prevalence of CTEV is estimated in the range of 0.5 to 2.0 cases/1,000 live births in LMIC (1). Most cases of clubfoot occur as an isolated birth defect and are known as 'idiopathic' because the cause is not known. The remaining 20% of cases are associated with other structural conditions such as arthrogryposis, syndromes and disorders of the nervous system, for example spina bifida (2). Male sex is consistently associated with an increased risk of clubfoot (3).

There is a global trend toward use of the minimally invasive Ponseti method (4) for the correction of CTEV, which consists of simultaneous correction of the components of the clubfoot deformity with manipulation and casting. A percutaneous tenotomy of the Achilles tendon is usually required to correct the residual equinus. A foot abduction brace (FAB) is then needed to maintain the corrected position until 4 years of age (5); the CTEV deformity has a strong tendency to recur after corrective treatment because the factors that initiate the deformity remain active as the child grows (6). Recurrence of elements of the deformity is therefore less common after the child is four years old as growth of the foot decreases in speed.

In low resource settings non-specialist health workers are trained as clubfoot therapists (7). They assess, diagnose, treat and follow up patients with CTEV (8). Several scoring systems have been described for CTEV; these include the Ponseti-Laaveg classification (9) and the Dimeglio classification (10), which are complicated to use. The Pirani score (11) is frequently used to assess success during the corrective phase of treatment, however it is not validated for use in children of walking age because it does not reflect the extent of the deformity. The Roye tool (12) measures patient based outcomes in a high-income setting and the Bangla tool (13) was developed to evaluate results in Bangladesh and requires mathematical calculations. There is no agreed assessment of severity of recurrence and elements of the deformity that recur are typically noted under clinical examination and observation of function.

There remains a need for a valid, repeatable and easy to administer tool that will allow clubfoot therapists to differentiate a good outcome of treatment from a less acceptable outcome that needs further intervention. In addition, a standardised method to assess parent reported outcomes after CTEV treatment is required. To address this gap, we aim to develop a user friendly, comprehensive tool to assess children of walking age

who have undergone Ponseti treatment for CTEV, including parent reported outcome measures about the key quality of life factors that affect the child and parent.

Materials and Methods

This study was conducted and reported according to established STARD (Standards for Reporting of Diagnostic Accuracy Studies) guidelines (14). (Additional File 1).

Defining the ACT tool

The Assessing Clubfoot Treatment (ACT) tool was developed through a Delphi process with 35 Ponseti method trainers in Africa. The Delphi study method, criteria, description of consensus and analysis are published elsewhere in detail (15). The most important criteria for successful CTEV correction were determined and found to be (i) a plantigrade foot, (ii) the ability to wear a normal shoe, (iii) no pain, and (iv) the parent is satisfied. A literature review was used to develop four possible answers for each of the four identified criteria, and a score given for each answer. The assessment tool was then pilot tested including contextual relevance.

The inter-observer variation for the ACT tool was assessed with two experienced physiotherapists who train and mentor clubfoot therapists in Zimbabwe, and are experienced in co-ordinating national clubfoot programmes. The interclass correlation coefficient (ICC) was calculated for agreement. The conventional interpretation was used: ≤0.40, poor consistency; 0.41 to 0.74, acceptable consistency; and ≥0.75 good consistency (16).

Study population

A cohort study was established of 218 children with idiopathic CTEV managed at Parirenyatwa Hospital, Harare. The results of manipulation and casting are published elsewhere (17). The cohort included all children with a diagnosis of idiopathic CTEV corrected by the Ponseti method at the study hospital between March 2011 and April 2013 (25 months). The only exclusion criterion was conditions other than idiopathic CTEV, for example clubfoot associated with a syndrome or comorbidity, e.g. spina bifida.

Cohort follow up

In January 2017, when patients were 3.5 – 5.0 years from initial casting, we attempted to follow up all children in the cohort. Phone numbers were extracted from clinic records and carers and their children were invited to participate in the study. Contact was attempted at least three times.

Study design

First, the ACT tool was administered independently by the two physiotherapists who were experienced in the management of CTEV in countries in Africa (examiners). Then within an hour a full clinical assessment was performed independently by the two examiners, which involved observation, physical assessment and functional performance review; this included assessment of passive and active range of motion (plantiflexion, dorsiflexion, eversion, inversion of the foot, and knee extension), muscle strength tests of the calf and evertors of the foot), heel raises, squatting ability and gait analysis (walking and running), and discussion with the carer of the child. This examination protocol led to a decision that referral of the child for further treatment (re-casting or surgical review) was required, or that no further intervention was needed. The two examiners then discussed their independent decisions and came to one joint management decision. The examiners were therefore not blind to the decision outcome. The usual process is a clinical assessment by either one or two physiotherapists. The joint management decision was chosen as the gold standard with which to compare the ACT tool. After the decision was recorded, the ACT score was calculated.

Data collection

The question about the plantigrade position of the foot was answered first by independent physical examination of the child in supine by the physiotherapists, with the knee extended and though the measurement of passive range of dorsiflexion of the hindfoot. The remaining three questions of the ACT score were answered by the carers about the child's pain, ability to wear shoes and satisfaction. The child followed verbal instructions to complete the functional performance review. In addition, data were collected using a self-administered healthcare satisfaction questionnaire (18) and a quality of life questionnaire (19). The questionnaires were available in English and Shona and were cognitively tested. Each measure was recorded by hand on a separate paper. The study protocol was pilot tested for suitability before use.

Data Management and Analysis

All data were entered into a Microsoft Excel 2000 (Microsoft Inc., Redmond, Washington) software package. Data were analysed using Stata 14.1 (Stata-Corp 4905, Lakeway Drive College Station, Texas 77845,USA.).

A descriptive analysis compared characteristics of the children who attended follow up with that of the whole cohort.

A comparative analysis of outcomes was explored between three groups of children (a) those who had not completed casting, (b) those who had completed casting and had < 2 years of bracing, and (c) those who completed casting and had 2+ years of bracing.

Sensitivity, specificity, positive and negative predictive values were calculated for the ACT score compared to the gold standard (good outcome or needs referral for further orthopaedic management). A Receiver Operating Characteristic (ROC) curve for the ACT score was created to demonstrate the trade-off between sensitivity and specificity (20).

The potential predictors of the ACT tool were explored. Proportions were calculated for the parent reported outcome measures of healthcare satisfaction and quality of life.

Ethics

The Medical Research Council of Zimbabwe (MRCZ) and the London School of Hygiene & Tropical Medicine (LSHTM) granted ethical approval. The caregiver provided informed written consent. Transport costs were reimbursed.

Results

The ACT tool consists of one question about the plantigrade position of the foot answered by physical examination and three questions answered by the carers about the child's pain, ability to wear shoes and carer satisfaction (15) (Table 1). There are 4 possible answers to each question with a corresponding score from 0 (severe problems) to 3 (no problems). The total score is calculated within a range of 0 to 12.

Table 1: ACT questions and score

Score	1.The foot is plantigrade	2.Does your child complain of pain in their affected foot?	3.Can your child wear shoes of your/their choice?	4.How satisfied are you with your child's foot?
0	Does not reach plantigrade, with additional adduction, cavus or varus	Yes and it often limits their activity	Never	Very dissatisfied
1	Does not reach plantigrade, no additional deformity	Yes and it sometimes limits their activity	Sometimes	Somewhat dissatisfied
2	Plantigrade achieved	Yes but it does not limit their activity	Usually	Somewhat satisfied
3	More than plantigrade i.e. some dorsiflexion	No	Always	Very satisfied

Inter-observer agreement

The interclass correlation coefficient (ICC) was 0.99 for questions 1 and 3 and 1.0 for questions 2 and 4.

Cohort follow-up

Sixty-eight children of the cohort of 218 children (31.2%) attended for assessment in early 2017. The total analysis time under observation was 171.3 years.

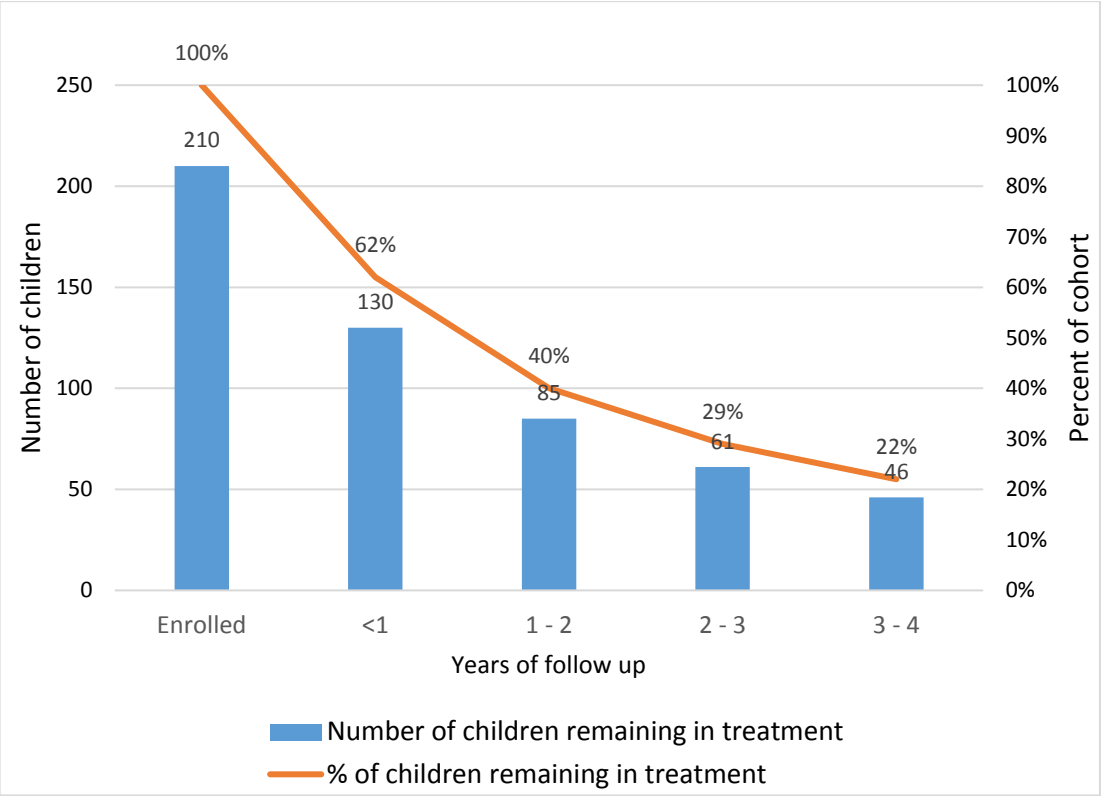
The follow up group is representative of the whole cohort in terms of sex, laterality, mean Pirani score at baseline, average number of casts applied, and tenotomy proportion (Table 2). They attended treatment for longer than those not seen, indicating increased compliance with treatment.

Table 2: Demographic details of cohort

	Total N (%)	Followed up N (%)	Not followed up N (%)	P-value
Total	218 (100%)	68 (31%)	150 (69%)	
Male Female	148 (68%) 70 (32%)	50 (73%) 18 (27%)	98 (65%) 52 (35%)	0.23
Bilateral Unilateral	119 (54%) 99 (45%)	35 (51%) 33 (49%)	84 (56%) 66 (44%)	0.53
Tenotomy**: Yes No	158 (73%) 57 (27%)	52 (76%) 16 (24%)	106 (72%) 41 (28%)	0.43
	Total(95%CI)	Followed up (95%CI)	Not followed up (95%CI)	P-value
Average age at first cast	14 months (12 – 17)	17months (11 – 23)	13 months (11 – 15)	0.16
Mean initial Pirani score L foot R foot	 3.8 (3.6 – 4.0) 3.7 (3.5 – 3.9)	 3.7 (3.4 – 4.0) 4.0 (3.6 – 4.3)	 3.8 (3.6 – 4.0) 3.6 (3.3 – 3.8)	 0.21 0.56
Average number of casts	7.2 (6.6 – 7.9)	6.9 (5.9 – 8.0)	7.4 (6.6 – 8.2)	0.44
Average months attending since first appointment	23 months (20 – 25)	30 months (26 – 35)	19 months (17 – 22)	0.0001*

**missing data from 3 children

By the end of the second year of treatment, 60% of the cohort had stopped attending clinic appointments (Figure 1).



*data missing for 8 children

Figure 1: Children attending clubfoot clinic (bracing and correction phase)*

Of the 68 children seen at follow up, 63 (93%) completed casting and were fitted with a brace, and 38 (56%) used a brace for more than 2 years.

Clinical assessment

All children were assessed with the reference standard and after the full clinical assessment by the two examiners 44/68 (65%) children were judged not to require any further intervention and 24/68 (35%) were judged to require further treatment (re-casting or surgical review). (Additional file 2) Where there was initial disagreement, consensus on the decision was reached through discussion. No adverse events occurred as a result of any of the outcome measures undertaken.

For the 38 children who finished casting and completed 2+ years of bracing 82% were judged to have a successful outcome (Table 3). Completion of casting and at least 2 years bracing was strongly associated with a successful outcome.

Table 3: Outcome at follow-up as judged independently by two expert physiotherapists

	Totals		Finished casting and 2+years bracing		Finished casting and <2yrs bracing		Did not finish casting	
Recruited at baseline**	**218****	*100%*	**83**	*39.5%*	**107**	*51%*	**20**	*9.5%*
Seen at follow-up	**68**	*31%*	**38**	*56%*	**24**	*35%*	**6**	*9%*
No intervention required	**44**	*65%*	**31**	*82%*	**12**	*50%*	**1**	*17%*
Referral for further orthopaedic intervention	**24**	*35%*	**7**	*18%*	**12**	*50%*	**5**	*83%*
P value for difference in proportion of those requiring intervention and those not*				0.001*		0.06		0.02

**missing data from 8 children (4%) *Fischers exact test

ACT tool

55/68 (81%) children achieved plantigrade, and in those who had completed 2+ years of bracing, this increased to 97% (37/38). Scores for parent reported outcome measures increased for children who had completed two years of bracing (Table 4).

Table 4: ACT score distribution

		Score 3	Score 2	Score 1	Score 0
		N (%)	N (%)	N (%)	N (%)
Total Cohort followed up (n=68)	Foot is plantigrade	33 (49%)	22 (32%)	7 (10%)	6 (9%)
	Complain of pain	44 (65%)	18 (27%)	5 (7%)	1 (1%)
	Wears shoe of choice	47 (69%)	13 (19%)	6 (9%)	2 (3%)
	Satisfied with foot	42 (62%)	14 (21%)	9 (13%)	3 (4%)
Completed casting (n=63)	Foot is plantigrade	31 (49%)	22 (35%)	7 (11%)	3 (5%)
	Complain of pain	43 (68%)	15 (24%)	5 (8%)	0 (0%)
	Wears shoe of choice	45 (71%)	12 (19%)	6 (10%)	0 (0%)
	Satisfied with foot	41 (65%)	12 (19%)	9 (14%)	1 (2%)
Completed ≥2 years bracing (n=38)	Foot is plantigrade	22 (58%)	15 (39%)	1 (3%)	0 (0%)
	Complain of pain	27 (71%)	9 (24%)	2 (5%)	0 (0%)
	Wears shoe of choice	27 (71%)	8 (21%)	3 (8%)	0 (0%)
	Satisfied with foot	28 (74%)	6 (16%)	4 (10%)	0 (0%)

72% (49/68) of the children followed up achieved a score of 9 or more. This proportion increased to 84% (32/38) in those who had completed 2+ years of bracing (Table 5).

Table 5: ACT score according to compliance with Ponseti treatment

ACT score	Total N	Score ≤6	Score 7-8	Score 9-10	Score 11-12
ACT score for total cohort	68	10 (15%)	9 (13%)	13 (19%)	36 (53%)
ACT score for those completing casting	63	8 (13%)	7 (11%)	13 (21%)	35 (55%)
ACT score for those completing casting and bracing for ≥2 years	38	2 (5%)	4 (11%)	9 (24%)	23 (60%)

Sensitivity and specificity

24 children required further intervention, of which 19 scored 8 or less on the ACT score (sensitivity: 79%) and the remaining children scored 9 or 10.

Of the 44 children who did not require further intervention on full clinical assessment, all scored 9 or more (specificity: 100%).

A score of 9 or more was found in 49 children, of which 44 were identified as not requiring further intervention (negative predictive value: 90%). Among the children who scored 8 or less, all 19 had been clinically assessed as requiring further intervention (positive predictive value: 100%).

If a score of 9 was used to predict the need for intervention instead of 8, the sensitivity increased from 79% to 83%, the specificity decreased from 100% to 87% and negative and positive predictive values from 90% to 80% and 100% to 91% respectively.

An ACT score of 8 or less correlates with the need for intervention and scores of 11 and 12 correlate with no need for further intervention. An ACT score of 9 or 10 warrants further review.

The ACT score ROC area is 0.97 (95%CI 0.94 – 1.00). (Figure 2) The closer the curve follows the left-hand border and then the top border of the ROC space, the more accurate the test.

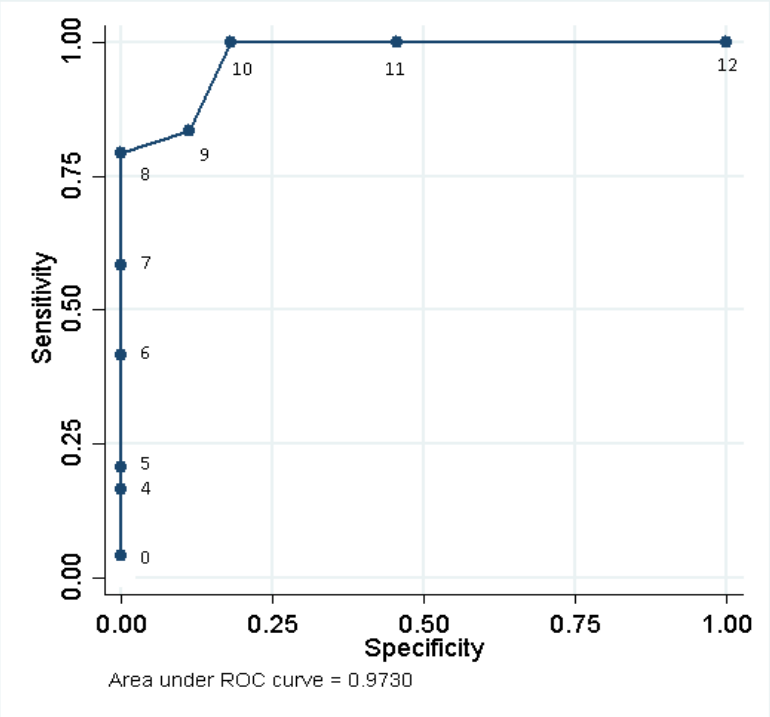


Figure 2: ROC display for ACT score

Factors affecting ACT score

Children who completed casting and were fitted with a brace had twelve times the odds (95%CI: 1.33 – 123.49 p = 0.03) of achieving a good outcome (score 9 -12) than those who did not. Those with 2+ years of brace wear had four times the odds (95% CI: 1.31 – 12.65 p = 0.02) of achieving a score of 9 -12 than those who used a brace for < 2 years.

Sex, side of CTEV, age at first clinic attendance, initial severity, number of casts, and tenotomy performed were not associated with an ACT score that predicted need for further intervention (score 0 - 8) (Table 6).

Table 6: Predictors of ACT score

Factor		Success / Borderline (ACT score 9-12) N (%)	Failure (ACT score 0-8) N (%)	Crude OR (95%CI)	P-value
Total		54 (79%)	14 (21%)		
Gender	Male	35 (71%)	14 (29%)	1.00	0.85
	Female	14 (74%)	5 (26%)	1.12 (0.34 – 3.70)	
Clubfoot	Bilateral	24 (69%)	11 (31%)	1.00	0.51
	Unilateral	25 (76%)	8 (24%)	1.43 (0.49 – 4.17)	
Age first attended clinic	<2 years	40 (74%)	14 (26%)	1.00	0.42
	≥ 2 years	9 (64%)	5 (36%)	0.57 (0.15 – 2.19)	
Initial severity (Pirani score)	< 3	34 (69%)	15 (31%)	1.00	0.43
	≥ 3	15 (79%	4 (21%)	1.65 (0.47 – 5.82)	
Number of casts	≥ 6	28 (68%)	13 (31%)	1.00	0.85
	<6	21 (78%)	6 (22%)	1.63 (0.53 – 4.98)	
Tenotomy	No	14 (74%)	5 (26%)	1.00	0.85
	Yes	35 (71%)	14 (29%)	0.89 (0.27 – 2.95)	
Completed casting and fitted with brace	No	1 (20%)	4 (80%)	1.00	0.03*
	Yes	48 (76%)	15 (24%)	12.8 (1.3 – 123.5)	
Completed casting with Pirani score ≤ 1	No	11 (55%)	9 (45%)	1.00	0.05*
	Yes	38 (79%)	10 (21%)	3.11 (1.01 – 9.56)	
Brace use	< 2 years	17 (57%)	13 (43%)	1.00	0.02*
	≥ 2 years	32 (84%)	6 (16%)	4.08 (1.31 – 12.65)	

Quality of life

There is a marked improvement in QoL in all areas for those who complete casting compared to those who did not (Additional File 3). An ACT score 9 -12 was associated with an increased QoL (p=0.002).

Healthcare Satisfaction

There was a tendency for parents whose children completed ≥2 years of bracing to be more satisfied (93%, 95%CI: 88 – 99) with the information given to them in the clubfoot clinic than those who did not (85%, 95%CI: 76 – 93) (Additional File 4) but this difference was not statistically significant.

Discussion

A simple tool to assist non-specialist health workers to identify a good outcome after treatment with the Ponseti method from an outcome that needs further management is required. The four-question ACT tool was shown to have a high sensitivity and specificity in identifying children who need additional intervention.

Gold standard

There is no accepted gold standard to assess the results of CTEV treatment. In the context of Africa, trained clubfoot therapists provide treatment. When children are seen during bracing, a decision on need for referral to an orthopaedic clinic is required. To assess whether the ACT tool could assist therapists in making that decision, the gold standard used was the agreement of two experienced physiotherapists after they had independently performed a full clinical assessment.

Comparison to previous studies

This study found that further intervention was indicated in 35% (24/68) of children and the success rate in those that completed casting and 2+ years of bracing was 82%. This indicates what can be achieved if there is good compliance with treatment and adequate follow-up. This is similar to high income settings where the probability of further intervention is reported as approximately 29% (21).

ACT tool and score

The ACT tool (one physical observation and 3 questions to the child's carer) takes approximately five minutes to perform. There was excellent agreement in the results of the test between two different observers.

A score of 8 or less indicated that the child needed referral, whereas a score of 11 or 12 indicated the child had a good outcome. One child with a score of 9 and three children with a score of 10 were judged to need referral for more casting and one child with a score of 10 to need referral for surgical review. These cases included curvature of the lateral border of the foot and review for a Tibialis Anterior transfer. There were no cases that recorded low parent satisfaction or parent reported pain when the foot achieved plantigrade or more.

Strengths and limitations

This was a cohort study with follow-up after first treatment of at least 3.5 years. Repeat phone calls facilitated attendance at the study clinic. The ACT tool was developed through an extensive Delphi process and literature review. The tool is simple and quick to administer and can be used by non-specialist health workers. It includes both physical observation and carer reported outcomes. The study protocol was pilot tested before use.

There were also study limitations. The results in those followed up are likely to be better than those for the cohort as they attended clinic appointments for longer and length of follow up is a predictor of good outcome. The tool is limited to one clinical examination, which restricts identification of pathology that is reliant on complex investigations. It is possible that results from the ACT tool may have influenced the decision to refer. Administering the tool first, but calculating the total score after the full clinical assessment, in addition to requiring agreement on the referral decision, should have reduced this potential for observer bias.

Implications

As non-specialist health workers regularly manage the treatment of CTEV in low resource settings there is a need to provide appropriate tools to allow measurement and evaluation of their treatments. Further work is required to evaluate the ACT tool in other situations and with other cadres of clubfoot therapists. Also further exploration to differentiate children who score 9 or 10 with a good outcome from those who need referral is warranted; in particular the tool is not sensitive in identifying children who have a curvature in the front of the foot but who score high due to parent satisfaction, good footwear use and absence of pain.

Recommendations

The use of the ACT score is to accurately inform and predict future management. It answers such questions as: (1) does the child need more treatment? (2) has the child been successfully treated? and (3) will the child's quality of life be improved? It is suitable for use in children who are of walking age.

We recommend that the ACT tool is used on a yearly basis after completion of casting and commencement of bracing, or if the non-specialist health worker has concerns regarding the outcome of CTEV treatment in a child of walking age. A score ≤8 predicts the need for further intervention. If a child scores 9 to 10, we recommend the clubfoot therapist identifies the primary reason and seeks a second opinion.

Conclusion

This paper contributes to the data on the measurement of CTEV treatment in low resource settings. The ACT tool includes a physical observation of the foot and parent reported outcome measures. A score ≤ 8 identifies children who need further intervention, and a score of 11 or 12 identifies children with a successful outcome. Further work is needed to distinguish the few children who have an ACT score of 9 or 10 and who require further treatment from those who have a successful outcome. There is an association between good outcome, high ACT score and higher quality of life.

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Additional File 1. STARD Checklist

Section and Topic	No	Recommendation	Page No
TITLE/ ABSTRACT/ KEYWORDS	1	Identify the article as a study of diagnostic accuracy (recommend MeSH heading 'sensitivity and specificity').	1 & 6
INTRO	2	State the research questions or study aims, such as estimating diagnostic accuracy or comparing accuracy between tests or across participant groups.	1 & 3
METHODS			
Participants	3	The study population: The inclusion and exclusion criteria, setting and locations where data were collected.	4
	4	Participant recruitment: Was recruitment based on presenting symptoms, results from previous tests, or the fact that the participants had received the index tests or the reference standard?	4 & 5
	5	Participant sampling: Was the study population a consecutive series of participants defined by the selection criteria in item 3 and 4? If not, specify how participants were further selected.	4 & 5
	6	Data collection: Was data collection planned before the index test and reference standard were performed (prospective study) or after (retrospective study)?	4 & 5
Test methods	7	The reference standard and its rationale.	5 & 7
	8	Technical specifications of material and methods involved including how and when measurements were taken, and/or cite references for index tests and reference standard.	4 & 5
	9	Definition of and rationale for the units, cut-offs and/or categories of the results of the index tests and the reference standard.	5
	10	The number, training and expertise of the persons executing and reading the index tests and the reference standard.	5
	11	Whether or not the readers of the index tests and reference standard were blind (masked) to the results of the other test and describe any other clinical information available to the readers.	5
Statistical methods	12	Methods for calculating or comparing measures of diagnostic accuracy, and the statistical methods used to quantify uncertainty (e.g. 95% CI)	6
	13	Methods for calculating test reproducibility, if done.	n/a
RESULTS			
Participants	14	When study was performed, including recruitment beginning, end dates	4
	15	Clinical and demographic characteristics of the study population (at least information on age, gender, spectrum of presenting symptoms).	7
	16	The number of participants satisfying the criteria for inclusion who did or did not undergo the index tests and/or the reference standard; describe why participants failed to undergo either test (a flow diagram is strongly recommended).	7
Test Results	17	Time-interval between the index tests and the reference standard, and any treatment administered in between.	5
	18	Distribution of severity of disease (define criteria) in those with the target condition; other diagnoses in participants without the target condition.	7
	19	A cross tabulation of the results of the index tests (including indeterminate and missing results) by the results of the reference standard; for continuous results, the distribution of the test results by the results of the reference standard.	8 & 9
	20	Any adverse events from performing the index tests or the reference standard.	7
Estimates	21	Estimates of diagnostic accuracy and measures of statistical uncertainty (e.g. 95% confidence intervals).	8 & 9
	22	How indeterminate results, missing data and outliers of the index tests were handled.	n/a
	23	Estimates of variability of diagnostic accuracy between subgroups of participants, readers or centers, if done.	8 & 9
	24	Estimates of test reproducibility, if done.	n/a
DISCUSSION	25	Discuss the clinical applicability of the study findings.	10 & 11

Additional File 2. Results of ACT score and treatment required

ACT score	No intervention required (N)	Re-casting required (N)	Surgical review required (N)
0			1
4			3
5			1
6		1	4
7		1	3
8		1	4
9	5	1	
10	3	3	1
11	12		
12	24		

Additional File 3. Results of quality of life questionnaire (Higher % = higher perceived quality of life)

Quality of life Dimension**	Did not complete casting N = 6	Completed casting, but <2 years bracing N = 24	Completed casting, and 2+ years bracing N = 38	Total cohort followed up** N= 62
	Mean (95% CI)	Mean (95% CI)	Mean (95% CI)	Mean (95% CI)
Physical functioning	68 (27 – 100)	88 (81 – 96)	96 (93 – 99)	93 (89 – 96)
Emotional functioning	69 (54 – 82)	84 (76 – 91)	89 (85 – 94)	87 (83 – 91)
Social functioning	75 (41 – 100)	85 (78 – 93)	95 (93 – 98)	91 (87 – 95)
School functioning	65 (15 – 100)	81 (72 – 91)	94 (89 - 99)	88 (83 – 93)
Psychosocial health	72 (47 – 96)	83 (77 – 91)	93 (90 – 96)	89 (86 – 93)
Total score	69 (31 – 100)	86 (80 – 92)	94 (92 – 97)	91 (87 – 94)

**data missing from 6 children

Additional File 4. Results of Healthcare Satisfaction Questionnaire (Higher % = higher satisfaction)

Healthcare satisfaction Dimension**	Cohort Mean % (95%CI) n=64	Complete casting Mean % (95%CI) n=60	Did not complete casting mean % (95%CI) n=4	P-value	Pirani ≤1 % (95%CI) (n=46)	Pirani ≥1 % (95%CI) (n=18)	P-value	Children completing ≥2 years of bracing (n=37)	Children who do not complete 2 years (n=27)	P-value
Information	83 (77 – 88)	84 (78 – 90)	70 (0 – 100)	0.49	82 (75 – 88)	85 (72 – 97)	0.56	88 (83 – 96)	75 (64 – 86)	**0.02***
Inclusion of family	82 (76 – 87)	82 (76- 87)	77 (21 – 100)	0.25	81 (75 – 87)	84 (70 – 97)	0.50	83 (76 – 90)	80 (71 – 90)	0.63
Communication	83 (77- 88)	83 (78 – 88)	74 (0 – 100)	0.64	82 (76 – 87)	85 (69 – 99)	0.53	83 (76-90)	82 (72 – 92)	0.87
Technical skills	88 (83 – 92)	88 (83 – 92)	88 (48 – 100)	0.42	87 (82 – 92)	90 (79 – 99)	0.45	87 (80 – 93)	89 (82 – 95)	0.67
Emotional needs	74 (66 – 82)	74 (66 – 82)	74 (0 – 100)	0.98	75 (66 – 83)	71 (50 – 92)	0.81	72 (61 – 83)	76 (64 – 88)	0.61
Overall satisfaction	90 (85 – 95)	91 (86 – 95)	74 (4 – 100)	0.98	91 (86 – 95)	87 (73 – 99)	0.59	93 (88 – 99)	85 (76 – 93)	**0.07***
Total score	83 (78 – 88)	83 (79 – 88)	76 (10 – 100)	0.18	83 (78 – 88)	82 (69 – 96)	0.82	84 (78 – 90)	81 (73 – 90)	0.54

**data from 4 children missing

Epilogue (Main findings and limitations)

The Assessing Clubfoot Treatment (ACT) tool consists of one question about the plantigrade position of the foot answered by physical examination and three questions answered by the caregiver about: the child's pain, the child's ability to wear shoes and carer satisfaction. It takes approximately five minutes to perform.

After full clinical assessment by the two physiotherapists (gold standard) 44/68 (65%) children were judged not to require any further intervention and 24/68 (35%) were judged to require further treatment (re-casting or surgical review).

A score of 8 or less with the ACT tool indicated that the child needed referral, whereas a score of 11 or 12 indicated the child had a good outcome. 72% (49/68) of the children followed up achieved a score of 9 or more. This proportion increased to 84% (32/38) in those who had completed two or more years of bracing.

The ACT tool was shown to have a high sensitivity and specificity in identifying children who need additional intervention when compared to full clinical assessment by two experienced physiotherapists. An ACT score of 8 or less demonstrated 79% sensitivity and 100% specificity in identifying children that required further intervention, with a positive predictive value of 100% and negative predictive value of 90%.

Predictors of success, as defined by an ACT of 9 or more, were (a) the child completed casting with a Pirani score ≤ 1, (b) the child completed casting and was fitted with a FAB, and (c) the child used a FAB for two years or more.

A major limitation of this study is that only 31% (68/218) of the original cohort of children attended for long-term follow-up. The implication of this selection bias is that the results in those followed up are likely to be better than those for the whole cohort as they attended clinic appointments for longer and length of follow up is a predictor of good outcome. The tool was applied only to children with idiopathic clubfoot and may not be applicable to children with

secondary clubfoot. The ACT tool and the full clinical assessment were administered by the same two people and it is possible that observer bias through administering the ACT tool first may have influenced the decision to refer taken in the full clinical assessment.

The ACT tool is limited to one clinical examination, which restricts identification of pathology that is reliant on complex investigations. In addition, the tool is not sensitive in identifying children who have a curvature in the front of the foot but who score high due to parent satisfaction, good footwear use and absence of pain.

Chapter 9. A comparison of outcome measures used to report clubfoot treatment with the Ponseti method: results from a cohort in Harare, Zimbabwe

From wearing braces to school shoes

Preamble

Clubfoot therapists often have no specialised physiotherapy or surgical support present in the clinics or nearby. It is important that clubfoot therapists monitor the results of treatment and know when to refer children for specialist review.

There are a variety of scores or clinical examination findings in use to assess outcome of clubfoot treatment. However, there is no consensus on which outcome measure clubfoot therapists should be trained to use. Also there are no agreed indications for referral, be it for review by a specialist or for further intervention, such as re-casting.

We therefore compared the results of the Ponseti method of clubfoot management at three to five years from initial correction using different outcome measures. We explored the ability of different assessment tools to discriminate between the need for referral for further intervention and a successful outcome.

This chapter includes a diagnostic accuracy study to compare the long-term results of Ponseti treatment in a cohort of children in one referral hospital in Zimbabwe.

Five tools that can be used by clubfoot therapists are evaluated. The tools assess success of treatment in children of walking age.

The submitted research paper investigates how the different outcome measures of clubfoot treatment can be used to identify the children needing referral compared to a comprehensive clinical assessment.

This paper was submitted to the journal BMC Musculoskeletal in March 2018 and is undergoing peer review.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?			
When was the work published?			
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion	n/a		
Have you retained the copyright for the work?*		Was the work subject to academic peer review?	

**If yes, please attach evidence of retention. If no, or if the work is being included in its published format, please attach evidence of permission from the copyright holder (publisher or other author) to include this work.*

SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	BMC Musculoskeletal
Please list the paper's authors in the intended authorship order:	Smythe T, Gova M, Muzarurwi R, Foster A, Lavy C
Stage of publication	Submitted

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I co-designed the study and designed the data collection protocol. I developed the tool algorithm and performed the statistical analysis. I wrote the first draft of the manuscript and prepared the revisions with comments from co-authors

Student Signature: _____ **Date:** 9/5/18

Supervisor Signature: _____ **Date:** 9/5/18

A comparison of outcome measures used to report clubfoot treatment with the Ponseti method: results from a cohort in Harare, Zimbabwe

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Target journal: BMC Musculoskeletal

Key words: clubfoot, CTEV, measurement, quality, Ponseti, evaluate, indicator, low resource

Funders: The Beit Trust, MiracleFeet, ZANE

Abbreviations:

ACT tool: Assessing Clubfoot Treatment tool

Background: There are various established scoring systems to assess the outcome of clubfoot treatment after correction with the Ponseti method. We used five measures to compare the results in a cohort of children followed up for between 3.5 to 5 years

Methods: In January 2017 two experienced physiotherapists assessed children who had started treatment between 2011 and 2013 in one clinic in Harare, Zimbabwe. The length of time in treatment was documented. The Roye score, Bangla clubfoot assessment tool, the Assessing Clubfoot Treatment (ACT) tool, proportion of relapsed and of plantigrade feet were used to assess the outcome of treatment in the cohort. Inter-observer variation was calculated for the two physiotherapists. A comparative analysis of the entire cohort, the children who had completed casting and the children who completed more than two years of bracing was undertaken. Diagnostic accuracy was calculated for the five measures and compared to full clinical assessment (gold standard) and whether referral for further intervention was required for re-casting or surgical review.

Results: 31% (68/218) of the cohort attended for examination and were assessed. Of the children who were assessed, 24 (35%) had attended clinic reviews for 4-5 years, and 30 (44%) for less than 2 years. There was good inter-observer agreement between the two expert physiotherapists on all assessment tools. Overall success of treatment varied between 56% and 93% using the different outcome measures. The relapse assessment had the highest unnecessary referrals (19.1%), and the Roye score the highest proportion of missed referrals (22.7%). The ACT and Bangla score missed the fewest number of referrals (7.4%). The Bangla score demonstrated 79.2% (95%CI: 57.8 – 92.9%) sensitivity and 79.5% (95%CI: 64.7 – 90.2%) specificity and the ACT score had 79.2% (95%CI: 57.8 – 92.9%) sensitivity and 100% (95%CI: 92 – 100%) specificity in predicting the need for referral.

Conclusion: At three to five years of follow up, the Ponseti method has a good success rate that improves if the child has completed casting and at least two years of bracing. The ACT score demonstrates good diagnostic accuracy for the need for referral for further intervention (specialist opinion or further casting).

Key words: clubfoot, CTEV, measurement, quality, Ponseti, evaluate, indicator, low resource

Background

Clubfoot, or congenital talipes equinovarus, is a condition that is present at birth in which the foot is in a rigid turned-in position. Corrective treatment of a high quality remains a key requirement for reducing disability and improving function related to the deformity. Over the past decades there has been an increase in the use of the minimally invasive Ponseti method to correct clubfoot (1). The Ponseti method requires correction of the deformity by sequential manipulation and serial casting (often with an achilles tenotomy) followed by long term use of a foot abduction brace at night to maintain the foot position (2). Despite the global trend toward increased use of the Ponseti method, there remains variation in how success of clubfoot treatment is measured (3, 4).

In low resource settings through Africa the Ponseti method is administered by locally trained therapists (5). These therapists often work alone and have no specialised physiotherapy or surgical support present in the clinics or nearby. It is important that they have a user friendly assessment system with agreed criteria for when treatment is not working and referral to a specialist for further management is indicated.

No globally accepted outcome scoring system exists to inform locally trained therapists of the need for referral for further intervention. The most frequently used approach to measuring whether the Ponseti method has been successful (or not) is clinical assessment. In sub-Saharan Africa 68% to 98% of cases are reported to have a successful outcome with the Ponseti method (4). This study aims to compare the results of the Ponseti method of clubfoot management at three to five years from initial correction using five different outcome measures. We explore the diagnostic accuracy of the outcome measures, which is the ability of the assessments to discriminate between the need for referral for further intervention and a successful outcome (6). For methodology review, outcome score results in this study are compared with a reference standard of 'true' treatment success status (defined by full clinical assessment). The results are categorised as true positive, false positive (referred but not needed), true negative, and false negative (should have been referred but was missed) (7). Sensitivity of the scoring system relates to the proportion of the children who need referral for further intervention and who are correctly classified by the outcome measure as requiring referral, and specificity is the proportion of children who do not need referral and who are correctly classified as not requiring referral by the outcome measure. Positive predictive value and negative predictive value are useful to understand the probability that a child with a given positive or negative outcome score

result has the need for referral for further intervention and are therefore correctly classified.

Methods

Study design and population

This study was conducted and reported according to established STARD (Standards for Reporting of Diagnostic Accuracy Studies) guidelines (8) (Additional File 1). A cohort study of 218 children with idiopathic clubfoot was conducted in 2016. The children were managed with manipulation and casting at Parirenyatwa Hospital, Harare and the results are published elsewhere (9). All children with a diagnosis of unilateral or bilateral idiopathic clubfoot who started treatment with the Ponseti method at the study hospital between 22nd March 2011 and 23rd April 2013 (25 months) were included in the cohort. The only exclusion criterion was foot conditions other than idiopathic clubfoot, for example clubfoot associated with neural-tube defects such as spina-bifida.

Sampling technique

The phone numbers of all carers of the cohort children were extracted from the clinic records in January 2017 and contact with them was attempted at least three times. Caregivers and their children were invited to attend the study. The children were between 3.5 and 5 years from initial casting.

Ethics, consent and permissions

Ethical approval for this study was granted by the Medical Research Council of Zimbabwe (MRCZ) and the London School of Hygiene & Tropical Medicine (LSHTM)(ref:11132 /RR/4725). All children and their caregivers were read an information sheet about the study and given an opportunity to ask questions. If they agreed to participate, written consent was taken from the caregiver who remained present throughout the assessment as per national requirements. Transport costs were reimbursed and referral services available in Harare were mapped pre-emptively to ensure appropriate onward referral for any children that required further intervention.

Data collection

Two physiotherapists who are experienced in co-ordinating national clubfoot programmes reviewed the assessment tools over three days for contextual relevance. The questionnaires were available in English and Shona and were cognitively tested. We used five outcome methods, three that give a score, and two that give a binary (success/failure) outcome. The Roye score (10) is a self-reported measurement that is used in high income settings, the Bangla (11) and the Assessing Clubfoot Treatment

(ACT) score (12) and (TBA, under review with the journal Medicine) combine physical assessment and parent reported outcome measures and have been developed for low resource settings. The Bangla score includes a functional assessment. The two binary outcomes were assessment of a plantigrade foot (5) and the relapse pattern (13). The assessment of a plantigrade positon of the foot was evaluated by the physiotherapists through physical examination of the child in supine; the knee was maintained in extension and the passive range of dorsiflexion of the hindfoot was measured. The study protocol was pilot tested for suitability in July 2016. Children were examined independently in January 2017 by the two physiotherapists and a decision was made if referral for further intervention (re-casting or surgical review) was required. Clinical examination composed observation, physical assessment and functional review; it included assessment of passive and active range of motion (plantiflexion, dorsiflexion, eversion, inversion of the foot, and knee extension), muscle strength tests of the calf and evertors of the foot), heel raises, squatting ability and gait analysis (walking and running).

Data Management and Analysis strategy

The data were entered into a Microsoft Excel 2000 (Microsoft Inc., Redmond, Washington) software package. Data were analysed using Stata 14.1 (Stata-Corp 4905, Lakeway Drive College Station, Texas 77845,USA). Statistical significance for the case series was set at the 95% confidence level. The inter-observer variation for the measurement of the physical assessment tools was assessed i.e. Intra-class correlation coefficient (ICC) ≥0.75 (10). Outcomes of children who had completed casting and ≥ two years of bracing were compared to all of the children who were followed up, and to those who had only completed casting. A two-tailed paired t-test was used to assess the mean difference between the outcome measures of Roye, Bangla and ACT scores. Fisher's exact test of independence was used to assess the difference in proportion of children with an outcome of relapse and plantigrade foot. The five measures were compared against the standard of whether referral for further intervention was required (for re-casting or surgical review) as defined by a consensus agreement of two expert physiotherapists with experience of managing clubfoot in Africa. Sensitivity, specificity, positive and negative predictive values were calculated for the five measures and compared to full clinical assessment (gold standard). The threshold for diagnostic accuracy was based on previous studies and was defined prior to the study. It was set at 70% for the three scores with continuous scales (14) and positive/negative for the binary outcomes (7).

Results

31% (68/218) of the cohort attended for review and were assessed. 50 (73%) children were boys and 18 (27%) were girls. There were 35 (51%) bilateral and 33 (49%) unilateral clubfeet. Tenotomies had been performed in 52 (76%) cases and the average number of casts to correction was 6.9 (5.9 – 8.0 casts). The total analysis time under observation was 171.3 years. The average length of time attending appointments from initial review was 30 months (26 – 35months). Of the children followed up, 24 (35%) attended clinic reviews for 4-5 years (Figure 1).

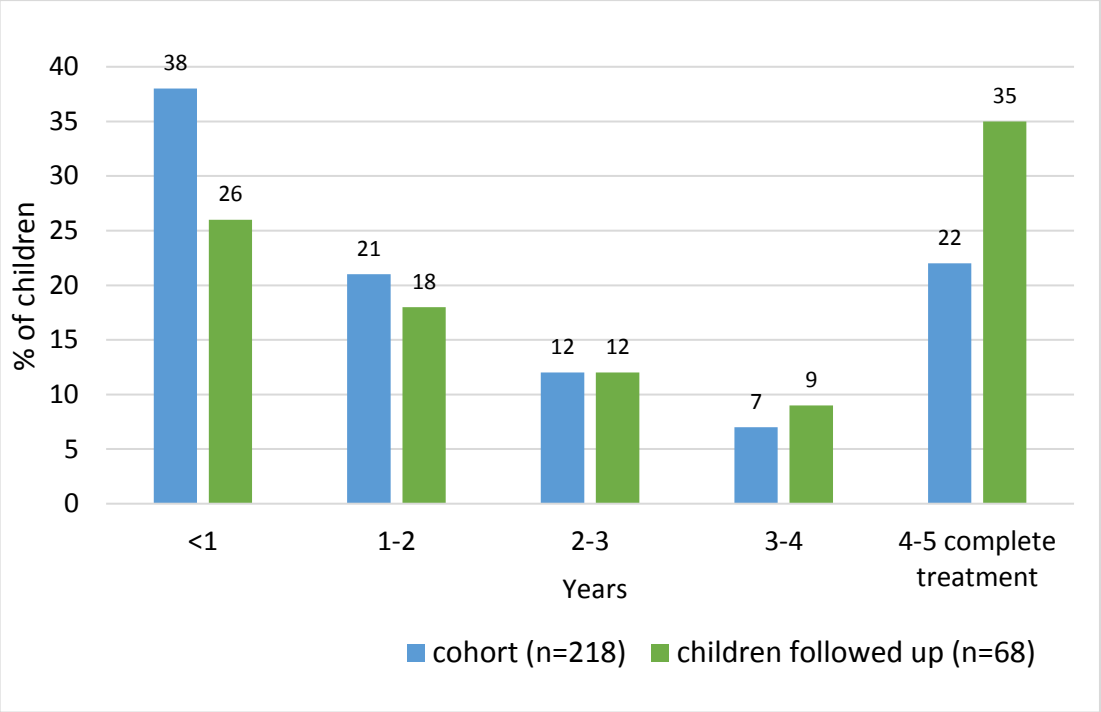

Figure 1: Length of time child attended clubfoot clinic appointments

There was good inter-observer agreement on the physical assessment of the Bangla tool, the ACT score tools and the relapse assessment, with an intra-class coefficient (ICC) of ≥ 0.82 on all criteria (Table 2). An ICC of 1.00 demonstrates perfect correlation.

Table 1: Inter-observer variation for outcome measures (ICC >75 = good consistency)

Outcome Measure	ICC	95%CI
Bangla Score		
1. Happy with child's feet?	0.96	0.94 - 0.98
2. Recommend to others?	1.00	1.00
3. Does child play with others?	1.00	1.00
4. Does child wear shoes of choice?	0.97	0.95 - 0.98
5. Does child have pain?	1.00	1.00
6. Squatting	1.00	1.00
7. Walking	1.00	1.00
8. Running	1.00	1.00
9. Up/down stairs	1.00	1.00
10a. Heel position L	0.94	0.88 - 0.97
10b. Heel position R	0.98	0.97 - 0.99
11a. Ankle range L	0.82	0.66 - 0.90
11b. Ankle range R	0.99	0.98 - 0.99
Relapse assessment		
1A - reduced DF	0.96	0.93 - 0.98
2A - fixed equinus	1.00	1.00
1B - dynamic supination, flex add	0.88	0.79 - 0.93
2B - fixed forefoot add	1.00	1.00
3 - 2 or more deformities	1.00	1.00
ACT score		
1. Foot is plantigrade	0.99	0.98 - 0.99
2. Does child complain of pain?	1.00	1.00
3. Can child wear shoes of choice?	0.99	0.99 - 1.00
4. How satisfied is the carer?	1.00	1.00

In all the children who were followed up (n=68) the success of treatment with different scores varied between 56% and 89% (Table 2). In the children who completed casting (63) it was between 57% and 93% (Table 3); and in the children who completed casting and at least two years of bracing (38) it was from 58% to 97% (Table 4). The individual category calculations for each outcome measurement are in additional files 2 - 5.

Table 2: Results of cohort of children followed up (n=68)

Outcome measure	Poor <49 N (%)	Fair: 50 – 69 N (%)	Good: 70 – 84 N (%)	V Good: 85 – 100 N (%)
Roye*	4 (6%)	3 (5%)	20 (30%)	39 (59%)
Total Roye*	7 (11%)		59 (89%)	
Bangla	12 (17%)	16 (24%)	15 (22%)	25(37%)
Total Bangla	28 (41%)		40 (59%)	
ACT score	7 (10%)	12 (18%)	13 (19%)	36 (53%)
Total ACT score	19 (28%)		49 (72%)	
	Cannot achieve plantigrade		Achieved plantigrade or better	
Plantigrade	13 (19%)		55 (81%)	
Any form of relapse	Yes		No	
	30 (44%)		38 (56%)	
Requires referral for further intervention	Yes		No	
	16 (24%)		52 (76%)	

*data missing for 2 children

Table 3: Results of cohort of children followed up who completed casting (n=63)

Outcome measure	Poor <49 N (%)	Fair: 50 – 69 N (%)	Good: 70 – 84 N (%)	V Good:85 – 100 N(%)
Roye*	2 (3%)	2 (3%)	19 (31%)	38 (63%)
Total Roye*	4 (7%)		57 (93%)	
Bangla	10 (16%)	15 (24%)	14 (22%)	24 (38%)
Total Bangla	25 (40%)		38 (60%)	
ACT score	5 (8%)	10 (16%)	13 (21%)	35 (55%)
Total ACT score	15 (24%)		48 (76%)	
	cannot achieve plantigrade		Achieved plantigrade or better	
Plantigrade	10 (16%)		53 (84%)	
Any form of relapse	Yes		No	
	27 (42%)		36 (57%)	
Requires referral for further intervention	Yes		No	
	14 (22%)		49 (78%)	

*data missing from 2 children

Table 4: Results of cohort of children followed up who completed >2 years bracing (n=38)

Outcome measure	Poor <49 N (%)	Fair: 50 – 69 N (%)	Good: 70 – 84 N(%)	V Good:85 – 100 N(%)
Roye*	1 (3%)	1 (3%)	10 (28%)	24 (66%)
Total Roye*	2 (6%)		34 (94%)	
Bangla	3 (8%)	10 (26%)	9 (24%)	16 (42%)
Total Bangla	13 (34%)		25 (66%)	
ACT score	1 (3%)	5 (13%)	9 (24%)	23 (60%)
Total ACT score	6 (16%)		32 (84%)	
	cannot achieve plantigrade		Achieved plantigrade or better	
Plantigrade	1 (3%)		37 (97%)	
Any form of relapse	Yes		No	
	16 (42%)		22 (58%)	
Requires referral for further intervention	Yes		No	
	5 (13%)		33 (87%)	

*data missing from 2 children

The results of success as defined by the outcome measures are illustrated in Figure 2, with the gold standard (full clinical assessment) results at the base of the figure. In the cohort, the relapse score and the Bangla tool had the lowest good outcome results of 56% and 59% respectively.

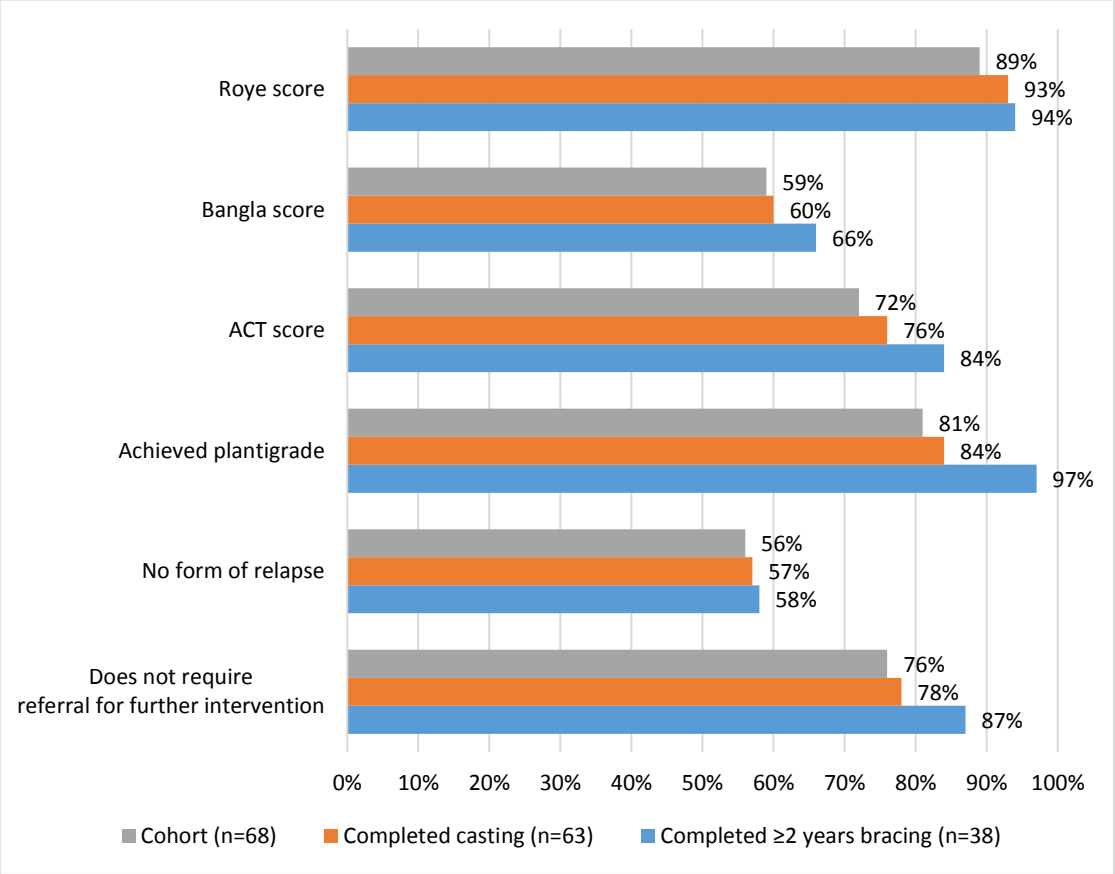

Figure 2: Comparison of outcomes to measure success

There was strong evidence for a difference between the outcomes of the Roye score and the Bangla score ($p<0.0001$), the Roye and the ACT score ($p=0.0013$), and the ACT and Bangla score ($p<0.0001$). It follows that none of these assessments can provide essentially the same estimate of success as the other measures.

There was a difference in the relative proportion of the cohort with relapse and plantigrade foot when assessed with Fischer's exact test ($p=0.012$). The binary outcomes are therefore not interchangeable.

No adverse events occurred as a result of any of the outcome measures undertaken. When compared to the standard of full clinical assessment and the subsequent decision on the need for referral for further intervention, the Roye score had a sensitivity of 31.8% (95%CI: 13.9 – 54.9%) and a specificity of 100% (95%CI: 92 – 100%), with positive and negative predictive values of 100% and 74.6% respectively.

The Bangla score demonstrated 79.2% (95%CI: 57.8 – 92.9%) sensitivity and 79.5% (95%CI: 64.7 – 90.2%) with 67.9% positive predictive and 87.5% negative predictive values, and the ACT score had 79.2% (95%CI: 57.8 – 92.9%) sensitivity and 100% (95%CI: 92 – 100%) specificity in predicting the need for referral, with positive and negative predictive values of 100% and 89.8% respectively. Of the 44 children that did not require referral for further intervention, all achieved plantigrade or more (positive predictive value: 100%) and of those who did require referral (n=24), 14 were identified with the plantigrade assessment (achieved less than plantigrade). The relapse score was most restrictive in identifying good outcome. False positive and false negative scores are displayed in Table 5.

Table 5: A comparison of measurement methods with the need for referral for further intervention

Method	Unnecessary referral (false positive) n(%)	Missed Referral (false negative) n(%)
Roye (n=66)	0 (0%)	15 (22.7%)
Bangla (n=68)	10 (14.7%)	5 (7.4%)
ACT (n=68)	1 (1.5%)	5 (7.4%)
Plantigrade (n=68)	0 (0%)	10 (14.7)
Relapse (n=68)	13 (19.1%)	6 (8.8%)

Discussion

The Ponseti method is often administered by locally trained therapists through Africa, who may have no specialised physiotherapy or surgical support present in the clinics. They require a user-friendly assessment system to identify when clubfoot treatment is not working and when refer to a specialist for further management is indicated. This study found that five scoring systems that are used to report outcomes of clubfoot treatment provided a wide spectrum of success (from 56% to 89% of cases) in a cohort with 3.5 – 5 years of follow up. When compared with the standard of clinical assessment, missed referrals ranged from 7.4% (Bangla and ACT scores) to 22.7% (Roye score). The measurements assess different aspects of clubfoot correction, from parent reported outcome measures (the Roye score) to scores that include physical assessment (Bangla and ACT) and single measurements (plantigrade foot and evidence of relapse). Success improves in all measures with the completion of casting and at least two years of bracing.

Comparison to previous studies

There are limited studies that compare measurement tools in the same patient against which to compare our findings. However, success of treatment in the cohort is similar to other studies in sub-Saharan Africa (between 63% and 98% of cases) (9).The process outcomes of noncompliance and surgical intervention, often defined as failure, are reported to vary from 7% - 61% and 3% - 39.4% (15) respectively. Additional clubfoot measurement tools include a scoring system that was described by Ponseti and Laaveg (16). The authors describe the functional results as satisfactory in 88.5% of feet and further studies with this scoring system describe success as 89.3% (17). The criteria includes the need for a goniometer and the tool was therefore not included in evaluation of this cohort.

Use of outcome measures

The ease of administration and rate of incorrect classification in the tools used to measure success need to be considered when selecting an outcome measure. In this study the threshold on an outcome scoring system was defined from previous literature as 70% for the Roye, Bangla and ACT scores and the result of yes/no for evidence of relapse and plantigrade foot. Single item scales for assessment of individual children require no further calculation and may be easier to use in clinics (such as plantigrade foot or evidence of relapse), however their simplicity may not allow a full assessment of success. Multi-scale items prove difficult to transform into useful statistics without technology and are unlikely to be routinely used in clinic. This study found no clear agreement between the different outcome measurements in use.

All of the assessments used in this study have limitations. The Roye score has been validated in high income settings and parents in our study reported difficulty in answering the question of “How often does your child have problems finding shoes that he or she likes?” as it was understood to be related to the availability of a variety of shoes. The Bangla score took the longest time to transform with statistical analysis. Acceptability and feasibility of the ACT score is needed to be studied in future research. With regard to the relapse score, Bhaskar et al considered ankle dorsiflexion <15 degrees with knee in extension as grade IA relapse. This may be a reason for the restriction in defining good outcome as an evaluation of 85 normal feet in children found that the mean ankle dorsiflexion was 12.8 degrees with knees in extension (18). Greater than 15 degrees may therefore be difficult to achieve.

Relationship between the outcome measures and clinical assessment

The Bangla and ACT tool were most helpful in predicting the need for referral for further intervention (specialist opinion or for further manipulation and casting). The five referrals that were missed with the ACT score were children who required review of a mobile curvature of the lateral border of the foot or supination in swing phase, neither of which are assessed with the score. Despite this, the ACT tool demonstrates the best diagnostic accuracy for the need for referral for further intervention.

Strengths and limitations of study

This study reports on five measurements of success in a cohort at 3.5 – 5 years from initial treatment with a total analysis time under observation of 171.3 years. Repeat phone calls facilitated assessments when caregivers were initially unavailable. Two independent raters reduced the likelihood of reporting bias and all outcome measures were verified by the reference standard. The threshold for diagnostic accuracy was based on previous studies and was defined prior to the study. There were also study limitations. No distinction between a clubfoot that may not have been fully corrected and a relapsed clubfoot was made, and all cases with elements of the deformity were classified with the relapse score, which may be a source of potential bias that underestimates the accuracy of the relapse score. The tools were chosen based on ease of use in low resource high volume clinics and were not all initially developed to identify need for referral for further intervention.

Implications for practice

Maintenance of the corrected foot position with a foot abduction brace (FAB) constitutes an important part of treatment. Barriers to routine measurement include limited time available for in-depth assessment in busy clinics and a lack of technology needed to measure and interpret results for more complicated functional assessments. Successful programmes need to demonstrate the quality of care that is delivered, account for the resources that are allocated and verify that children are receiving the best possible care in relation to these resources (19). It follows that clubfoot clinics should measure the outcomes of their interventions for comparison to other models of care or to other types of service delivery. Task shifting and task sharing between orthopaedic and non-specialised health workers in some clinics means that outcome measures are even more important as teams expand. As older children are being treated with the principles of the Ponseti method (20) and the method has been shown to be cost effective (21), expert guidance on assessment and measurement in clubfoot care is needed. Healthcare workers starting to work in clubfoot care require training on

what measurement instruments to use, how to use them and what to do with the results. Proposed solutions to service delivery include increasing provider's knowledge and skill (22) and this includes how success is measured. Implementing outcome measures and improving quality inevitably involves change and supervision and mentorship is required through this process.

Conclusion

It is important that trained clubfoot therapists monitor the results of treatment and know when to refer children for specialist review. This study found no clear agreement between the different scores in use. When compared to the normal practice of full clinical assessment, the measurement tool with the best evidence for diagnostic accuracy was the ACT tool.

List of abbreviations

ACT: Assessing Clubfoot Treatment

ICC: Intra-class correlation coefficient

Declarations

Ethics approval and consent to participate: Ethical approval for this study was granted by the Medical Research Council of Zimbabwe (MRCZ) and the London School of Hygiene & Tropical Medicine (LSHTM)(ref:11132 /RR/4725). All children and their caregivers were read an information sheet about the study and given an opportunity to ask questions. If they agreed to participate, written consent was taken from the caregiver who remained present throughout the assessment as per national requirements.

Consent for publication: not applicable

Availability of data and material: All data generated or analysed during this study are included in this published article [and its supplementary information files]. The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests: The authors declare that they have no competing interests

Funding: The Beit Trust, MiracleFeet, ZANE as scholarship to TS

Authors' contributions: TS, CL and AF designed the study. TS, MG and RM designed the study protocol. TS analysed and interpreted the data. TS wrote the first draft of the manuscript. MG, RM, AF and CL reviewed and edited the manuscript. All authors read and approved the final manuscript.

Acknowledgements: Zimbabwe Ministry of Health and Child Welfare, Parirenyatwa clubfoot clinic staff, Ryan Bathurst ZSCP, Debra Mudariki, Memory Mwadziwana and Mediatrice Mutsambi for data collection

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Appendix 1. The STARD 2015 list

Section and topic	No	Item
Title or abstract		
	1	Identification as a study of diagnostic accuracy using at least one measure of accuracy (such as sensitivity, specificity, predictive values or AUC) **A comparison of outcome measures used to report clubfoot treatment with the Ponseti method: the predictive ability of need for further intervention**
Abstract		
	2	Structured summary of study design, methods, results and conclusions
Introduction		
	3	Scientific and clinical background, including the intended use and clinical role of the index test **In low resource settings through Africa the Ponseti method is administered by locally trained therapists. These therapists often work alone and have no specialised physiotherapy or surgical support present in the clinics or nearby. It is important that they have a user friendly assessment system with agreed criteria for when treatment is not working and referral to a specialist for further management is indicated.**
	4	Study objectives and hypotheses **We aim to:** **(1) compare the results of the Ponseti method of clubfoot management at three to five years from initial correction using five different outcome measures.** **(2) explore the diagnostic accuracy of the outcome measures (the ability of the assessments to discriminate between the need for referral for further intervention and a successful outcome)**
Methods		
Study design	5	Whether data collection was planned before the index test and reference standard were performed (prospective study) or after (retrospective study) **Data were collected before the index test and reference standard were performed**
Participants	6	Eligibility criteria **Cohort study: there was one set of eligibility criteria for all study participants**
	7	On what basis potentially eligible participants were identified (such as symptoms, results from previous tests, inclusion in registry) **All children with a diagnosis of unilateral or bilateral idiopathic clubfoot who started treatment with the Ponseti method at the study hospital between 22nd March 2011 and 23rd April 2013 (25 months) were included in the cohort.**
	8	Where and when potentially eligible participants were identified (setting, location and dates) **Treated at Parirenyatwa Hospital, Harare between 22nd March 2011 and 23rd April 2013**
	9	Whether participants formed a consecutive, random or convenience series **The included study participants are a consecutive series of all patients evaluated for eligibility at the study location and satisfying the inclusion criteria**

Test methods	10a	Index test, in sufficient detail to allow replication **The index tests were performed as per guidelines in the published papers and references provided**
	10b	Reference standard, in sufficient detail to allow replication **Children were examined independently in January 2017 by the two physiotherapists and a decision was made if referral for further intervention (re-casting or surgical review) was required.** **Details include: Clinical examination composed observation, physical assessment and functional review; it included assessment of passive and active range of motion (plantiflexion, dorsiflexion, eversion, inversion of the foot and knee extension), muscle strength tests of the calf and evertors of the foot), heel raises, squatting ability and gait analysis (walking and running).**
	11	Rationale for choosing the reference standard (if alternatives exist) **The most frequently used approach to measuring whether the Ponseti method has been successful (or not) is clinical assessment. In sub-Saharan Africa 68% to 98% of cases are reported to have a successful outcome with the Ponseti method (4)**
	12a	Definition of and rationale for test positivity cut-offs or result categories of the index test, distinguishing pre-specified from exploratory **References given and limitations outlined in the discussion section:** **The threshold for diagnostic accuracy was set at 70% for the three scores with continuous scales (14) and positive/negative for the binary outcomes (7).**
	12b	Definition of and rationale for test positivity cut-offs or result categories of the reference standard, distinguishing pre-specified from exploratory **The threshold for diagnostic accuracy was based on previous studies and was defined prior to the study. It was set at 70% for the three scores with continuous scales (14) and positive/negative for the binary outcomes (7)**
	13a	Whether clinical information and reference standard results were available to the performers or readers of the index test **Categorisation of information for each test is provided in the web appendices**
	13b	Whether clinical information and index test results were available to the assessors of the reference standard **The clinical information and test results were available to the assessors of the reference standard and the reference standard was consensus on a decision for further referral or not**
Analysis	14	Methods for estimating or comparing measures of diagnostic accuracy **Sensitivity, specificity, positive and negative predictive values were calculated for the five measures and compared to full clinical assessment (gold standard). The results are categorised as true positive (TP), false positive (FP) (referred but not needed), true negative (TN), and false negative (FN)(should have been referred but missed).**
	15	How indeterminate index test or reference standard results were handled **Two assessments by individual physiotherapists and subsequent consensus was required for the standard reference result to avoid indeterminate results**
	16	How missing data on the index test and reference standard were handled **Missing data were excluded and noted on all tables of results**

	17	Any analyses of variability in diagnostic accuracy, distinguishing pre-specified from exploratory **Post hoc analyses was not performed**
	18	Intended sample size and how it was determined **Sample size was based on of the number of children first attending a single clinic over a period of 25 months and the follow up of these children. 31% (68/218) of the cohort attended for review and were assessed**
Results		
Participants	19	Flow of participants, using a diagram **Reference of flow diagram in original cohort analysis is given and explanation of sampling technique included "The phone numbers of all carers of the cohort children were extracted from the clinic records in January 2017 and contact with them was attempted at least three times. Caregivers and their children were invited to attend the study. The children were between 3.5 and 5 years from initial casting."**
	20	Baseline demographic and clinical characteristics of participants **Included in the first paragraph of results**
	21a	Distribution of severity of disease in those with the target condition **Included in the first paragraph of results**
	21b	Distribution of alternative diagnoses in those without the target condition **n/a due to selection criteria**
	22	Time interval and any clinical interventions between index test and reference standard **Independent assessment occurred on the same day between the index test and the reference standard so as to avoid any change in condition of the clubfoot position**
Test results	23	Cross tabulation of the index test results (or their distribution) by the results of the reference standard **Outlined in results and full tables included in web appendix**
	24	Estimates of diagnostic accuracy and their precision (such as 95% CIs) **Provided in results**
	25	Any adverse events from performing the index test or the reference standard **No adverse events occurred as a result of any of the outcome measures undertaken**
Discussion		
	26	Study limitations, including sources of potential bias, statistical uncertainty and generalisability **Included in discussion**
	27	Implications for practice, including the intended use and clinical role of the index test **Included in discussion**
Other information		
	28	Registration number and name of registry **This cohort study was not registered**
	29	Where the full study protocol can be accessed **The design and rationale of the study was not previously published**
	30	Sources of funding and other support; role of funders **Included in acknowledgements**

Appendix 2. Summary of outcomes: Roye Score

The total score includes all 10 items. Scores are computed by reverse-coding, summing the responses, and then dividing the result by the number of items completed, ie, scores are a mean of the responses. For ease of interpretation, the scores are linearly transformed to a 0 to 100 scale with 100 being best, so that 1 = 0, 2 = 33.3, 3 = 66.7, and 4 = 100 for questions with four possible answers. The one yes-or-no item (ever complains of pain in affected foot) was coded yes = 0 or no = 100.

Table 1: Results of Roye score for cohort (N=66) *results from 2 children missing as attended without primary carer

Roye score (cohort followed up, n=66*)	Very satisfied N (%)	Somewhat satisfied N (%)	Somewhat dissatisfied N (%)	Dissatisfied N (%)		Individual Score Mean % (95%CI)	Subscale Mean % (95%CI)
1. satisfied with status of foot	42 (64)	15 (23)	5 (7)	4 (6)	Satisfaction	81 (74 - 89)	85 (80 - 90)
2. satisfied with appearance	43 (65)	14 (21)	5 (8)	4 (6)		82 (75 - 89)	
	Never N(%)	Sometimes N(%)	Usually N(%)	Always N(%)			
3. How often is your child teased?	54 (82)	8 (12)	4 (6)	0 (0)		91 (87 - 96)	
4. How often does your child have problems finding shoes that fit?	49 (74)	10 (15)	3 (5)	4 (6)		86 (79 - 93)	
5. How often does your child have problems finding shoes that he or she likes?	45 (68)	15 (22)	3 (5)	3 (5)		85 (78 - 91)	
	No N (%)	Yes N(%)					
6. Does your child ever complain of pain in his or her [affected] foot?	26 (39)	40 (61)			Function	61 (49 - 73)	87 (82 - 91)
	Not at all limited N(%)	Somewhat limited N(%)	Moderately limited N(%)	Very limited N(%)			
7. How limited is your child in his or her ability to walk?	60 (91)	5 (8)	1 (1)	0 (0)		96 (94 - 99)	
8. How limited is your child in his or her ability to run?	52 (79)	10 (15)	3 (5)	1 (1)		90 (84 - 96)	
	Never N(%)	Sometimes N(%)	Usually N(%)	AlwaysN(%)			
9. How often does your child complain of pain during heavy exercise?	46 (70)	18 (27)	1 (1.5)	1 (1.5)		88 (84 - 93)	
10. How often does your child complain of pain during moderate exercise?	61 (92.5)	4 (6)	1 (1.5)	0 (0)		97 (94 - 100)	

Table 2: Results of Roye score for those who completed casting (N=61)

Roye score (completed casting) n=59	Very satisfied N (%)	Somewhat satisfied N (%)	Somewhat dissatisfied N (%)	Dissatisfied N (%)		Individual Mean % (95%CI)	Subscale Mean % (95%CI)
1. satisfied with status of foot	41 (67.2)	13 (21.3)	5 (8.2)	2 (3.3)	Satisfaction	84 (77 - 91)	87 (83 - 92)
2. satisfied with appearance	42 (68.8)	12 (19.7)	5 (8.2)	2 (3.3)		85 (78 - 91)	
	Never N(%)	Sometimes N(%)	Usually N(%)	Always N(%)			
3. How often is your child teased?	52 (85.2)	7 (11.5)	2 (3.3)	0 (0)		94 (90 - 98)	
4. How often does your child have problems finding shoes that fit?	48 (78.7)	8 (13.1)	2 (3.3)	3 (4.9)		89 (82 - 95)	
5. How often does your child have problems finding shoes that he or she likes?	42 (68.9)	14 (22.9)	3 (4.9)	2 (3.3)		86 (79 - 92)	
	No N (%)	Yes N (%)					
6. Does your child ever complain of pain in his or her [affected] foot?	39 (63.9)	22 (36.1)			Function	64 (52 - 76)	88 (84 - 92)
	Not at all limited N (%)	Somewhat limited N (%)	Moderately limited N (%)	Very limited N (%)			
7. How limited is your child in his or her ability to walk?	56 (91.8)	5 (8.2)	0 (0)	0 (0)		97 (95 - 100)	
8. How limited is your child in his or her ability to run?	49 (80.3)	10 (16.5)	1 (1.6)	1 (1.6)		92 (87 - 97)	
	Never N (%)	Sometimes N (%)	Usually N (%)	Always N (%)			
9. How often does your child complain of pain during heavy exercise?	44 (72.1)	16 (26.2)	0 (0)	1 (1.7)		90 (85 - 94)	
10. How often does your child complain of pain during moderate exercise?	58 (95.1)	3 (4.9)	0 (0)	0 (0)		98 (97 - 100)	

Table 3: Results of Roye score for those who completed ≥2 years bracing (N=38)

Roye score (completed ≥2 years bracing) n=36	Very satisfied N (%)	Somewhat satisfied N (%)	Somewhat dissatisfied N (%)	Dissatisfied N (%)		Individual Mean % (95%CI)	Subscale Mean % (95%CI)
1. satisfied with status of foot	27 (75%)	7 (19%)	1 (3%)	1 (3%)	Satisfaction	89 (81 -97)	89 (82 – 95)
2. satisfied with appearance	28 (78%)	6 (16%)	1 (3%)	1 (3%)		90 (82 – 97)	
	Never N(%)	Sometimes N(%)	Usually N(%)	Always N(%)			
3. How often is your child teased?	32 (89%)	3 (8%)	1 (3%)	0 (0%)		95 (91 – 100)	
4. How often does your child have problems finding shoes that fit?	27 (75%)	5 (14%)	1 (3%)	3 (8%)		85 (75 – 95)	
5. How often does your child have problems finding shoes that he or she likes?	25 (69%)	7 (19%)	2 (6%)	2 (6%)		84 (75 – 94)	
	No N (%)	Yes N (%)					
6. Does your child ever complain of pain in his or her [affected] foot?	27 (75%)	9 (25%)			Function	75 (60 – 90)	91 (87 – 96)
	Not at all limited N (%)	Somewhat limited N (%)	Moderately limited N (%)	Very limited N (%)			
7. How limited is your child in his or her ability to walk?	34 (94%)	2 (6%)	0 (0%)	0 (0%)		98 (96 – 100)	
8. How limited is your child in his or her ability to run?	29 (81%)	6 (17%)	0 (0%)	1 (3%)		92 (84 – 98)	
	Never N (%)	Sometimes N (%)	Usually N (%)	Always N (%)			
9. How often does your child complain of pain during heavy exercise?	28 (78%)	8 (22%)	0 (0%)	0 (0%)		93 (88 – 97)	
10. How often does your child complain of pain during moderate exercise?	34 (94%)	2 (6%)	0 (0%)	0 (0%)		98 (96 – 100)	

*data from 2 children missing

Appendix 3. Summary of outcomes: Bangla score

'Grading'	Very good	Good	Fair	Poor
%	85-100	70 - 85	60-70	<50

A. Parent rating subscore /5 where yes = +1, don't know = 0 and no = -1
B. Gait assessment subscore /4 where yes = +1, not fully – 0, no = -1
C. Clinical examination /2 where valgus/>DF = +1, straight/90 degrees = 0 and varus/<0 df = -1

Scores for bilateral cases were halved to achieve same scale/foot for section C/clinical examination

Total score converted to % indicated quality/grade of individual child's results from treatment

Table 1: Results of Bangla score for entire cohort (n=68)

A. Parent rating	Yes +1 N (%)	Don't know 0 N (%)	No -1 N (%)	Mean scores (%)	Rating Number
1. Happy with child's feet	57 (84)	2 (3)	9 (13)	71	"Good"
2. Recommend to others?	68 (100)	0 (0)	0 (0)	100	
3. Does child play with others?	68 (100)	0 (0)	0 (0)	100	
4. Does child wear shoes of choice?	53 (78)	0 (0)	15 (22)	56	
	Yes -1 N (%)	Don't know 0 N (%)	No +1 N (%)		
5. Does child have pain?	22 (32)	0 (0)	46 (68)	35	
Parental Rating sub score				72%	
B. Gait assessment	Yes +1 N (%)	Not fully/ with assistance 0 N (%)	No -1 N (%)	Mean scores (%)	"Very good"
6. Squatting	66 (97)	0 (0)	2 (3)	94	
7. Walking	67 (99)	1 (1)	0 (0)	99	
8. Running	67 (99)	0 (0)	1 (1)	98	
9. Up/down steps	67 (99)	0 (0)	1 (1)	98	
Gait assessment sub score				97%	
C. Clinical examination	Valgus +1 N (%)	Straight 0 N (%)	Varus -1 N (%)	Mean scores (%)	"Fair"
10. Heel position - left * n=53	2 (4)	32 (60)	19 (36)	32	
Heel position - right ** n=48	4 (8)	28 (58)	16 (34)	25	
	>0 dorsiflexion +1 N (%)	0/90 degrees 0 N (%)	<0 dorsiflexion N (%)	Mean scores (%)	
11. Ankle range - left* n=53	28 (53)	17 (32)	8 (15)	38	
Ankle range - right** n=48	23 (48)	15 (31)	10 (21)	27	
Clinical examination sub score (35 bilateral)				61%	
Total score				**77%**	"Good"

Table 2: Bangla score results from children who completed casting (n=61)

A. Parent rating	Yes +1 N (%)	Don't know 0 N (%)	No -1 N (%)	Mean scores (%)	Rating Number
1. Happy with child's feet	52 (85)	2 (3)	7 (12)	75	"Very good"
2. Recommend to others?	61 (100)	0 (0)	0 (0)	100	
3. Does child play with others?	61 (100)	0 (0)	0 (0)	100	
4. Does child wear shoes of choice?	49 (80)	0 (0)	12 (20)	59	
	Yes -1 N (%)	Don't know 0 N (%)	No +1 N (%)		
5. Does child have pain?	18 (30)	0 (0)	43 (70)	40	
Parental Rating sub score				75%	
B. Gait assessment	Yes +1 N (%)	Not fully/ with assistance 0 N (%)	No -1 N (%)	Mean scores (%)	"Very good"
6. Squatting	60 (98)	1 (2)	0 (0)	97	
7. Walking	61 (100)	0 (0)	0 (0)	100	
8. Running	61 (100)	0 (0)	0 (0)	100	
9. Up/down steps	61 (100)	0 (0)	0 (0)	100	
Gait assessment sub score				99%	
C. Clinical examination	Valgus +1 N (%)	Straight 0 N (%)	Varus -1 N (%)	Mean scores (%)	"Very good"
10. Heel position - left * n=46	2 (4)	28 (61)	16 (35)	38	
Heel position - right **n=42	3 (7)	25 (60)	14 (33)	41	
	>0 dorsiflexion +1 N (%)	0/90 degrees 0 N (%)	<0 dorsiflexion N (%)	Mean scores (%)	
11. Ankle range - left* n=46	25 (54)	16 (35)	5 (11)	42	
Ankle range - right** n=42	21 (50)	14 (33)	7 (17)	33	
Clinical examination sub score (30 bilateral)				77%	
Total score				**84%**	"Very good"

Table 3: Bangla score results from children who completed ≥2 years bracing (n=38)

A. Parent rating	Yes +1 N (%)	Don't know 0 N (%)	No -1 N (%)	Mean scores (%)	Rating Number
1. Happy with child's feet	34 (89%)	0 (0%)	4 (11%)	79	"Very good"
2. Recommend to others?	38 (100%)	0 (0%)	0 (0%)	100	
3. Does child play with others?	38 (100%)	0 (0%)	0 (0%)	100	
4. Does child wear shoes of choice?	31 (82%)	0 (0%)	7 (18%)	63	
	Yes -1 N (%)	Don't know 0 N (%)			
5. Does child have pain?	28 (74%)	0 (0%)	10 (26%)	47	
Parental Rating sub score				**78**	
B. Gait assessment	Yes +1 N (%)	Not fully/ with assistance 0 N (%)	No -1 N (%)	Mean scores (%)	"Very good"
6. Squatting	38 (100%)	0 (0%)	0 (0%)	100	
7. Walking	38 (100%)	0 (0%)	0 (0%)	100	
8. Running	38 (100%)	0 (0%)	0 (0%)	100	
9. Up/down steps	38 (100%)	0 (0%)	0 (0%)	100	
Gait assessment sub score				100	
C. Clinical examination	Valgus +1 N (%)	Straight 0 N (%)	Varus -1 N (%)	Mean scores (%)	"Very good"
10. Heel position - left * n=46	1 (4%)	22 (73%)	7 (23%)	20	
Heel position - right **n=42	1 (4%)	17 (68%)	7 (28%)	24	
	>0 dorsiflexion +1 N (%)	0/90 degrees 0 N (%)	<0 dorsiflexion N (%)	Mean scores (%)	
11. Ankle range - left* n=46	19 (63%)	10 (33%)	1 (4%)	60	
Ankle range - right** n=42	15 (60%)	10 (40%)	0 (0%)	60	
Clinical examination sub score (30 bilateral)				82	
Total score				**76**	"Very good"

Appendix 4. ACT score

Table 1: ACT score categories

Score	The foot is plantigrade	Does your child complain of pain in their affected foot?	Can your child wear shoes of your/their choice?	How satisfied are you with your child's foot?
3	More than plantigrade ie some dorsiflexion	No	Always	Very satisfied
2	Plantigrade	Yes but it does not limit their activity	Usually	Somewhat satisfied
1	Does not reach plantigrade	Yes and it sometimes limits their activity	Sometimes	Somewhat dissatisfied
0	Less than plantigrade with adduction, cavus or varus	Yes and it often limits their activity	Never	Very dissatisfied

Table 2: Results of ACT score distribution

		3	2	1	0
		Children N (%)	Children N (%)	Children N (%)	Children N (%)
Cohort followed up (n=68)	Foot is plantigrade	33 (49%)	22 (32%)	7 (10%)	6 (9%)
	Complain of pain	44 (65%)	18 (27%)	5 (7%)	1 (1%)
	Wears shoe of choice	47 (69%)	13 (19%)	6 (9%)	2 (3%)
	Satisfied with foot	42 (62%)	14 (21%)	9 (13%)	3 (4%)
Completed casting (n=63)	Foot is plantigrade	31 (49%)	22 (35%)	7 (11%)	3 (5%)
	Complain of pain	43 (68%)	15 (24%)	5 (8%)	0 (0%)
	Wears shoe of choice	45 (71%)	12 (19%)	6 (10%)	0 (0%)
	Satisfied with foot	41 (65%)	12 (19%)	9 (14%)	1 (2%)
Completed ≥2 years bracing (n=38)	Foot is plantigrade	22 (58%)	15 (39%)	1 (3%)	0 (0%)
	Complain of pain	27 (71%)	9 (24%)	2 (5%)	0 (0%)
	Wears shoe of choice	27 (71%)	8 (21%)	3 (8%)	0 (0%)
	Satisfied with foot	28 (74%)	6 (16%)	4 (10%)	0 (0%)

**Interpretation of ACT score in similar manner to Roye score - the scores are linearly transformed to a 0 to 100 scale with 100 being best, so that 1 = 0, 2 = 33.3, 3 = 66.7, and 4 = 100

Appendix 5. Relapse score

Table 1: Assessment of relapse

Relapse type	Cohort Children (n=68) N (%)	Children completed casting (n=63) N (%)	Children completed ≥2years bracing (n=32)* N (%)
No relapse	37 (55%)	36 (57%)	18 (56%)
1A (decrease in ankle dorsiflexion from 15degrees to neutral with knee extension)	13 (19%)	13 (21%)	6 (19%)
2A Dynamic forefoot adduction or supination of foot	1 (1%)	1 (2%)	0 (0%)
1B Fixed equinus of any degree (passive correction to neutral not possible)	5 (7%)	4 (6%)	4 (13%)
2B Fixed adduction of forefoot and midfoot (fixed lateral curvature)	0 (0%)	0 (0%)	0 (0%)
3 (Two or more fixed deformities)	12 (18%)	9 (14%)	4 (13%)

*missing data from 6 children

Epilogue (Main findings and limitations)

In the children who were followed up (n=68), the success of treatment with different assessment tools varied between 56% and 93%.

The relapse score and the Bangla tool had the lowest successful outcome results of 56% and 59% respectively. The relapse score had the highest proportion of unnecessary referrals (19.1%) and the Roye score the highest proportion of missed referrals (22.7%), when compared to full clinical assessment.

The ACT score and the Bangla score missed the fewest number of children who needed referral (7.4%). The ACT score had no unnecessary referrals. When compared to full clinical assessment, the Bangla score demonstrated 79.2% (95%CI: 57.8 – 92.9%) sensitivity and 79.5% (95%CI: 64.7 – 90.2%) specificity and the ACT score had 79.2% (95%CI: 57.8 – 92.9%) sensitivity and 100% (95%CI: 92 – 100%) specificity in predicting the need for referral.

Study limitations include (a) the chosen tools were based on ease of use in low resource high volume clinics, and not all tools were initially developed to identify need for referral for further intervention; (b) no distinction was made between a clubfoot that may not have been fully corrected and a clubfoot that had elements of recurrence; and (c) the tools were only used to assess idiopathic clubfoot; findings may not be applicable to children with secondary clubfoot.

Chapter 10. Indicators to assess the functionality of clubfoot clinics in low resource settings: a Delphi consensus approach and pilot study.

Data collection and clubfoot clinic charts

Preamble

Evaluation of clinical services is central to the concept of quality improvement. Trained clubfoot therapists deliver services in designated clubfoot clinics in Zimbabwe and there is little comparative data on clubfoot clinic quality, or which indicators to measure.

Criteria to assess the functionality of these clubfoot clinics are needed to identify the areas that require attention to improve the overall service. For example, both a clinic with two excellent clubfoot therapists but no POP, and a clinic with all the equipment required for clubfoot treatment but no trained therapists are inadequate.

The study therefore investigates the components required for a good clubfoot service and clinic in Africa. The strengths and weaknesses of the current clubfoot clinics in Zimbabwe are identified by the extent to which these components are being provided. Recommendations are made on areas for future research.

This chapter comprises a submitted research paper, which includes two studies. First, a Delphi method was used to create a consensus definition on a set of indicators to assess the functionality of a clubfoot clinic in Africa. Second, a cross-sectional survey using ten indicators was performed of the 12 clubfoot clinics in Zimbabwe to assess their functionality and to make recommendations for improvement.

This paper was published in the journal International Health after peer review in May 2018.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?	International Health		
When was the work published?	May 2018		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion	n/a		
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Supervisor Signature: _____ **Date:** 9/5/18

Int Health
doi:10.1093/inthealth/ihy033

ORIGINAL ARTICLE

Indicators to assess the functionality of clubfoot clinics in low-resource settings: a Delphi consensus approach and pilot study

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Received 1 December 2017; revised 26 March 2018; editorial decision 11 April 2018; accepted 11 April 2018

Background: This study aims to determine the indicators for assessing the functionality of clubfoot clinics in a low-resource setting.

Methods: The Delphi method was employed with experienced clubfoot practitioners in Africa to rate the importance of indicators of a good clubfoot clinic. The consistency among the participants was determined with the intraclass correlation coefficient. Indicators that achieved strong agreement (mean≥9 [SD <1.5]) were included in the final consensus definition. Based on the final consensus definition, a set of questions was developed to form the Functionality Assessment Clubfoot Clinic Tool (FACT). The FACT was used between February and July 2017 to assess the functionality of clinics in the Zimbabwe clubfoot programme.

Results: A set of 10 indicators that includes components of five of the six building blocks of a health system—leadership, human resources, essential medical equipment, health information systems and service delivery—was produced. The most common needs identified in Zimbabwe clubfoot clinics were a standard treatment protocol, a process for surgical referrals and a process to monitor dropout of patients.

Conclusions: Practitioners had good consistency in rating indicators. The consensus definition includes components of the World Health Organization building blocks of health systems. Useful information was obtained on how to improve the services in the Zimbabwe clubfoot programme.

Keywords: clubfoot, congenital talipes equinovarus, Delphi, Ponseti, quality, Zimbabwe

Introduction

Measurement of the functionality (capability and suitability) of health facilities is central to the concept of quality improvement. It allows identification of opportunities to strengthen and improve health services and quality of care. Measurements of quality are difficult to compare and interpret[1] due to the complex nature of health facilities. Internationally comparable measures are scarce and there is a lack of data on facility quality assessments in low-resource settings.[2]

Clubfoot, or congenital talipes equinovarus (CTEV), is a rigid deformity of the foot and one of the most common congenital musculoskeletal malformations that affects mobility.[3] Incidence is estimated as 1.2 per 1000 live births.[4] Clubfoot correction of a high quality continues to be a key requirement for reducing disability and improving function related to the congenital foot deformity. The minimally invasive Ponseti technique[5] is now the most common method of treatment, and involves manipulation and casting and a regime of bracing. In high-income settings, this technique radically reduces the need for extensive corrective surgery and the corrective phase is reported to have a success rate of 98%.[6] Although the definition and measurement of success of treatment are being explored in resource-constrained settings,[7–9] the provision of appropriate care is difficult to define, measure and evaluate. It is increasingly evident that patient outcomes are not solely a function of efficacious clinical interventions and practices but are also affected by the quality of non-clinical services. Evidence and consensus on what clinic qualities to measure are lacking.

A structured consensus technique that may be used to reach agreement about health care quality indicators[10] is the Delphi method. The anonymous opinions of participants are sought through a sequential process[11] and this allows equal weight to be given to all participants.[12,13] The collated group responses are fed back to participants after the completion of each round of questionnaires. Establishing consensus does not ensure validity; however, agreement provides a basis for establishing criteria that are likely to have clinical sensibility.[14]

Strategies to improve services for children with clubfoot require consideration of the clubfoot clinic within the health system and the paradigm shift from condition-specific interventions to interventions that strengthen health systems. The interdependence between caregiver demand for clubfoot services, the identification and referral of clubfoot within the context of primary health care, the service provision of the clubfoot clinic and the enabling environment of the broader health facility is required for good clubfoot treatment. Within this health system–oriented approach, appropriate measures are needed to assess the functionality of clubfoot clinics.[15] There are few formal monitoring systems in place to inform this planning.[16] This study aims to determine the indicators for assessing the functionality of clubfoot clinics in low-resource settings by establishing a consensus among expert Ponseti trainers in the Africa region and then to use those indicators to assess the Zimbabwe clubfoot programme.

Materials and methods

Study design

This study was designed and reported according to the recommended guidelines for selection of health care quality indicators.[10] A Delphi method was employed with experienced clubfoot practitioners (Ponseti technique) throughout Africa to rate the importance of indicators of a 'good Ponseti clubfoot clinic'. Based on the final consensus definition, a set of questions was developed to form the Functionality Assessment Clubfoot

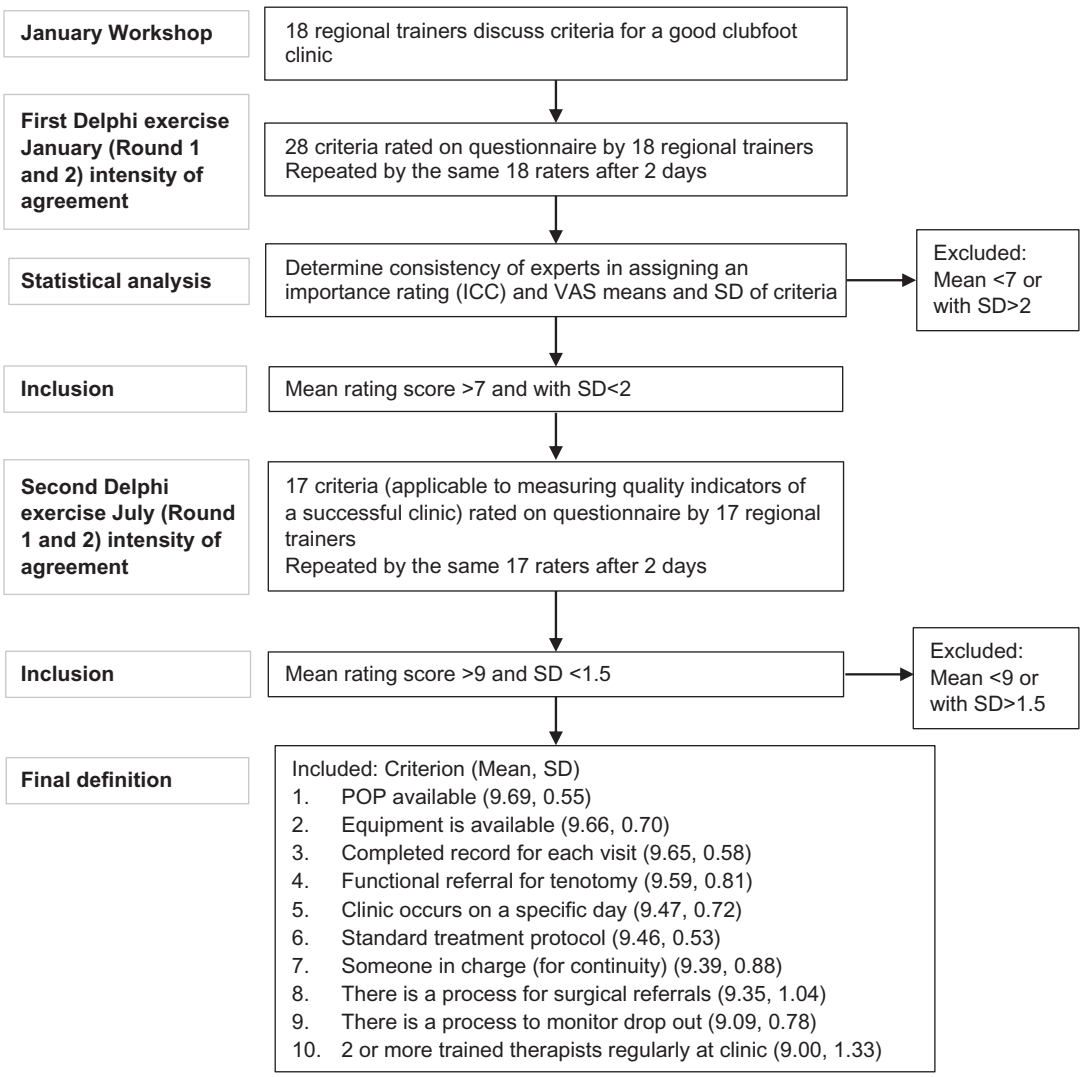

Figure 1. Flow chart of criterion selection.

Table 1. The FACT

Domain	Criteria	Rating				Score
		0	1	2	3	
Leadership	There is an identifiable person in charge of the clinic	In the past 8 weeks: There was no identifiable person in charge of the clinic	There was an identifiable person in charge of five or fewer clinics	There was an identifiable person in charge of six to seven clinics	There was an identifiable person in charge of every clinic	
Human resources	There are regularly two or more Ponseti-trained health care workers (HCWs) available at each clinic	In the past 8 weeks: There were fewer than two trained HCWs available at every clinic	There were two or more trained HCWs available in five or fewer clinics	There were two of more trained HCWs available in six to seven clinics	There were two or more trained HCWs available at every clinic	
Essential medical equipment	There is always plaster of Paris available	In the past 8 weeks: Plaster of Paris was not available for three or more clinics	Plaster of Paris was not available for two clinics	Plaster of Paris was not available for one clinic	There was always plaster of Paris available	
	There is always equipment (such as bucket and scissors/blade, tenotomy set, correct size braces) available	In the past 8 weeks: A bucket, scissors/blade, tenotomy set or correct size braces were not available for three or more clinics	A bucket, scissors/blade, tenotomy set or correct size braces were not available for two clinics	A bucket, scissors/blade, tenotomy set or correct size braces were not available for one clinic	There was always a bucket and scissors/blade, tenotomy set, correct size braces available	
Service delivery	There is a standard treatment protocol	There is no standard protocol for treatment of idiopathic clubfoot in children <2 y of age, the older child with clubfoot or non-idiopathic clubfoot	There is a standard verbal treatment protocol but nothing in writing for treatment of idiopathic clubfoot in children <2 y of age, the older child with clubfoot and non-idiopathic clubfoot	There is a written protocol for treatment of idiopathic clubfoot in children <2 y of age, the older child with clubfoot and non-idiopathic clubfoot, but it is not consistently used	There is a standard written protocol for treatment of idiopathic clubfoot in children <2 y of age, the older child with clubfoot and non-idiopathic clubfoot and it is followed consistently	
	There is a functioning referral system for tenotomy	In the two most recent tenotomy cases: There was no clinician to whom children requiring a tenotomy could be referred	The children were referred for tenotomy and there is no record of the outcome	The children were referred but the tenotomy was not completed as anticipated	The children were referred for tenotomy and it was completed as anticipated	
	There is a process for surgical referrals	In the two most recent cases who required surgery: There was no clinician to whom children requiring a surgical review could be referred	The children were referred and there is no record of the outcome	The children were referred and there is a record of the outcome without a plan for follow-up	The children were referred for surgery and there is a record of the outcome with a plan for follow-up	

Continued

Table 1. *Continued*

Domain	Criteria	Rating				Score
		0	1	2	3	
	There is a process to monitor dropout of patients	There is no process to monitor dropout of patients	There is a verbal process to monitor dropout of patients but nothing in writing	There is a written protocol to monitor dropout of patients but it is not consistently followed	There is a written protocol to monitor dropout of patients and it is consistently followed	
	The clinic occurs on a specific day, at minimum weekly	In the past 8 weeks: The clinic did not have an identified day for clubfoot treatment	The clinic has been held on the identified day(s) for less than six of the weeks	The clinic has been held on the identified day(s) for six to seven of the weeks	The clinic has been held on the identified day(s) every week	
Health information system	There is a completed clinic record for each patient visit	In the last 10 clinic records: ≤5 of the clinic records were filled in completely	6–7 of the records were filled in completely	8–9 of the clinic records were filled in completely	All of the clinic records were filled in completely	
Total score						

Clinic Tool (FACT). The FACT was used between February and July 2017 to assess the functionality of 12 clinics of the Zimbabwe national clubfoot programme.

Delphi method to develop a consensus definition

In the first Delphi exercise conducted in January 2016, 18 experienced Ponseti method clubfoot practitioners who are trainers in 10 national clubfoot programmes in Africa attended a workshop. They included orthopaedic surgeons, physiotherapists and orthopaedic technicians. The participants were chosen based on their knowledge and experience of the topic and willingness to participate.[17] Facilitated discussion in the workshop identified criteria (indicators) likely to demonstrate a well-functioning Ponseti clubfoot clinic. A questionnaire was then developed consisting of 28 potentially relevant indicators. It was pilot tested for understanding. The 18 trainers were invited to participate and rate each of the 28 indicators for their relative importance. A 10 cm visual analogue scale (VAS) with the anchors 'completely unimportant' and 'extremely important' was used. The paper questionnaires were completed by hand. The second round occurred 2 days later. The trainers were given the mean score and standard deviation (SD) for each indicator from the first round and asked to repeat the rating for the second round. No indicator was excluded and no discussion was allowed among the participants.

The consistency among the 18 trainers was determined with the intraclass correlation coefficient (ICC). The ICC is interpreted as follows: ≤0.40, poor consistency or large variation in opinion; 0.41–0.74, acceptable consistency; and ≥0.75, good consistency.[18]

In July 2016, 6 months after the first workshop, a second workshop was held with 17 different trainers of the Ponseti technique (from 10 countries). The experts were not considered to differ from the first group and their attendance in July was based on convenience of the workshop schedule. Indicators generated in January 2016 that had a mean VAS >7 (on the 10-point scale) with an SD <2 were included (21 indicators) in the questionnaire of the second Delphi exercise.

The two-round Delphi exercise, with an identical process to that undertaken in January 2016, was used to reach a consensus on indicators to assess the functionality of clubfoot clinics in low-resource settings and therefore define a well-functioning Ponseti clubfoot clinic. Of the 17 indicators rated, those that scored ≥9 with an SD <1.5 were considered to have high agreement and form the consensus definition. As there is variability in the measurement of distribution of scores in studies that use the Delphi method,[19] the thresholds for the VAS mean and SD were decided *a priori* and the cut-offs were selected to include indicators with high agreement.

The study methodology and course of action for the management of responses are outlined in Figure 1. The questionnaires answered in the second rounds of Delphi exercises 1 and 2 are in Supplementary files Appendix 1 and 2, respectively.

Assessment of clubfoot clinics in Zimbabwe using the consensus definition

A questionnaire based on the 10 indicators that met the criteria for the consensus definition was developed and piloted. The

questionnaire was reviewed by the Zimbabwe national clubfoot clinical supervisor and the researchers. Each question has four response categories, scored from 0 to 3, where 3 is the ideal result. The maximum clinic score is 30. A response matrix was developed to analyse the results (Table 1).

Data collection

Data were collected between February and July 2017 during routine clubfoot clinic mentoring visits by the clinical supervisor of the Zimbabwe Sustainable Clubfoot Programme. The questionnaire was paper based and completed by hand. Routine discussion with clinic staff, observation and review of equipment and clinic records provided data for completion of the questionnaire. The clubfoot clinic staff were informed of the outcome and subsequent recommendations discussed with the team. Data were collected from all 12 national clubfoot clinics.

Table 2. Indicators for a functioning clubfoot clinic ranked by mean score and SD from the second round: Delphi exercise 1, January 2016

	Indicator	Mean	SD
1	100% foot abduction brace uptake post-tenotomy	9.68	0.42
2	Functioning referral for tenotomy	9.49	0.67
3	Completed record for each visit	9.37	0.49
4	Someone in charge (for continuity)	9.32	0.63
5	Two or more trained therapists regularly at the clinic	9.28	0.76
6	Standardized treatment protocol	9.20	0.88
7	Plaster of Paris available	9.19	0.85
8	Clinic occurs on a specific day	9.13	1.12
9	Equipment is available	8.94	1.17
10	Counsellor contact with 90% of families at each clinic	8.89	0.78
11	There is a process for surgical referrals	8.61	1.38
12	Clinicians refer if there is no change in Pirani score for four visits	8.61	1.59
13	The nearest maternity ward refers all cases	8.51	1.96
14	Clinicians refer if >10 casts	8.41	1.24
15	The family is followed up if an appointment is missed	8.32	1.96
16	There is a clubfoot champion in charge of the clinic	7.96	2.64
17	90% of patients wear a foot abduction brace at 4 months	7.92	2.10
18	Children are <6 months of age at the first treatment	7.52	1.69
19	Braces are received within 2 weeks of ordering	7.52	2.53
20	Tenotomy coverage >90%	7.42	2.35
21	Relapse rate <10%	7.25	2.00
22	Tenotomy coverage >70%	7.10	2.00
23	Relapse rate <20%	5.99	2.05
24	Only 10% drop out after 6 months of bracing	5.95	1.82
25	Relapse rate not >30%	5.88	1.98
26	Only 25% drop out after 6 months	4.25	1.86
27	<70% tenotomy coverage	3.95	2.05
28	50% drop out after 6 months of bracing	2.83	2.19

Data management and analysis

All data were entered into an Excel 2000 (Microsoft, Redmond, WA, USA) spreadsheet. All data were managed and analysed using Stata 14.2 (StataCorp, College Station, TX, USA). A descriptive analysis compared the characteristics of the clinics. Proportions were calculated and a comparative analysis of criteria was explored.

Ethics

Ethical approval was granted by the Medical Research Council of Zimbabwe and the London School of Hygiene & Tropical Medicine.

Results

Delphi method to develop a consensus definition

In Delphi exercise 1 (January 2016) the response rate of trainers to the questionnaires was 94.4% (17/18) and in exercise 2 (July 2016) the rate was 94.1% (16/17).

Table 3. Indicators for a functioning clubfoot clinic ranked by mean score and SD from the second round: Delphi exercise 2, July 2016

	Indicators	Mean	SD
1	Plaster of Paris available	9.69	0.55
2	Equipment available	9.66	0.70
3	Completed record for each visit	9.65	0.58
4	Functional referral for tenotomy	9.59	0.81
5	Clinic occurs on a specific day	9.47	0.72
6	Standardized treatment protocol	9.46	0.53
7	Someone in charge (for continuity)	9.39	0.88
8	There is a process for surgical referrals	9.35	1.04
9	There is a process to monitor dropouts	9.09	0.78
10	Two or more trained therapists regularly at the clinic	9.00	1.33
11	The nearest maternity ward refers all cases	8.99	0.84
12	Clinicians refer if no change in Pirani score for four visits	8.96	1.04
13	Tenotomy coverage >70%	8.76	0.89
14	The family is followed up if an appointment is missed	8.71	1.06
15	Counsellor contact with 90% of families at each clinic	8.68	1.18
16	Clinicians refer if >10 casts	8.53	2.31
17	Children are <6 months of age at the first treatment	7.30	2.19

The consistency of Ponseti method trainers in Africa in rating indicators to assess the functionality of Ponseti clubfoot clinics was good. The first Delphi ICC had an external consistency of 0.96 (95% [CI 0.94–0.98]) and the second Delphi ICC had an external consistency of 0.84 (95% CI 0.70–0.93).

From the initial 28 indicators, 17 met the inclusion criteria for Delphi exercise 2. The results of the rating of each indicator by trainers in Africa are shown in Table 2 for Delphi exercise 1 (January 2016) and Table 3 for Delphi exercise 2 (July 2016).

Ten indicators in the second Delphi exercise met the *a priori* definition (mean score ≥9 [SD <1.5]) for inclusion in the consensus definition: plaster of Paris available, equipment is available, completed record for each visit, functional referral for tenotomy, clinic occurs on a specific day, standard treatment protocol available, someone in charge (for continuity), there is a process for surgical referrals, there is a process to monitor dropouts and two or more trained therapists are regularly at the clinic.

Assessment of clubfoot clinics in Zimbabwe using the consensus definition

The average clubfoot clinic score was 21 (range 16–26) (Table 4). All clubfoot clinics had an identifiable person in charge, two or more Ponseti clubfoot therapists that were regularly available and the clinic occurred on a specific day, at a minimum weekly (Figure 2). Indicators of a standard treatment protocol, a process for surgical referral and a process to monitor dropout of patients were overall the lowest scoring indicators of the clinics.

Regarding the health system building blocks, the indicators for leadership and human resources achieved full scores, while service delivery had the lowest score in all clinics (Table 4).

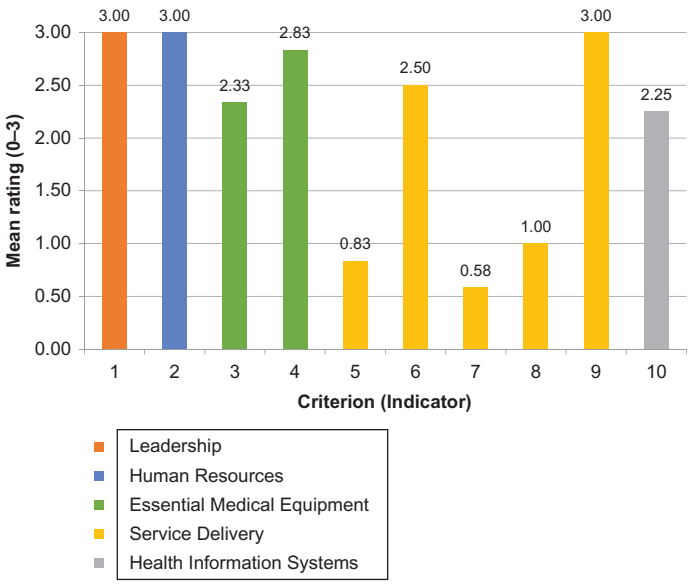

Figure 2. Mean scores for the 12 clubfoot clinics.
Legend:
(1) There is an identifiable person in charge of the clinic.
(2) There are regularly two or more Ponseti-trained health care workers available at each clinic.
(3) There is always plaster of Paris available.
(4) There is always equipment (e.g. bucket and scissors/blade, tenotomy set, correct size braces) available.
(5) There is a standard treatment protocol.
(6) There is a functioning referral system for tenotomy.
(7) There is a process for surgical referrals.
(8) There is a process to monitor dropout of patients.
(9) The clinic occurs on a specific day, at minimum weekly.
(10) There is a completed clinic record for each patient visit.

Discussion

This study determined the opinions of experts from 11 countries in Africa about the indicators for a successful Ponseti clubfoot clinic within health facilities. The aim of the Delphi method was to define criteria to evaluate the functionality of clubfoot clinics in low-resource settings, therefore regional trainers of the Ponseti method were deemed the most appropriate experts to participate in this context. A questionnaire was developed, based on the consensus indicators, to evaluate the functionality of the Zimbabwe clubfoot clinics.

Delphi exercise

The trainers had good consistency in rating indicators to assess the functionality of a clubfoot clinic. The consensus indicators include components of five of the six World Health Organization (WHO) building blocks of health system strengthening,[20] namely leadership, human resources, essential medical equipment, health information systems and service delivery.

Assessment of 12 clinics

The FACT took 15 min to complete and was undertaken as part of routine supervision visits. Of the 10 indicators piloted, leadership and human resources were found to score the highest in the Zimbabwe national clubfoot programme, with service delivery demonstrating the greatest need for improvement.

Comparison to other literature/previous studies

The indicators developed in this study reflect the WHO building blocks for health systems.[20] To our knowledge there are no quality indicators for clubfoot clinics published and evaluated in the literature; however, individual non-governmental organizations regularly use checklists and reporting templates for accountability and quality improvement purposes.

Strengths and limitations

This study has used many experts, in the context of Africa, to develop and rate criteria (indicators) of clubfoot clinic qualities that are viewed to be the most important. The response rate of the survey was high (94%). There are also study limitations. The panel in this study was selected for their expertise but may not be representative of all Ponseti treatment practitioners. Previous research has shown that panel composition influences ratings.[21] In addition, indicators will never completely capture the richness and complexity of a health system. Their design must be understood in

Table 4. Individual clinic scores

Health system building block	Leadership	Human resources	Medical equipment		Service delivery					Health information system	
Indicator	1	2	3	4	5	6	7	8	9	10	Total
Clinic ID	Identifiable person in charge	Two trained therapists	Plaster of Paris	Bucket/scissors/ blade/tenotomy set/braces	Standard treatment protocol	Tenotomy referral	Process for surgical referrals	Process to monitor dropouts	Clinic occurs on a specific day	Completed clinic record	Clinic score out of 30
1	3	3	3	2	0	2	1	1	3	3	21
2	3	3	3	2	0	3	0	1	3	3	21
3	3	3	1	3	1	2	0	1	3	2	19
4	3	3	1	3	1	3	2	1	3	3	23
5	3	3	1	3	1	0	0	1	3	1	16
6	3	3	3	3	1	3	1	1	3	3	24
7	3	3	3	3	1	3	0	1	3	3	23
8	3	3	1	3	1	2	0	1	3	0	17
9	3	3	3	3	1	3	0	1	3	3	23
10	3	3	3	3	1	3	0	1	3	2	22
11	3	3	3	3	1	3	3	1	3	3	26
12	3	3	3	3	1	3	0	1	3	1	21
Mean score	3.00	3.00	2.33	2.83	0.83	2.50	0.58	1.00	3.00	2.25	

context and there may be some indicators that we have not considered that may be important. For example, there was no consensus on the number of patients and one finding has been that hospitals caring for greater numbers of patients with similar conditions tend to have better outcomes for surgical procedures.[22,23] Consequently, while the questionnaire can provide valuable information, other data may be relevant to have a more complete understanding of the clubfoot clinic.

Implications

The data from this study provide useful information to assist in monitoring and improving services for children with clubfoot in low-resource settings. For example, clubfoot service provision in Zimbabwe utilizes a task-shifting approach, where trained 'clubfoot therapists' undertake manipulation and casting and brace reviews, and medical officers complete tenotomies. Tenotomies are undertaken in the outpatient clinic under local anaesthetic, and the decision of when to verbally refer for tenotomy rests with the clubfoot therapist. Surgical procedures that extend beyond a tenotomy require written referral to a specialist. This study highlights the need for defined care pathways to monitor standard procedures and contribute to a robust referral system.

Indicators are summary measures and no matter how valid they are, they will rarely by themselves motivate people to change. Using the questionnaire, clubfoot programme staff determined if all necessary elements were in place for a successful clinic that can deliver clubfoot care. Based on the results of the assessment, the need for standard treatment protocols, a process for surgical referrals and a process to monitor dropouts were identified in every clinic in Zimbabwe. These processes were discussed and a plan to address them in every clinic was created. The usefulness of this tool in clubfoot clinics is therefore twofold: (1) to assess the design and implementation of effective clubfoot clinic programmes and (2) to monitor services and highlight the improvements needed as the programme develops.

Future research

It is important that these indicators are tested in other situations to see if they are valid. The definition of indicators in greater detail (e.g. the development of a specific equipment list that includes braces and essential equipment for tenotomy) will identify gaps in resources and allow for clubfoot clinics to be appropriately equipped. Also, research questions such as how provider performance can be improved, responsiveness to change and why some clinics perform better than others may provide further insights into quality improvement.

Approaches to encourage completion of both the treatment and bracing phases include designing education resources with parents, who have unique insights about their challenges and situations but are often excluded from the design process about issues that directly affect their lives. An understanding of how caregivers can best be supported, when and where counselling is needed and mechanisms for delivery (e.g. peer group, one to one) warrant further investigation.

A well-equipped and managed clubfoot clinic may still provide poor care. A child's experience of care and parent-reported outcomes provide valuable insights on quality of care but are rarely measured. Agreement on how to measure these outcomes using metrics that are robust, comparable and financially efficient is required.

Conclusion

Appropriate measures are required to determine clubfoot clinic qualities and to compare different clinics. Using the Delphi method among experts from across Africa, we found a consensus for the most important indicators of a good clubfoot (Ponseti method) clinic. The consensus definition includes 10 indicators covering five of the six WHO building blocks of health system strengthening: leadership, human resources, essential medical equipment, service delivery and health information systems. The FACT indicators recommended are accompanied by descriptions of their technical properties, and methodological guidance is provided for their assessment.

In the Zimbabwe clubfoot clinics, the area of leadership scored the highest and the area of service provision had the greatest room for improvement. This article contributes to the data on clinic indicators globally. The data from this study provide useful information on planning services and may direct health care planning towards the areas of need.

Supplementary data

Supplementary data are available at *International Health* online (http://inthealth.oxfordjournals.org).

Authors' contributions: TS, AF and CL conceived the study. TS, AF and CL designed the study protocol. TS and DM collected data. TS analysed and interpreted the data. AF, CL and DM critically revised the manuscript for intellectual content. All authors read and approved the final manuscript.

Acknowledgements: The authors are grateful to the Zimbabwe Ministry of Health and Child Welfare for their support. A further thanks goes to all of those clubfoot practitioners whose motivation and skills shape the success of the national clubfoot programme. Lastly, we thank the Zimbabwe Sustainable Clubfoot Programme, without whose commitment and generosity, none of this would have been possible.

Funding: This work was supported by the Beit Trust, ZANE and MiracleFeet, who are gratefully acknowledged for scholarship funding provided to TS.

Competing interests: None declared.

Ethical approval: Ethical approval was granted by the Medical Research Council of Zimbabwe and the London School of Hygiene and Tropical Medicine.

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Appendix 1. Delphi Questionnaire (Round1)

How important are the following for a **good** clubfoot clinic:

How important are the following for a **good** clubfoot clinic:

1. The clubfoot clinic occurs on a specific day, at a minimum weekly (av:8.9)

0 1 2 3 4 5 6 7 8 9 10

2. The tenotomy rate is less than 70% (av 3.7)

0 1 2 3 4 5 6 7 8 9 10

3. The children first attend clinic under 6 months of age (av 7.3)

0 1 2 3 4 5 6 7 8 9 10

4. There is 25% drop out after 6 months of bracing (av 4.7)

0 1 2 3 4 5 6 7 8 9 10

5. There is always equipment available eg a bucket, scissors (av 8.9)

0 1 2 3 4 5 6 7 8 9 10

6. There is someone in charge of the clinic (av 9.1)

0 1 2 3 4 5 6 7 8 9 10

7. There is a place to refer to for surgery (av 8.5)

0 1 2 3 4 5 6 7 8 9 10

8. There are regularly two or more trained Ponseti therapists available at each clinic (av 8.9)

0 1 2 3 4 5 6 7 8 9 10

9. The tenotomy rate is more than 90% (av 6.9)

0 1 2 3 4 5 6 7 8 9 10

10. The relapse rate (as identified as requiring re-casting) is not above 30% (av 6.2)

0 1 2 3 4 5 6 7 8 9 10

11. The tenotomy rate is more than 80% (av 6)

0 1 2 3 4 5 6 7 8 9 10

12. There is a functioning referral system for tenotomy (av 9.3)

0 1 2 3 4 5 6 7 8 9 10

13. Braces can be obtained within two weeks of request (av 7.7)

0 1 2 3 4 5 6 7 8 9 10

14. There is always plaster of paris available (av 5.6)

0 1 2 3 4 5 6 7 8 9 10

15. There is 10 % drop out after 6 months of bracing (av 5.6)

0 1 2 3 4 5 6 7 8 9 10

16. The relapse rate (as identified as requiring re-casting) is below 10% (av 7.5)

0 1 2 3 4 5 6 7 8 9 10

17. There is 50% drop out after 6 months of bracing (av 3.3)

0 1 2 3 4 5 6 7 8 9 10

18. There is counsellor contact for 90% of patients on a clinic day (av 8.7)

0 1 2 3 4 5 6 7 8 9 10

19. There is referral if more than 10 casts are applied (av 8)

0 1 2 3 4 5 6 7 8 9 10

20. 90% of patients still use the SFAB appropriately at 4 months (av 8.5)

0 1 2 3 4 5 6 7 8 9 10

21. There is a completed record for each patient visit (av 9.4)

0 1 2 3 4 5 6 7 8 9 10

22. There is a standardised clinic schedule (av 8.9)

0 1 2 3 4 5 6 7 8 9 10

23. There is 100% FAB uptake post tenotomy (av 9.5)

0 1 2 3 4 5 6 7 8 9 10

24. The child is referred if there is no change Pirani score for 4 weeks (av 8.7)

0 1 2 3 4 5 6 7 8 9 10

25. The relapse rate (as identified as requiring re-casting) is below 20%(av 7)

0 1 2 3 4 5 6 7 8 9 10

26. The person in charge of the clinic is a champion for clubfoot

0 1 2 3 4 5 6 7 8 9 10

27. The close maternity hospitals refer their clubfoot patients

0 1 2 3 4 5 6 7 8 9 10

28. Each time a patient misses an appointment it is followed up

0 1 2 3 4 5 6 7 8 9 10

Appendix 2. Delphi Questionnaire (Round 2)

How important are the following for a **good** clubfoot clinic:

1. The clubfoot clinic occurs on a specific day, at a minimum weekly (av:9.2)

0 1 2 3 4 5 6 7 8 9 10

2. The children first attend clinic under 6 months of age (av 7.7)

0 1 2 3 4 5 6 7 8 9 10

3. There is always equipment available such as a bucket, scissors, water (av 9.3)

0 1 2 3 4 5 6 7 8 9 10

4. There is an identifiable person in charge of the clinic (av 9.3)

0 1 2 3 4 5 6 7 8 9 10

5. There is a process for surgical referrals (av 9.1)

0 1 2 3 4 5 6 7 8 9 10

6. There are regularly two or more trained Ponseti therapists available at each clinic (av 8.5)

0 1 2 3 4 5 6 7 8 9 10

7. The relapse rate (as identified as requiring re-casting) is not above 30% (av 8.2)

0 1 2 3 4 5 6 7 8 9 10

8. The tenotomy rate is more than 70% (av 8.5)

0 1 2 3 4 5 6 7 8 9 10

9. There is a functioning referral system for tenotomy (av 9.4)

0 1 2 3 4 5 6 7 8 9 10

10. Braces can be obtained within two weeks of request (av 8.9)

0 1 2 3 4 5 6 7 8 9 10

11. There is always plaster of paris available (av 9.6)

0 1 2 3 4 5 6 7 8 9 10

12. There is no more than 20 % drop out after 6 months of bracing (av 8.8)

0 1 2 3 4 5 6 7 8 9 10

13. There is a process to monitor drop out of bracing (av 9.1)

0 1 2 3 4 5 6 7 8 9 10

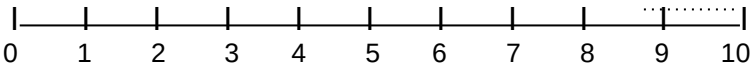
14. There is counsellor contact for 90% of patients on a clinic day (av 8.4)

0 1 2 3 4 5 6 7 8 9 10

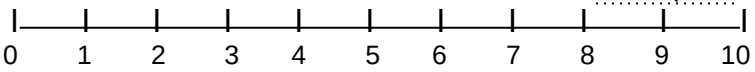
15. The child is referred if more than 10 casts are applied (av 8.9)

0 1 2 3 4 5 6 7 8 9 10

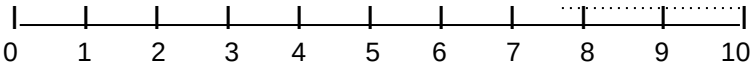
16. There is a completed record for each patient visit (av 9.7)



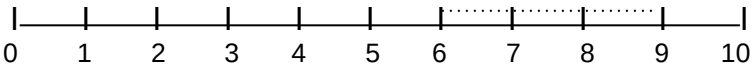
17. There is a standardised clinic schedule (av 9.2)



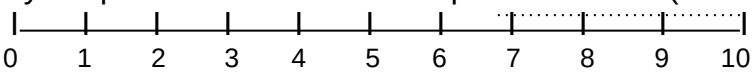
18. The child is referred if there is no change Pirani score for 4 weeks (av 8.9)



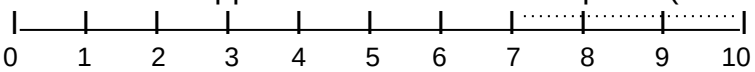
19. The person in charge of the clinic is a champion for clubfoot (av 8.1)



20. The close maternity hospitals refer their clubfoot patients (av 8.5)



21. Each time a patient misses an appointment it is followed up (av 8.6)



Appendix 3. Quality Assessment Form

Domain	Criteria	Rating				Score
		0	1	2	3	
Leadership	There is an identifiable person in charge of the clinic	In the past 8 weeks:				
		There was no identifiable person in charge of the clinics	There was an identifiable person in charge of 5 or fewer clinics	There was an identifiable person in charge of 6-7 clinics	There was an identifiable person in charge of every clinic	
Human resources	There are regularly two or more Ponseti trained health care workers (HCW) available at each clinic	In the past 8 weeks:				
		There were fewer than 2 trained HCWs available at every clinic	There were 2 or more trained HCWs available in 5 or fewer clinics	There were 2 of more trained HCWs available in 6 -7 clinics	There were 2 or more trained HCWs available at every clinic	
Essential medical equipment	There is always plaster of paris available	In the past 8 weeks:				
		Plaster of paris was not available for 3 or more clinics	Plaster of paris was not available for 2 clinics	Plaster of paris was not available for one clinic	There was always plaster of paris available	
	There is always equipment (such as bucket and scissors/blade, tenotomy set, correct size braces) available	In the past 8 weeks:				
		A bucket, scissors/blade, tenotomy set or correct size braces were not available for 3 or more clinics	A bucket, scissors/blade, tenotomy set or correct size braces were not available for 2 clinics	A bucket, scissors/blade, tenotomy set or correct size braces were not available for one clinic	There was always a bucket and scissors/blade, tenotomy set, correct size braces available	
Service delivery	There is a standard treatment protocol	There is no standard protocol for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot or non-idiopathic clubfoot	There is a standard verbal treatment protocol but nothing in writing for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot and non-idiopathic clubfoot	There is a written protocol for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot and non-idiopathic clubfoot but it is not consistently used	There is a standard written protocol for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot and non-idiopathic clubfoot and it is followed consistently	

	There is a functioning referral system for tenotomy	In the 2 most recent tenotomy cases:				
		There was no clinician to whom children requiring a tenotomy could be referred	The children were referred for tenotomy and there is no record of the outcome	The children were referred but the tenotomy was not completed as anticipated	The children were referred for tenotomy and it was completed as anticipated	
	There is a process for surgical referrals	In the 2 most recent cases who required surgery:				
		There was no clinician to whom children requiring a surgical review could be referred	The children were referred and there is no record of the outcome	The children were referred but the surgery was not completed as anticipated	The children were referred for surgery and it was completed as anticipated	
	There is a process to monitor drop out of patients	There is no process to monitor drop out of patients	There is a verbal process to monitor drop out of patients but nothing in writing	There is a written protocol to monitor drop out of patients but it is not consistently followed	There is a written protocol to monitor drop out of patients and it is consistently followed	
	The clinic occurs on a specific day, at minimum weekly	In the past 8 weeks:				
		The clinic did not have an identified day for clubfoot treatment	The clinic has been held on the identified day/s for less than 6 of the weeks	The clinic has been held on the identified day/s for 6-7 of the weeks	The clinic has been held on the identified day/s every week	
Health information system	There is a completed clinic record for each patient visit	In the last 10 clinic records:				
		5 or less of the clinic records were filled in completely	6-7 of the records were filled in completely	8-9 of the clinic records were filled in completely	All of the clinic records were filled in completely	
Total score						

Epilogue (Main findings and limitations)

The consistency of Ponseti method trainers in Africa in rating indicators to assess functionality of Ponseti clubfoot clinics was good. The set of 10 indicators include components of five of the six building blocks of a health system: leadership, human resources, essential medical equipment, health information systems and service delivery.

A questionnaire was developed consisting of the 10 indicators; each scoring from 0 to 3, giving a total from 0 to 30 where 30 is the ideal result.

The most common needs identified in the Zimbabwe clubfoot clinics related to service provision; they were (a) a standard treatment protocol, (b) a process for surgical referrals, and (c) a process to monitor drop out of patients.

Limitations to this study design include lack of guidelines on thresholds to determine consensus, therefore the thresholds for the VAS mean and SD were decided *a-priori* in an effort to determine indicators that were rated with a high mean value and minimal variance. The Delphi panel consisted of expert trainers in the Ponseti technique and are not representative of non-specialised health workers that deliver clubfoot care. Additional data will be required to have a complete understanding of clubfoot clinic functionality, as indicators never capture the richness and complexity of a health system.

Additional Information: Strategies to contain costs for children with clubfoot and their families need to be considered. The sixth health system building block (Financing) is not included in the FACT tool as it was not identified in the facilitated discussion workshop that was used to identify indicators likely to demonstrate a well-functioning Ponseti clubfoot clinic. This is a limitation of the FACT tool, and additional finance data are required to allow a more complete understanding of the functionality of the clubfoot clinic.

Chapter 11. The development of a training course for clubfoot treatment in Africa: learning points for course development

Small group practical sessions with the ACT project

Preamble

Two trained clubfoot therapists are required to deliver the Ponseti method of clubfoot treatment (Chapter 10). There is a lack of trained health workers to provide treatment for clubfoot in Africa and there is no standard training course for these clubfoot therapists.

The Africa Clubfoot Training (ACT) project was developed in response to requests to build local training capacity in the Africa region, and to develop training materials with standard elements for novice clubfoot therapists.

Information on the content of this training course is required to optimise the task-shifting and task-sharing role of clubfoot therapists.

In addition, there is a paucity of data to inform dialogue on the development of medical training in LMIC. Qualitative and quantitative data that are used to identify what elements of the training work, what needs to be improved and how this process may occur, are rarely reported. These mixed methods are required to inform the design of content, and to improve understanding of the factors that influence training outcomes.

This chapter comprises a mixed methods (both qualitative and quantitative) study that describes the development of a training course for clubfoot therapists in Africa. It outlines the inputs that the Africa Clubfoot Training project used to create the training course, the processes by which these were achieved, and it describes the content of the course.

This paper was submitted to BMC Medical Education in September 2017 and is undergoing revision after peer review.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

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Student Signature: [redacted] **Date:** 14/8/18

Supervisor Signature: [redacted] **Date:** 14-08-18

Smythe et al. BMC Medical Education (2018) 18:163
https://doi.org/10.1186/s12909-018-1269-0

BMC Medical Education

RESEARCH ARTICLE Open Access

The development of a training course for clubfoot treatment in Africa: learning points for course development

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Abstract

Background: Clubfoot is a common congenital musculoskeletal disorder that causes mobility impairment. There is a lack of trained mid-level personnel to provide clubfoot treatment in Africa and there is no standard training course. This prospective study describes the collaborative and participatory approach to the development of a training course for the treatment of clubfoot in children in resource constrained settings.

Methods: We used a systems approach to evaluate the development of the training course.
Inputs: The research strategy included a review of context and available training materials, and the collection of data on current training practices. Semi-structured interviews were conducted with seven expert clubfoot trainers. A survey of 32 international and regional trainers was undertaken to inform practical issues. The data were used to develop a framework for training with advice from two technical groups, consisting of regional and international stakeholders and experts.
Process: A consensus approach was undertaken during workshops, meetings and the sharing of documents. The design process for the training materials took twenty-four months and was iterative. The training materials were piloted nine times between September 2015 and February 2017. Processes and materials were reviewed and adapted according to feedback after each pilot.

Results: Fifty-one regional trainers from Africa (18 countries), 21 international experts (11 countries), 113 local providers of clubfoot treatment (Ethiopia, Rwanda and Kenya) and local organising teams were involved in developing the curriculum and pilot testing. The diversity of the two technical advisory groups allowed a wide range of contributions to the collaboration.
Output: The resulting curriculum and content comprised a two day basic training and a two day advanced course. The basic course utilised adult learning techniques for training novice providers in the treatment of idiopathic clubfoot in children under two years old. The advanced course builds on these principles.

Conclusion: Formative research that included mixed methods (both qualitative and quantitative) was important in the development of an appropriate training course. The process documentation from this study provides useful information to assist planning of medical training programmes and may serve as a model for the development of other courses.

Keywords: Clubfoot, Congenital talipes equinovarus, Ponseti, Africa, Training, Clinical skill, Course development

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Background

Clubfoot, or congenital talipes equinovarus, is a common congenital disorder that causes mobility impairment if untreated. It is a structural and functional deformity where the child is born with the foot turned inwards [1]. The Ponseti method [2, 3] is promoted as an effective and low cost treatment of clubfoot [4]. This minimally invasive method includes a correction phase and a maintenance phase, and can be delivered by trained mid-level health care providers in resource constrained settings [5]. The correction phase of treatment involves the simultaneous correction of three components of the clubfoot deformity, with the equinus (downward pointing of the foot) corrected last. The manipulated foot is held in a series of long leg (toe to groin) plaster of paris casts, with the knee at 90 degrees. Manipulation and casting usually occurs weekly. The plaster cast retains the degree of correction and allows the soft tissue time to remodel. Once the cavus, adductus and varus have been corrected, an outpatient procedure to cut the Achilles tendon (heel tendon), known as a tenotomy, is usually needed to correct the rigid downward position of the foot. The final cast remains for 3 weeks to allow the Achilles tendon to re-grow in this lengthened position. To prevent recurrence of the deformity, it is recommended that a foot abduction brace is worn for 23 h/day for 3 months following correction, and then at night until the child is 4 years old [6].

Despite the worldwide spread of the Ponseti method for clubfoot [7], there is a discrepancy between what is known about effective clubfoot management and its availability to children with clubfoot in Africa. A major barrier is trained health care providers. The World Health Organization (WHO) recognises a health workforce crisis in Africa [8] and the need to share resources to narrow the gap between the ideal health workforce and the capacity of training institutions [9].

The Africa Clubfoot Training (ACT) project was developed in response to requests to build local training capacity in the Africa region and to develop training materials with standard elements for novice clubfoot treatment providers. The project aimed to align with local priorities though joint planning and co-ordination, and to establish a long-lasting partnership and community of practice. It proposed to:

i. Develop a simplified training course for novice clubfoot treatment providers with standard elements
ii. Strengthen training and delivery capacity for clubfoot treatment in sub-Saharan Africa through providing national clubfoot trainers with standardised and evaluated training materials
iii. Build capacity for clubfoot training and mentoring through developing an integrated training of trainers course

Historically a narrow technical focus without contextual understanding and limited teamwork [9, 10] have led to a lack of agreement on curricula and relevant frameworks for medical education. The tendency of professionals to act in isolation compounds this difficulty [9]. While frameworks for successful partnership networks and principles of good collaboration are reported [10–14], there is a paucity of data to inform dialogue on the development of medical training [15]. Many training courses include components of knowledge, skill and competency [16], however formative research used to inform the design of content is rarely reported [17]. Formative research is systematic and process oriented. It allows the collection of data (both qualitative and quantitative) and analysis of what elements of the training work, what needs to be improved and how this process may occur [18]. It aims to develop and improve training design from an early stage when opportunities for influence are likely to be greatest, and it allows improved understanding of the factors that influence training [19].

There is little evidence of the effectiveness of training and mentoring health care providers in the management of clubfoot in Africa, or how such interventions can be best designed to be feasible and appropriate. This prospective study describes a systems approach [20] to the design and implementation of a health related training course. We describe the inputs and processes [21] that the ACT project used to create the training course, and how these were achieved. The lessons learned may be useful for development and implementation of other medical training courses.

Methods

Inputs

Partnership of stakeholders

The ACT project is a partnership between the Nuffield Department of Orthopaedics, Rheumatology and Musculoskeletal Sciences (NDORMS) at the University of Oxford, the Global Clubfoot Initiative (GCI), CURE Clubfoot and CURE Ethiopia Children's Hospital, in co-ordination with the Ministries of Health in Ethiopia and Rwanda. These partners formed a core project team and informed project planning and course design through comprehensive knowledge of regional context and needs. The Tropical Health Education Trust (THET) funded the ACT project through the Health Partnership Scheme funded by the UK Department for International Development. The London School of Hygiene & Tropical Medicine provided monitoring and evaluation expertise.

Comprehensive needs analysis

A situational needs analysis and scoping meeting was undertaken in March 2015 with the core project team. The current understanding, context and needs for a training course were mapped extensively.

Review of existing training materials and current practice

Initially, a review of available training materials, teaching methods and processes was undertaken. Two researchers in Public Health then conducted semi-structured interviews with regional and international trainers in the Ponseti method of clubfoot management to collect data on current training practices and challenges. The interviews were transcribed and coded to identify common themes. Following the interviews, an online survey was created and regional and international trainers, purposively selected as experts in the field, were invited to participate. The survey was designed to identify which training practices provided desired clubfoot management behaviours and potential areas for improvement. It aimed to understand practical issues, such as whom to include in the training, current knowledge and skills gaps, and follow up mentoring requirements (Additional file 1). Throughout this review process, published materials and current literature were explored [22] to identify best practice in both clubfoot management and principles of adult learning (for example, providing effective feedback) [23].

Expert consultation

A wide spectrum of experts were involved in the decisions of what should be included in the training and how this should be achieved. The experts were invited to participate in one of two technical advisory groups (TAG). The Africa TAG consisted of trainers in clubfoot management (Ponseti technique) throughout Africa and experts from non-governmental organisations (NGOs) that currently support clubfoot management in Africa; representatives from CBM, CURE, International Committee of the Red Cross (ICRC), MiracleFeet, Mobility Outreach International (MOI) and Ponseti International Association (PIA) were invited to participate. The UK TAG consisted of members of the UK Clubfoot Consensus Group [24] and medical educationalists. The roles of each TAG were outlined at the beginning of the project and included the review of course material for appropriate training delivery, technical and methodological accuracy, and suitability for delivery in a low-income setting.

Training of trainers

Regional trainers were identified to deliver and pilot the 'Basic Provider Course' (BPC) training and supervise new providers. The trainers were chosen based on willingness to participate, knowledge of the topic and their strategic role as key clubfoot trainers in their country. They included orthopaedic surgeons, physiotherapists, physician assistants, prosthetists and orthotists, nurses and orthopaedic technicians. The mean length of time that the trainers had used the Ponseti method was 7.8 years (95%CI 6.5–9.0) and the average number of trainings delivered was 6.4 (95%CI 4.4–8.4). The regional trainers attended a two-day, customised "Train the Trainer" (TTT) course delivered by clubfoot experts and immediately delivered the two-day 'Basic Provider Course' (BPC) themselves, to reinforce newly learned principles and to give feedback on the BPC structure and content.

Equipment

Rubber models and skeleton models were required. A list of material inputs (e.g. plaster of paris, underwrap, tenotomy kits, buckets) was created over the pilot trainings to assist future training courses and is outlined in Additional file 2.

Patients

Children with clubfoot, under the age of 6 months, were assessed and treated by providers under close supervision. All caregivers were read an information sheet about the training and given an opportunity to ask questions. If they agreed to participate, written consent was taken. The caregiver was required to provide written consent and to remain present throughout the assessment as per national requirements. Transport costs were reimbursed and referral services available were mapped pre-emptively to ensure appropriate onward referral for any children that required further intervention.

Process

Plan: Partnership development plan, workshops, meetings, draft documents

All partners that formed the core project team formally committed to a partnership development plan at the beginning of the project. The BPC and APC training goals and design were developed from the extensive needs analysis, project planning, literature search, interview and survey data. They were presented at web meetings with the two TAGs and subsequently commented on over a two-week period. The goals and design were modified accordingly and topics where consensus was not reached were noted. Meetings occurred after each pilot with the core project team and draft documents were shared electronically for further comments and suggestions, after which they were amended. This occurred seven times. The APC was presented and discussed with regional trainers on the final day of two

BPC pilots. Figure 1 outlines the timescale of key activities.

Design: consensus approach

A list of topics that required consensus was generated where evidence in the literature was lacking or unclear. Two additional web meetings were held with the TAGs prior to the delivery of the pilot training to discuss the topics where consensus was required. As the aim for the BPC was to create a course that was simple to understand for novice providers, materials with a standardized approach on the controversial areas in clubfoot treatment were required. Further consensus forums were therefore scheduled in the pilots for discussion with regional trainers.

Development: iterative approach

The BPC content and structure was edited twice before the first pilot in September 2015. The training material was piloted four times. Semi-structured interviews with trainers and trainees were conducted during each pilot training and collated with feedback from verbal debrief

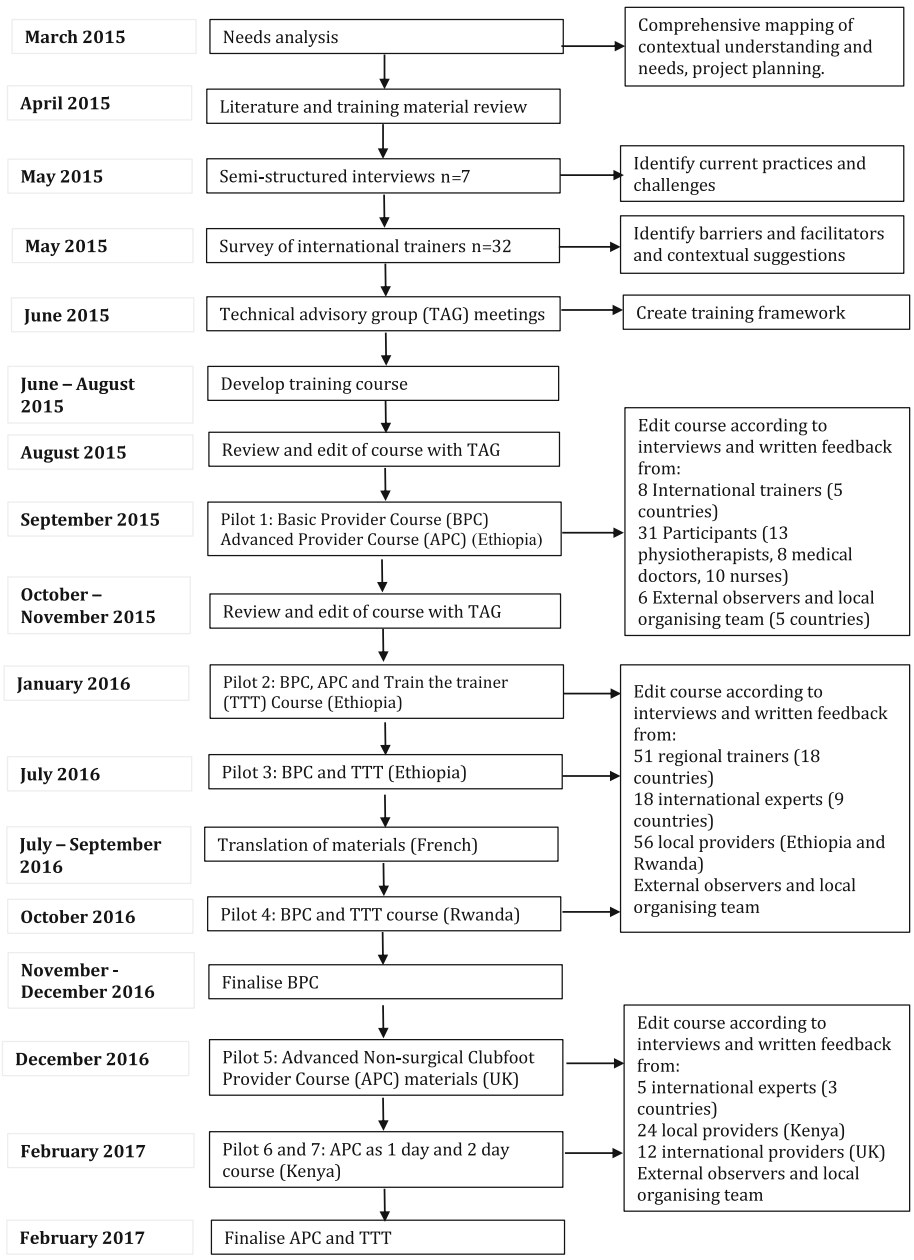

Fig. 1 Timescale of key activities in the development of the training course

sessions and written questionnaires. The training materials were reviewed and adapted according to feedback after each pilot. This process evaluation allowed further development of the training material and substantial changes were made to the timing and content of the training course. The comprehensive needs analysis allowed a good foundation for the format of the course, however at the beginning of the project partnership, the precise content of the training course was unknown. Through the iterative approach, it was identified precisely where novice providers required support after basic training and the APC content was developed into a 2 day course from the recommendations made at the end of the BPC. The process evaluation for the five APC pilots was the same as for the BPC.

Delivery

The two-day BPC was piloted with eight international trainers in September 2015. The content, materials, supervisory and mentorship structure [16] of the BPC was then piloted with 51 regional trainers in Ethiopia (2 courses) and Rwanda (1 course) and included both Anglophone and Francophone trainers. The one-day APC was piloted in Ethiopia (2 courses) and the UK (1 course) and a one-day and two-day version of the APC course was piloted in Kenya.

Monitoring

A theory of change (ToC) was developed to hypothesise the pathways that may contribute to behaviour change in the BPC training (Fig. 2). Regional trainers attending the TTT course completed a knowledge MCQ and confidence matrices immediately pre- and post-course, and a further confidence matrix after applying teaching skills in delivering a BPC.

The ToC was adjusted throughout the twenty-four months as further assumptions and contextual factors were considered. The standard elements to be delivered in training were defined in order to inform a skills checklist, knowledge and confidence pre- and post-training. Quantitative data were collected through pre- and post-training questionnaires to monitor efficacy of training and to identify areas for improvement and increased learning opportunities. Knowledge was assessed before and after the course with a pilot tested single best answer multiple-choice questionnaire (Additional file 3). Long-term knowledge retention was not assessed. Confidence in key skills in clubfoot management was assessed before and after the course with a pilot tested confidence matrix. Extensive written and verbal feedback was obtained from national, regional and international trainers following each pilot to identify areas for improvement.

Ethics, consent and permissions

The London School of Hygiene & Tropical Medicine granted ethical approval for this study ref.:10412/RR/3466. Informed written consent was obtained from all participants.

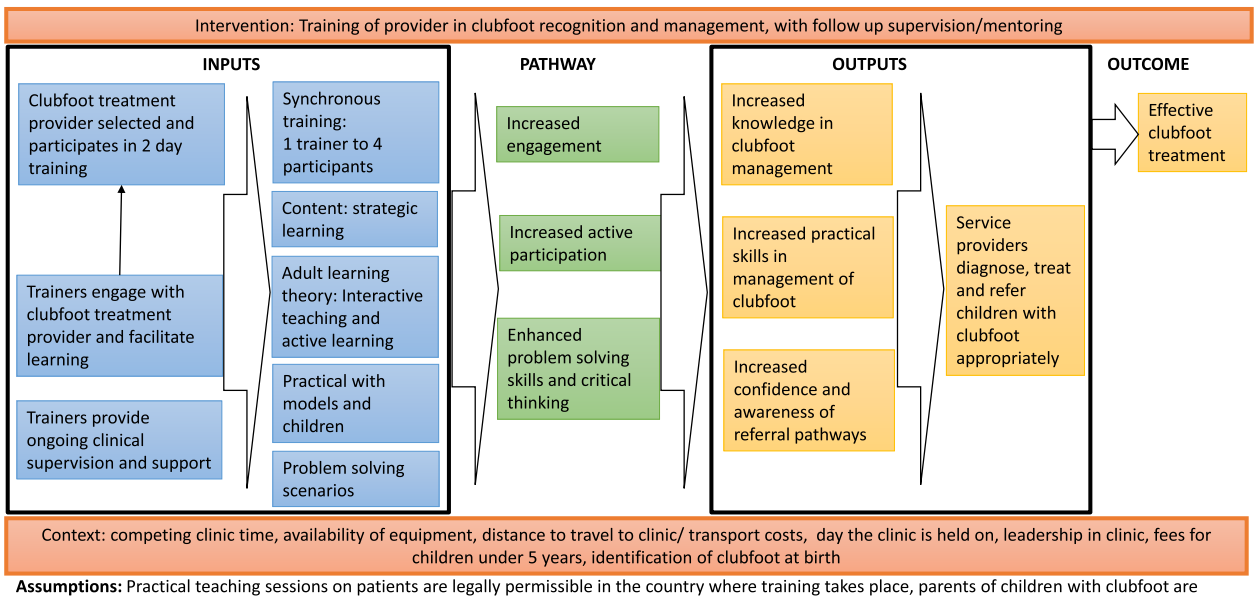

Fig. 2 Theory of change for the development of the BPC training course

Results

Training goals

The design process for the curriculum and content of the training course for clubfoot treatment in Africa took twenty-four months (March 2015 – February 2017) and was iterative. The comprehensive needs analysis proposed a project design that included goals to:

i. Create and pilot a simplified basic course with key messages and increased practical opportunities and videos, with the use of a standardised teaching method to promote critical thinking and problem solving
ii. Create and pilot an advanced course and an integrated training of trainers
iii. Pilot the test materials with trainers from throughout Africa to ensure input to material development and enhance acceptability, feasibility and ownership
iv. Ensure manuals include logistics for administration staff and briefings for the trainers to promote the link between training and follow up supervision and mentoring.
v. Purposively select pilot locations and regional trainers based on extensive knowledge and capacity
vi. Translate the materials into French due to needs of the region
vii. Integrate principles of adult learning.

Participation

Of the 26 international and regional trainers invited to the first interviews, seven (27%) were available to participate. Participants reported the need for training materials with consistent language and messages, and understandable learning outcomes. The importance of practical components in training and follow up mentoring were highlighted, as was the need to facilitate expectations of what practical skills a provider may develop through participation in the training programme.

Thirty-two of 100 trainers (32%) responded to the online survey. Collectively, the 32 trainers had held courses in 54 countries. Twenty of the 32 trainers (63%) had delivered more than 10 courses. Seventy percent had used their own materials for training and 63% had used materials from the Global Clubfoot Initiative (GCI). Eighty-three percent advised on a separate basic and advanced provider course. The need for refresher training and mentoring was a key theme, in addition to requests for targeted training at an appropriate level for novice clubfoot providers. The creation of opportunities to share the previously developed curricula, best practices and lessons learned were common goals.

Output, basic provider course

The resulting BPC has a focus on adult learning techniques for the training of novice providers in the treatment of idiopathic clubfoot in children less than 2 years of age. The final curriculum and content of the BPC focuses on a problem solving approach with group work and practical exercises to promote optimal learning and facilitate active engagement. It is designed to be delivered face to face over 2 days, or as fourteen stand-alone modules, with a sequence of learning tasks that progress from simple to more complex. If delivered as stand-alone modules, this would be in response to a specific training need or contextual issues that were identified by a clinic or individual. It contains clearly defined content and learning objectives for the development of required skills for the novice provider (Additional file 4 outlines the training courses). The structure of the BPC allows for the teaching of content through small group and other interactive activities and communication is promoted through built-in open questions to stimulate dialogue. The learning outcomes for novice local providers were based on the diversity of cadre to be trained and the need for a training course with standard elements.

Learning outcomes BPC

The BPC was therefore designed with three main learning outcomes; providers of the Ponseti method of clubfoot treatment will be able to:

i. Explain the clubfoot deformity and the method of correction using the Ponseti technique;
ii. Demonstrate practical skills in treating a child under 2 years presenting with idiopathic, previously untreated clubfoot; and
iii. Identify more complex cases, when treatment is not progressing as would be expected and when to seek guidance with these cases.

Output, advanced provider course

The APC was designed to build on the knowledge, competence and opportunities for mentoring that were developed through the BPC. The main objectives include:

i. To refresh understanding and skills in basic Ponseti treatment, and to add advanced knowledge to these
ii. To develop understanding and skills in non-surgical management of challenging cases such as atypical, recurrent, neglected, and secondary clubfoot
iii. To facilitate exchange of knowledge through case discussion of challenging cases
iv. To establish a common approach to measuring and improving quality of care in clinics, to encourage reflection on what is and what is not working well

in own practice / clinic setting, and to identify priority actions

v. To promote consideration of how parents can be supported to promote treatment adherence during clubfoot treatment.

Application of formative research

Consensus topics included, but were not limited to, the definition of recurrence of the clubfoot deformity, the definition of complex and atypical clubfoot, Pirani score and positioning, and the brace review protocol. Consensus on the format, content and structure was gained after the fourth pilot course. The need for a third training course that includes surgical management was identified.

As planning progressed, new questions were identified. Feedback included comments on individual slides, clarity and context of images, organisation and order of topics, consistency of language, context specific details, level of difficulty, and the identification of further practical opportunities.

The diversity of advisory groups and trainers allowed a wide range of contributions to the collaboration, but also complexity in the management of different interests. Table 1 outlines the results of the demographic analysis of the pilot courses.

The following examples demonstrate the type of information collected in the formative research and how this information was used to inform decisions about the approach to the training message.

First, the need to develop a training of trainers was developed as part of the grant application. The need to model best practice when training was further raised in the interviews and TAG meetings. In response, the TTT was developed to build the foundation for best practice training techniques. On the basis of the content and the observed need to be closely supervised and assisted in practical sessions, a 1:4 trainer to participant model was adopted when delivering the BPC. Closer supervision of trainees was required given the increased emphasis on interactive learning and the inclusion of detailed practical sessions.

Second, the proposed 1 day 'Advanced Provider Course' that was identified in the needs analysis was modified to the 2 day 'Advanced Non-Surgical Clubfoot Treatment Course' over the pilots. Challenges with scope of content were raised in the September 2015, January 2016 and July 2016 pilots. Small group discussions and responses from questionnaires advised that trainees with a good level of skill in clubfoot management (Ponseti technique), who are actively receiving mentoring and supervision, require an environment in which to refresh understanding and skills in basic clubfoot management and to delve deeper into how and why the technique works, in addition to sharing tips and advice through discussion of complicated cases. Consequently, it was agreed that changes to the APC were needed. A low-cost pilot in the UK was undertaken in December 2016, ahead of the pilot in Kenya in January 2017, to maximise the opportunity for success. The one-day advanced course was therefore developed with the intentional focus on non-surgical intervention. It was extended to 2 days to allow adequate time for discussion, reflection, practice and development of practical skills, based on pilot course feedback by trainers and participants. It was noted that a further course on surgical management is required.

Knowledge and skills gained

Fifty-one regional trainers and 113 national clubfoot treatment providers (Ethiopia, Rwanda and Kenya) were trained through the course development. Results of a knowledge MCQ and confidence matrix completed pre- and post-training by local providers are demonstrated in Table 2.

Correct multiple choice questionnaire (MCQ) answers increased from 59 to 80%, (BPC participants) and 59 to 73%, (APC participants) after training. Self-reported confidence increased from 57 to 89% (BPC participants) and 85 to 95% (APC participants) (Table 2).

Discussion

This study outlines the formative research that informed the design of a training course for clubfoot providers in resource constrained settings. A two-day training course for novice clubfoot providers (Ponseti method) and two-day training course for advanced clubfoot providers were developed. Decisions were made using a data driven approach with a comprehensive contextual understanding, and involved key stakeholders. The ACT project delivered consensus on the content and quality of training material through partnership with technical advisory groups, regional trainers and local participants. The local organising teams, who were not involved in the training design, provided insight on practical issues and it is likely that their involvement will increase sustainability. In-depth needs analysis, interviews, surveys, opinion polls, group discussions with experts, consensus meetings, piloting the materials and observation allowed the development of a training course that aligned with regional priorities.

Strengths

From the beginning of the ACT project there was agreement on the focus of the training course. The partnership development plan ensured commitment to health system partnerships and partnerships in practice. The flexibility in design of the course allowed the decision-making process to be iterative and the training

Table 1 Demographics of the pilot courses

Pilot number	1	2	3	4	5	6	7	Total[a]
City, Country	Addis Ababa, Ethiopia	Addis Ababa, Ethiopia	Addis Ababa, Ethiopia	Kigali, Rwanda	London, UK (1 day)	Nairobi, Kenya (1 day)	Nairobi, Kenya (2 day)	
Dates of pilot trainings	23–24 September 2015	25–29 January 2016	25–29 July 2016	24–28 October 2016	2 December 2017	26 January 2017	24–25 January 2017	21 days
Type of pilot training	BPC and APC	BPC, APC and TTT	BPC and TTT	BPC and TTT	APC	APC	APC	
Number of International experts delivering training	8	9	9	9	5	5	5	21
Countries represented by International experts	Ethiopia, Norway, Netherlands, UK, Zimbabwe	Australia, Ethiopia, UK, Zimbabwe	Australia, Ethiopia, Kenya, Tanzania, UK, Zimbabwe	Australia, Canada, Cameroon, DRC, Rwanda, Switzerland, UK, Zimbabwe	UK	UK, Kenya, Australia	UK, Kenya, Australia	13
Number of organisers for training course	4	3	3	3	2	3	3	12
Countries represented (organisers)	Australia, Ethiopia, UK	Ethiopia, UK	Ethiopia, UK	Rwanda, UK	UK	Kenya, Zambia	Kenya, Zambia	4
Number of regional trainers	0	18	17	16	0	1	1	51
Number of countries represented (trainers)	Trainers were ACT faculty	10	10	7	1	3	3	18
Names of countries represented (regional trainers)		Ethiopia, Ghana, Kenya, Liberia, Malawi, Mozambique, Rwanda, Tanzania, Zambia, Zimbabwe	Ethiopia, Cameroon, DRC, Ghana, Kenya, Malawi, Sierra Leone, South Africa, Zambia, Zimbabwe	Burundi, Niger, DRC, Senegal, Cameroon, Rwanda, Togo				18
Cadres of trainers: Surgeon	5 M	11 M	9 M	5 M	0	0	0	30
Physiotherapist	2F, 1 M	1F, 5 M	1F, 2 M	8 M	0	0	0	20
Medical doctor	0	0	0	1F, 1 M	0	0	0	2

Table 1 Demographics of the pilot courses *(Continued)*

Pilot number	1	2	3	4	5	6	7	Total[a]
Clinical officer	0	1 M	3 M	0	0	0	0	4
Nurse/other	0	1F	2 M	1 M	0	0	0	4
Number of local providers trained	20	17	18	21	12	12	13	113
Cadres of providers trained: Surgeon	3 M	3 M	0	1 M	2 M, 1F	0	0	10
Physiotherapist	1F, 7 M	1F, 3 M	1F, 9 M	3F, 9 M	2 M, 6F	1 M, 2F	2 M, 1F	48
Nurse	7F	4F, 5 M	2F, 1 M	2 M	1F	1 M	0	23
Doctor	2 M	1 M	1F, 4 M	4 M	0	0	1 M	13
Other	0	0	0	0	0	6 M, 2F	5 M, 4F	17

M male, *F* female

[a]Several trainers were involved with multiple trainings. The total is the number of individuals involved and does not count a person more than once

Table 2 Knowledge and confidence of participants before and after training

	Pre-course mean (95%CI)	Post-course mean (95%CI)
BPC participants		
MCQ	59% (53–65)	80% (74–85)
Confidence	57% (50–64)	89% (86–92)
APC participants		
MCQ	59% (49–68)	73% (63–82)
Confidence	85% (79–92)	95% (92–98)

materials to be modified as new information emerged. For example, the process allowed the development of two courses and identification that a further course focusing on surgical management was required, and the APC was expanded from a one-day to a two-day course in order to meet training needs as identified by contextual understanding, the training experts and participants. In addition, pooled resources (human, institutional and financial) contributed to the success of the training course design and the constant dialogue between colleagues, experts and stakeholders with different backgrounds facilitated exposure to different views and approaches [25, 26]. Stakeholder meetings, webinars and pilot trainings were used as arenas for decision-making on consensus issues and the various mediums were useful to build regional ownership.

Limitations

A limitation of the training course is the absence of a pass/fail competency [27]. Instead, a skills checklist (Additional file 5) was developed to allow follow-up mentoring to identify areas of strength and weakness in novice providers. The skills checklist allows a skill gap analysis and follow up in supervision clinics [16]. Consensus was that the course would provide the initial foundation for the delivery of clubfoot treatment and further mentorship is required; as such a competency pass/fail was deliberately avoided. Seventy-three percent (19/26) of the experts invited for an interview and 68 % (68/100) of international trainers invited to complete the survey did not participate. The survey participants were also self-selected and selection bias in the development of the training curriculum cannot be ruled out. With regards translation, the forward translation was undertaken by a health professional, however the expert panel participating in the backward translation consisted of the regional francophone trainers. This research is limited to the description of how a training course was designed and the collaborative process undertaken, and intentionally does not evaluate the long-term outcomes or impact of the training course. The authors were closely involved in the implementation of the ACT project and in the development of the manuscript, which may lead to researcher bias in the results of this study.

Lessons learnt

1. Within the ACT collaboration, each partner had unique characteristics without which the partnership would not have be possible (e.g. human resources, technical expertise, funding ability, awareness of context); common shared values and interests among the partners were essential.
2. The rigor and focus of the Health Partnership Scheme grant brought partners together with a sense of purpose and common goals.
3. Minutes that outlined the understanding of proceedings and action points to be undertaken after each meeting allowed an organised approach to communication and accountability.
4. The management of expectations and activities was aided by clearly defined objectives for the training that were regularly assessed and modified when necessary.
5. The constant evaluation of all aspects of the training materials, and their potential limitations, was promoted to inform the design and development of the training course.
6. The use of a variety of methods to deliver the training messages (such as demonstration, pair work, practical opportunities) was important to help select the anticipated behaviour changes.

The work we describe took 2 years to develop and will require further evaluation of regional implementation and impact.

Challenges

The creation of partnerships between non-governmental organisations (NGOs) that are often in competition for funding, combined with avoiding a culture of individualism, took leadership and patience on all sides. Establishing consensus among multiple stakeholders was one of the most difficult and time-intensive steps, and required negotiation skills as well as a shared commitment to the ultimate goal. Meetings with international and regional stakeholders were limited by time as participation was on a volunteer basis. Practically, different time-zones and inconsistent internet access required web meetings to have thorough advanced planning. Barriers to volunteering with the ACT project faculty included difficulty in National Health Service staff taking time away from the UK, as observed in other projects [28].

Main findings as related to previous literature

The importance of formative research for the design of successful interventions extends from the provision of neonatal care in rural Ghana [17], to hygiene promotion programmes [29] and programmes to reduce the treatment gap for mental health disorders in low resource settings [30]. Principles of good collaboration have been proposed to include (i) development across a diverse group of stakeholders, (ii) establishment of a community of practice, (iii) strengthened links between the project, communities, and ministries of health, and (iv) enhanced mutual respect for different cultures and contexts [14]. However, while interventions regularly include various forms of training, there are few published examples of the design process undertaken to create the training.

Implications

Major gaps in evidence are highlighted through this research. There is little information available on the formation of training courses for health care workers in resource constrained settings. The accurate identification of processes that assist and facilitate learning and application of new skills within appropriate contexts is required. Further evaluation of the interaction between knowledge and competence is needed. In addition, evaluation of how different components of a training programme are understood by different cadres is required, as is the carry-over of skills after the training course.

Recommendations for implementation of the training course

1. Training with practice-based learning requires direct supervision and multiple opportunities for the clubfoot providers to demonstrate competency in practice over time. In this example, the training is designed to occur in a 1 to 4 (trainer to provider) model to allow providers adequate participation and supervised practice in assessment and treatment.
2. Supportive supervision and mentoring 'on the job' is needed to meaningfully build on initial skills gained in training.
3. The sharing of knowledge of existing curricula, programmes and systems will increase opportunities globally to build regional capacity and increase access to interdisciplinary services.
4. Innovative ways to address the potentially limited access to clubfoot models or equipment suggested in this training warrants further research (e.g. through the use of mobile health tools).
5. National investments in provider training and in supporting the health system are required for scaling up and sustaining clubfoot treatment.

Conclusion

Formative research with mixed methods (both quantitative and qualitative) was essential for the development of the training courses. Consensus meetings were central to the harmonisation of aims and goals of the ACT project. A broad spectrum of multidisciplinary stakeholders, beyond those with clinical expertise, shaped the success of the training project through a shared vision and mutual accountability. Knowledge increased in both novice and advanced participants after training. Self-reported confidence increased on all measures tested after participating in the training. The process data from this study provide useful information to assist planning of medical training programmes and may serve as a model for the development of other courses.

Additional files

Additional file 1: Survey of current training practices. Includes questions and topic guides for survey that aimed to understand practical issues, current knowledge and skills gaps, and follow up mentoring requirements. (DOCX 21 kb)

Additional file 2: Considerations for equipment and location. A list of material inputs that was created over the pilot trainings to assist the planning and organisation of future training courses. (DOCX 76 kb)

Additional file 3: Example of the basic provider course MCQ. The pilot tested single best answer multiple-choice questionnaire. (DOCX 25 kb)

Additional file 4: Finalised training course components. Outlines the components of the training courses. (DOCX 15 kb)

Additional file 5: Skills checklist. The checklist developed to allow follow-up mentoring to identify areas of strength and weakness in novice providers. (DOCX 17 kb)

Additional file 6: Acknowledgements. Acknowledgements of the multi-disciplinary team involved in the ACT project. (DOCX 15 kb)

Abbreviations

ACT: Africa Clubfoot Training; APC: Advanced provider course; BPC: Basic provider course; GCI: Global clubfoot initiative; MCQ: Multiple choice questionnaire; NDORMS: Nuffield Department of Orthopaedics, Rheumatology and Musculoskeletal Sciences; NGO: Non-Government Organisation; TAG: Technical advisory group; THET: Tropical Health Education Trust; ToC: Theory of change; TTT: Train the trainer; WHO: World Health Organisation

Acknowledgements

The authors are grateful to the co-ordinators and organisers in Ethiopia and Rwanda who facilitated training logistics. A further thanks to all of those who participated in the course, whose motivation and skills shaped the success of the project. We thank the Ethiopian Ministry of Health and Rwandan Ministry of Health and Human Resources for Health for their support of trainee attendance. We thank the ACT project Core Working Group, Africa Technical Advisory group, UK Technical Advisory group and the UK Clubfoot Consensus Group, without whose commitment and generosity, none of this project would have been possible. Lastly, we acknowledge and thank the multidisciplinary ACT team involved in assembling, writing, piloting, reviewing and rewriting the material included in Additional file 6.

Funding
The ACT project was supported by the Tropical Health Education Trust and funded through the Health Partnership Scheme, which is funded by the UK Department for International Development (DFID) for the benefit of the UK and partner country health sectors. The Beit Trust are acknowledged for scholarship funding provided to TS.

Availability of data and materials
Supporting data can be accessed in Additional Files. Further data used and analysed during the current study are available from the corresponding author on reasonable request.

Authors' contributions
CL, GL, LH, RO, TS conceived the study; CL, GL, LH and TS designed the study protocol; BA, GL, LH, RO and TS developed the outcome measures; GL, RO and TS carried out the analysis and interpretation of these data. TS and CL drafted the manuscript; BA, GL, LH, and RO critically revised the manuscript for intellectual content. All authors read and approved the final manuscript. TS and CL are guarantors of the paper.

Ethics approval and consent to participate
This research was performed in accordance with the Declaration of Helsinki and The London School of Hygiene & Tropical Medicine granted ethical approval for this study ref.:10412 /RR/3466. Informed written consent was obtained from all participants.

Consent for publication
Not Applicable.

Competing interests
The authors declare that they have no competing interests.

Publisher's Note
Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

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Received: 5 September 2017 Accepted: 26 June 2018
Published online: 13 July 2018

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Additional File 1. Survey on current training practices

1. Please tell us about your experiences of conducting Ponseti training...
How many Ponseti training courses have you been a trainer on in the past?
What level of Ponseti courses have you taught? (beginner/advanced courses, or both)
Which countries were the Ponseti courses held in?
2. When delivering Ponseti courses, what training materials have you used?
GCI training materials
Your own training materials
Other training materials (Please specify below)
3. If you have used the GCI training materials please comment on how they could be improved.
4. In your experiences of training, what is the best format to deliver a Ponseti training course:
1-part course: Teach all theoretical and practical aspects at once
2-part course: Start with a preliminary 'beginners course' followed by an 'advanced' or 'refresher' course some time later.
Comments
5. If you were to deliver a 2-part course starting with a preliminary 'beginners course' followed by an 'advanced' or 'refresher' course what modules should be included in each course:
Please suggest any additional modules that should be included
6. What 'red flags' or potential complications of treatment would you make course participants aware of during a beginner's course? (tick all that apply)
Atypical or complex appearance and features
Cast slips
Casting sores
Under-correction of the foot
Over-correction of the foot
Casting errors such as pronating or pushing the foot into dorsiflexion
Relapse of foot deformity
Patient dropping out of treatment
Others (please list below in comments box)
Other (please specify)
7. What hands-on practice elements should be included in a Ponseti course? (tick all that apply)
Practice manipulation on clubfoot skeleton models
Manipulation and casting using rubber foot models
Manipulation and casting on patients
Brace fitting on patients
Pirani scoring on patients
Any other practical exercises you would recommend? (please list in the comments box below)
Other (please specify)
8. Ponseti course practicals...
For practical exercises, what is the ideal number of trainees per trainer?
How many children should each person practice casting on, ideally?
9. Is a 2-3 day Ponseti course with theoretical and practical elements sufficient for participants to learn how to apply the Ponseti technique effectively?
Yes/No
If not, what additional elements would you suggest?
10. Do you have any other comments or suggestions on the best ways to deliver training in the Ponseti technique?

Additional File 2. Considerations for equipment and location

1. Choice of course location and venue : consider – regional versus central, cost, space for both lectures and practicals, noise, ease of catering, accommodation nearby, experts
2. Create an agenda that is realistic, allowing for set-up time each day, breaks, travel times, practical sessions, debriefing
3. Organize equipment – projector, laptop, power blocks, flipcharts, buckets, plinths, scissors, plaster cutters
4. Organize consumables – plaster, padding, braces, gloves, tenotomy supplies
5. Organize resources – electronic copy of slides, manuals, handouts, blank patient records, certificates of attendance
6. Set up the day before the course – room set-up, check equipment is working, organize handouts / manuals / stationery, ensure course materials are on laptop, consider layout / equipment for different sessions
7. Consider publicity / media for the course
8. Organize accommodation, catering, and transport
9. Arrange a faculty / team meeting before the course – allocate topics / sessions / roles:
    a. Course leader
    b. Timekeeping and liaise with catering team
    c. Allocated instructor for each session
    d. Preparation for practical sessions including group sizes and room layout (before course and on the day)
    e. Technical support (computers)
    f. Invitations, registration, and paperwork
    g. Giving each other feedback
    h. Coordinating patients and families
    i. Set-up and set-down

Additional File 3: Example of the Basic Provider Course MCQ

Basic Ponseti Provider Course – Multiple Choice Questionnaire (MCQ)

There are 12 questions, with 1 correct answer for each question. There is no negative marking. Please answer every question, and guess the correct answer if you are not sure.

1. Which of the following is not a clinical component of clubfoot?

   A. Equinus
   B. Abductus
   C. Varus
   D. Cavus

2. Which of the following is not a goal of the Ponseti Method?

   A. A functional foot
   B. A plantargrade foot
   C. To use modified shoes
   D. A painfree foot

3. According to the Pirani score, when should a tenotomy be performed?

   A. When the midfoot score is more than 1
   B. When the midfoot score is 0
   C. When the hindfoot score is 0
   D. When the lateral head of talus score is 1

4. Which of the following is not assessed in the Pirani score?

   A. Posterior Crease
   B. Empty Heel
   C. Degrees of Abduction
   D. Medial Crease

5. Which combination of elements are required by the Ponseti method to successfully manage a clubfoot?

   A. Manipulation and foot abduction brace
   B. Manipulation, casting, Achilles tendon tenotomy and foot abduction brace
   C. Casting and Achilles tendon tenotomy
   D. Manipulation, casting and Achilles tendon tenotomy

6. What is the Pirani score NOT helpful for?

   A. Using a common language to discuss patients with other clubfoot providers
   B. Informing your next treatment
   C. Deciding when to discharge your patient
   D. Monitoring correction of deformity

7. Which is NOT required for successful Ponseti casting?

   A. Discussing the care of the cast with parents
   B. An aim for the cast
   C. Two people to cast
   D. Manipulation of the foot after casting

8. Why is moulding of the cast important?

A. To make the cast look nice for the parents
B. To keep the manipulated position of the foot
C. To give you something to do as the cast dries
D. To strengthen the cast so that the child can walk on it

9. What position does the first Ponseti cast maintain?

A. Dorsiflexion of the ankle
B. Abduction
C. Pronation
D. Supination

10. What deformity of clubfoot does the first Ponseti cast correct?

A. Adductus
B. Cavus
C. Equinus
D. Varus

11. The 'lateral head' of which bone is the fulcrum for manipulation with the Ponseti method?

A. Calcaneum
B. Lateral Malleolus
C. Navicular
D. Talus

12. What is INCORRECT in your assessment of brace quality?
The brace should have:

A. Shoes attached to bar at 50° abduction
B. Dorsiflexion of 10°-15°
C. A well rounded heel cup
D. An inspection hole on the medial side

Additional File 4. Finalised training course components

Course	Components
Basic Provider Course	Introduction to clubfoot and Ponseti management Anatomy and definitions Pirani score Clubfoot Deformity and Ponseti method of manipulation Practical session with rubber and skeletal models Applying Ponseti casts Demonstration of Pirani score and casting on 2 children Practical session with casting on rubber models Tenotomy Bracing and relapse Practical session on assessment Manipulation and casting on patients Practical session on brace fitting and tenotomy When to stop and re-think treatment
Advanced Non-surgical Clubfoot Treatment Course	Review of clubfoot management (Ponseti Method) Why the method works (alternative hand holds and kinematic coupling) Practical exercises with skeletal models Common errors Atypical cases including video and case study Management of recurrence Treating older children Syndromic cases Practical exercises with casting on rubber models Group assessment of patients and formulation of treatment plant Demonstration of treatment on patients Clinic and treatment quality Parent education and adherence

Additional File 5. Skills checklist

Training skills check	Skills demonstrated under supervision	Observed		Comments
		Yes	No	
Discuss Ponseti treatment with parents	Uses demonstration or descriptions to communicate to parents			
	Discusses the plan for treatment			
	Asks if the parents have questions			
Assessment Pirani score	Completes entire Pirani score form including date and initials			
	Interprets and analyses findings to establish treatment plan			
Manipulation of child's foot	Good position of child and parent			
	Identifies lateral head of talus, correct handhold			
	Manipulates foot into accurate position to correct deformity			
	Uses gentle handling, 'soft hands'			
Casting of child's foot	Cast over toes			
	Lower half of cast applied first			
	Cast up to groin and gluteal fold			
	Moulds cast until dry			
	Knee at 90 degrees			
	Toes visible in casts			
	Regular talking with moulder			
Application of brace	Correct measurement of foot and selection of brace			
	Looks at heel cup to ensure heel is in brace correctly			
	Accurately conveys information regarding relapse			
	Makes appointment for review of brace			

Additional File 6: Acknowledgements

The multi-disciplinary ACT team involved in assembling, writing, piloting, reviewing and rewriting the material includes:

Endashaw Abera, Kinfe Araya, William Guy Atherton, Birhanu Ayana, Ryan Bathurst, Rachel Buckingham, Alexis Buunaaim, John Cashman, Christopher Carter, Augustine B. Chiewolo, Sandram Chimangeni, Naomi Davis, Marieke Dreise, , Roderick Duncan, Mesfin Etsub, Jennifer Everhart, Solomon Fasika Demissie, Gregory Firth, Rick Gardner, Prosper Guo-Moh, Ben Gwilliam, Lin Habimana, Soeur Odette Habimana, Jean Claude Habyarimana, Linda Hansen, Moussa Moise Henri Martin, Alison Hulme, Nathaniel Sallu Kargbo, Stephen Kariuki, Christian Katembo Kamavu, Kakule Katenge Joseph, Pascal Kayishema, Simplice Kighoma Vuhaka, Charles V. Alain Kinkpé, Peter Klungsøyr, Joseph Korpisah, Koffi Kouwekou, Giorgio Lastroni, Chris Lavy, Grace Le, Benjamin Lwayivweka Ngahangondi, Chouchou Safi Matsoro, Deborah McMillan, Samuel Maina, Paul Mang'oli, Stephen Mannion, Osman Ibrahim Mohamud, Karen Moss, Ilho Moyo, Debra Mudariki, Ibrahim Issaka Niandou, Isidor Ngayomela, Marie-Caroline Nogaro, Tim Nunn, Henry Ndasi, Jean-François Negrini, André Georges Nguene Nyemb, Emmanuel Nsengiyumva, Isaac Otieno, Rosalind Owen, Safalao Phalira, Norgrove Penny, Rebecca Radcliffe, Scott Reichenbach, Sampson Sarpong-Peprah, Prem Saggurthi, Peter Matthias Schmauch, Jana Shih, Kamwanda Sililo, Tracey Smythe, Michiel Steenbeek, Unisa A Tarawallie, Tim Theologis, Joseph Theuri, Tewodros Tilahun, Tchaa Hodabalo Towoezim, Michael Uglow, Esperance Uwizeye, Bernard Uzabakiriho, Andrew Wainwright, Situmbeko Wambulawae, Denise Watson and Kagnew Wubishet.

In developing these materials we acknowledge the foundational work of many individuals over the years that the Ponseti technique has been practiced in Africa. We are very grateful for material shared by many in the group above, and we also thank the following for a combination of expertise, advice, inspiration and photographs: CBM, Winfried Danke, Fred Dietz, Matthew Dobbs, Marieke Dreise, Bryce Flurie, Vikas Gupta, John Herzenberg, Iris Lohan, A F Lourenco, Colin MacFarlane, Jennifer McCahill, José Morcuende, Vince Mosca, Monica Noguiera, Norgrove Penny, Shafique Pirani, Ignacio Ponseti, David Scher, Marc Sinclair, David Spiegel, Lynne Staheli, Michiel Steenbeek, UK Clubfoot Consensus Group, Miraclefeet and Walk for Life Bangladesh

Epilogue (Main findings and limitations)

Fifty-one regional trainers and 113 national clubfoot therapists (Ethiopia, Rwanda and Kenya) were trained through the course development.

The final curriculum and content of the two-day basic course (BPC) focuses on a problem solving approach with group work and practical exercises to promote the application of learned techniques.

The advanced course was designed to build on the knowledge, competence and opportunities for mentoring that were developed through the BPC.

Limitations to the study include survey participants that were self-selected and therefore selection bias cannot be ruled out. The authors were closely involved in the implementation of the ACT project and in the development of the manuscript, which may lead to observer bias in the results of this study.

A further limitation of the evaluation of this training is the absence of a pass/fail competency. Instead, a skills checklist was developed to allow follow-up mentoring of novice providers.

Chapter 12. The feasibility of a training course for clubfoot treatment in Africa: a mixed methods study

Practising casting with rubber models in Addis Ababa, Ethiopia

Preamble

Process data from the ACT project was important to understand the delivery of an appropriate and suitable training course for clubfoot therapists. This training programme includes standard elements and continued mentorship, and has the potential to contribute to improving treatment of clubfoot in children. Information on the feasibility of the training programme in Africa and evaluation of short-term outcomes is required to assess implications for training effectiveness and scale up.

We undertook a prospective, mixed methods evaluation of the ACT project that was based on a model designed to assess the feasibility of public health interventions (Bowen et al 2010). The model includes eight facets of feasibility: acceptability, demand, implementation, practicality, adaption, integration, expansion and limited efficacy.

This chapter includes a feasibility study of the ACT project that builds on the learning points for course development (Chapter 11). It presents the short-term results and outcomes of a training programme for clubfoot therapists in Africa and makes recommendations for implementation.

This research paper was submitted to the journal PLoS ONE in March 2018 and is undergoing peer review.

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RESEARCH PAPER COVER SHEET

SECTION A – Student Details

Student	Tracey Heather Smythe
Principal Supervisor	Professor Allen Foster
Thesis Title	Evidence to improve clubfoot services in Africa with Zimbabwe as a case study

SECTION B – Paper already published

Where was the work published?			
When was the work published?			
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SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	PLOS ONE
Please list the paper's authors in the intended authorship order:	Smythe T, Owen R, Le G, Uwizeye E, Hansen L, Lavy C
Stage of publication	Submitted

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I co-designed the data collection tools, extracted the quantitative data, completed quantitative data analysis, drafted the manuscript, prepared the subsequent revisions with consideration of comments from co-authors

Student Signature: _____ **Date:** 9/5/18

Supervisor Signature: _____ **Date:** 9/5/18

The feasibility of a training course for clubfoot treatment in Africa: a mixed methods study

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Target Journal: PLoS ONE

Figures: 2

Abstract 274 words, Text 3,098 words

Funders: Scholarship awarded to TS by The Beit Trust and ZANE.

Abstract

Background

There is no available training programme with standard elements for health workers treating clubfoot in Africa. Standardised training with continued mentorship has the potential to improve management of clubfoot. We aimed to evaluate the feasibility of such a training programme among clubfoot providers in Africa, and assess implications for training effectiveness and scale up.

Method

We used participatory research with trainers from 18 countries in Africa over two years to devise, pilot and refine a 2-day basic and a 2-day advanced clubfoot treatment course. (The Africa Clubfoot Training or 'ACT' Course.) The pilots involved training 113 participants. Mixed methods (both qualitative and quantitative) were used for evaluation. We describe and synthesise the results using the eight elements proposed by Bowen et al (2010) to assess feasibility. All participants completed feedback questionnaires, and interviews were conducted with a subset of participants. We undertook a narrative description of themes raised in the participant questionnaires and interviews. Descriptive statistics were used to compare pre- and post-course scores for confidence and knowledge.

Results

113 participants completed pre and post-course measures (response rate 100%). Mean participant confidence increased from 63% (95%CI: 59 – 69%) to 88% (95%CI: 86 – 90%) post course. Mean participant knowledge increased from 55% (95%CI: 51 – 60%) to 78% (95%CI: 76 – 81%) post course. No difference was found in mean for either subscale of cadre or sex. The qualitative analysis generated themes under four domains: 'practical learning in groups', 'interactive learning', 'relationship with the trainer' and 'ongoing supervision and mentorship'

Conclusion

The Africa Clubfoot Training package to teach health care workers to manage clubfoot is likely to be feasible in Africa. Future work should evaluate its impact on short and long term treatment outcomes and a process evaluation of implementation is required.

Introduction

Clubfoot is a common congenital disorder that causes mobility impairment if untreated. The Ponseti method for clubfoot management is recommended for the correction of the fixed foot deformity [1] and can be effectively delivered by mid-level personnel [2]. The treatment consists of sequential correction of the deformity through manipulation and casting, with a tenotomy in most cases, followed by the use of a foot abduction brace to maintain correction [3]. To date, no structured training programme with standard elements exists in Africa.

General strategies to improve clinical practice include didactic teaching, printed educational materials, audit and feedback, interactive workshops, use of local opinion leaders and computerised decision support systems. The effects of these interventions vary from minor to moderately large [4]. There is limited exploration of the development of training for clubfoot providers in the literature, with existing research largely restricted to outcomes of treatment. Evidence suggests that multifaceted training strategies have a higher efficacy, particularly when tailored to address specific barriers and settings [5] and the development of the Africa Clubfoot Training (ACT) project was therefore underpinned by adult learning theory, which is essentially experiential [6, 7].

Our preliminary research suggested that the training developed for clubfoot treatment providers should include a more basic and repetitive introductory element, understandable learning outcomes, practical skill development and elements to improve problem solving [8]. Pre-existing training materials were deemed by users to be too complex for mid-level learners and to contain an excess of information. The materials were thought to be confusing in the way that complex clinical theories (such as kinematic coupling) were presented to participants with a lower level of anatomical knowledge, and to provide insufficient links between assessment and treatment. The challenges of teaching mixed cadres of clinicians were noted, as well as the need for a clearly structured learning experience. The resulting ACT course therefore included two-day basic and advanced training courses, which consist of large group presentations, small group discussions, practical exercises and problem solving scenarios, and a training of trainers course. The content of the basic and the advanced course is included in Supplementary Information files (S1 and S2 files respectively). Materials included training manuals, presentation slides, videos, practical exercises and guidelines for supervision and mentoring.

The feasibility of such training has not previously been reported. Understanding the

feasibility of delivering the training, and the experiences of participants is crucial for the intervention to be effective beyond the pilot phase. To address this gap, we aimed to evaluate the development of the ACT course.

Method

Study Design

We conducted a mixed methods evaluation between March 2015 and February 2017, based on a model proposed by Bowen et al (2010). The model is designed to assess the feasibility of public health interventions [9] and we evaluated the training from the eight facets of acceptability, demand, implementation, practicality, adaption, integration, expansion and limited efficacy. We evaluated both qualitative and quantitative data to gain a deeper understanding of the research findings.

Setting

Participants were clubfoot treatment providers from Ethiopia, Rwanda, the UK and Kenya. Trainers were international experts and Anglophone and Francophone regional trainers from throughout Africa. The training materials were appraised on nine occasions. The Basic Provider Course (BPC) was trialled four times (Ethiopia x3, Rwanda x1) and the Advanced Provider Course (APC) was trialled five times (Ethiopia x2, UK x1, Kenya x2)

Recruitment

All participants were sent information about the training prior to the start. There were no exclusion criteria and we invited all participants attending the training to participate in the evaluation.

Data collection

The extensive needs analysis and project planning, including the project funding application and minutes from the project team meetings, provided contextual background to inform our training design and application. The needs analysis included mapping of existing clubfoot service delivery and training opportunities, and information collected from key stakeholders including national and regional clubfoot programme directors, existing trainers and clubfoot service providers, and discussion with parents.

Semi-structured interviews with key informants and trainers started prior to the first training and continued throughout all nine training courses. The interviews undertaken with participants in the training courses were lightly structured and allowed

respondents to express in their own ways and pace. The interview schedule was subdivided into three parts:
(i) Questions concerning the process and structure of the training, to understand the demand for and acceptability of the training;
(ii) General questions about practical training opportunities to identify relevant activities to implement;
(iii) Specific questions about gaining knowledge and skills, to inform how to integrate mentoring.

Participants completed paper questionnaires by hand pre- and post-training. The key sections in the questionnaires included: participant demographics, the ranking of seven aspects of the course from most to least important, the most useful thing that was learned on the course and how to improve the materials. The questionnaires also included pre- and post-training confidence and knowledge on management of clubfoot. The confidence question format was statements to be scored on a five point Likert scale, ranging from “not at all confident” to “completely confident”, and the knowledge section was a single correct answer multiple choice question. A repeated-measures design examined changes immediately after the training session. Satisfaction questions were included on the end line questionnaire (S3: Pre and post training questionnaire).

National clubfoot supervisors completed a paper based skills checklist (S4: Skills checklist) in routine supervision visits to the participant’s clinic within three months of completing the course (Ethiopia and Rwanda).

A six-month follow up electronic survey was sent to all regional trainers who participated in the ACT courses held in January and July 2016 to understand uptake.

Data management and analysis
Two researchers in Public Health collated the responses to the open-ended questions into a word document and undertook a qualitative content analysis using the questions as the codes; similar responses were grouped, groups were titled and the number of responses counted. Using conventional content analysis, coding groups were derived directly from the text data [10]. We analysed satisfaction questions thematically and undertook a narrative description of themes raised in trainer questionnaires, interviews and the electronic survey. As the pilot training courses were conducted over a number of months, with the materials being revised after each pilot, the themes were examined for evidence of change over time in the participants' perceptions of the training.

All quantitative data were entered into a Microsoft Excel 2000 (Microsoft Inc., Redmond, Washington) software package and managed and analysed data using Stata 14.2 (StataCorp 4905, Lakeway Drive College Station, Texas 77845, USA). Categorical and nominal/ordinal data were analysed using simple descriptive statistics, including calculation of proportions, and exploration of a comparative analysis of cadre (background of participant) and sex was undertaken. Linear regression was then used to examine the associations between post-test score and the variable of interest (cadre and sex) after adjustment for pre-test score. For the composite confidence score, all participants' Likert scale scores were added together, and the means and standard deviations (SD) were calculated.

Detailed descriptive statistics for the participants are provided in our research that reports on the development of the training course [8] and included in S5: descriptive statistics of participants.

Ethics

This study was approved by the Ethics Committee of the London School of Hygiene & Tropical Medicine, ref:10412 /RR/3466. Informed written consent was obtained from all participants.

Results

Acceptability

The content of the training materials and style of learning were found to be effective and appreciated by participants.

> *'this was exactly the material that I needed to advance my knowledge at this stage'.* (Participant, Advanced Course)

Participant satisfaction improved when the limitations and parameters of the course were explained clearly in advance, as this allowed participants to manage their own expectations of the course.

Initial discussions on content resulted in debates on some areas, for example, on how much to correct a clubfoot before scoring its severity, about which there is little in the published literature. Contentious points were recorded and discussed with all stakeholders and consensus gained on presentation in the training [8]. In the final pilot courses, these consensus points were presented without generating much discussion, indicating their acceptability to the users.

The programme deliverers determined the training to be suitable through direct observation of engaged participants; supervision was provided by trainers (ratio of 1 trainer to 4 participants), which assisted the consistent engagement of all participants. The training approaches were found to be highly acceptable to participants, with several commenting that they appreciated the time for building relationships with the trainers and the informal learning and mentoring that occurred during the course.

The opportunity for hands on practical training was valued:

> *'Faculty with good physical dexterity, explained well positioning of the thumb, index and middle finger while correcting.'* – Participant casting on model legs

However, in the final versions of the training, when timing that allowed for practical sessions was maximised, requests from participants to have more hands on practice remained.

Some practical sessions included treatment of children with clubfoot. The factors that reduced acceptability for families included: insufficient explanation to parents, crowding of participants, taking photographs without permission, and parents' embarrassment in being questioned in front of a large group. Clear guidance to manage these situations was subsequently developed in response.

Demand

The needs analysis and pre-ACT consultation with existing trainers revealed a high level of demand for the development of a standardised training course. Emphasis on supervision and mentoring was built into all components of the training course, as identified in the needs analysis and requested by users. The majority of participants requested follow up training:

> "*very useful and will definitely help us…repeat training is necessary*" (Participant quotation).

Participants requested that a training structure be developed in their country so that regular refresher training and advanced training is available, and that their colleagues attend future courses.

Implementation

Trainers reported the challenge of presenting materials that they themselves had not written, and the need for very clear guidance around the topics to cover when delivering each section of the training, the timings of each component and the

instructions for interactive and practical sessions. As this guidance was refined, trainers' satisfaction with the materials improved. Participants noted that the training delivery was aided with an identified course leader to direct each course; specific guidance for course leaders was developed and included in the training materials, and mentoring by more experienced trainers was given to new course leaders of ACT courses, to develop local course leadership capacity.

Practicality

Environmental considerations for the training include adequate space, audio-visual equipment, ventilation, accommodation and refreshments. Participants reported that these issues played a very important role in the learning experience overall. Much organisation and support of local administration staff was required to identify appropriate participants and children with clubfoot and to assist in the daily running of the training course, the implementation of which was not assessed outside of the nine pilots. In addition, the increased volume of children for the clubfoot programmes after the training required consideration of local staffing requirements to absorb additional cases.

Adaption

The course materials were piloted in English in Ethiopia, Kenya and UK and in French in Rwanda, and provided many opportunities to consider both local contexts and their more global applications. After the initial pilots, trainers noted that some of the more active learning styles were less familiar to many local participants. The materials were designed to support an interactive and practical skills-based learning experience, including small group discussions, problem-based learning, case studies, teaching of practical skills, reflexive practice, action planning, using evaluation tools, and structured feedback on teaching, practical and communication skills. Ten clear and memorable key 'take-home' messages for the basic course were identified and agreed by the development team that were actively repeated and reinforced through the course. Trainers and participants reported enjoying using a variety of learning styles and finding these beneficial.

This training was provided to mixed groups of professionals and had the potential to lead to tensions due to medical hierarchies. Cadres of lead trainers were therefore mixed to model good relationships and communication, e.g. between physiotherapists and orthopaedic surgeons. In some cases, specific instructions were given to overcome professional boundaries that might affect treatment quality – e.g. any

clinician assisting a tenotomy was encouraged by the surgeon trainer to prompt the surgeon if the tenotomy was not considered to be complete.

Integration

16/35 (45.7%) of national trainers responded to the electronic survey sent at six months following attendance at either the January or July 2016 pilots. All (100%) of the respondents reported that they had been involved in training other healthcare workers in clubfoot treatment and used the materials provided. This included workshops, courses and lectures and the majority of trainers had trained between 16 and 25 providers in this time.13/16 (81%) of the trainers had been involved in mentoring other healthcare workers in clubfoot treatment since delivering the ACT course.

The respondents noted that the materials for the course facilitated change in the way that they teach, and the most commonly reported change involved the way of teaching a practical skill. All trainers wanted to integrate the ACT materials into their current training programme. Reasons given included the simplicity of the theoretical and practical material, that the materials underwent many reviews with stakeholders, that principles of adult learning are incorporated, and that the standardised approach is useful.

Expansion

Training with the ACT materials occurred in 20 countries (Burkina Faso, Burundi, Cameroon, Congo-Brazzaville, DRC, Ethiopia, Ghana, Guinea Bissau, Kenya, Madagascar, Malawi, Niger, Rwanda, Senegal, South Africa, Tanzania, Togo, Uganda, Zambia, and Zimbabwe) between April 2017 and February 2018. The training augmented existing regional programmes in these countries and program co-ordinators were encouraged to formalise the continuing professional development (CPD) gained through the ACT course on a local level. However, challenges include trainers not recruiting enough cases for the training, and requests to deliver the advanced course directly after the basic course with the same participants, as well as not understanding exactly what materials are needed to deliver the training.

Limited efficacy

The BPC training elicited change in the knowledge and confidence of the participants. Mean participant confidence increased from 63% (95%CI: 59 – 69%) to 88% (95%CI: 86 – 90%) post course. Mean participant knowledge increased from 55% (95%CI: 51 – 60%) to 78% (95%CI: 76 – 81%) post course. (Figs 1 and 2).

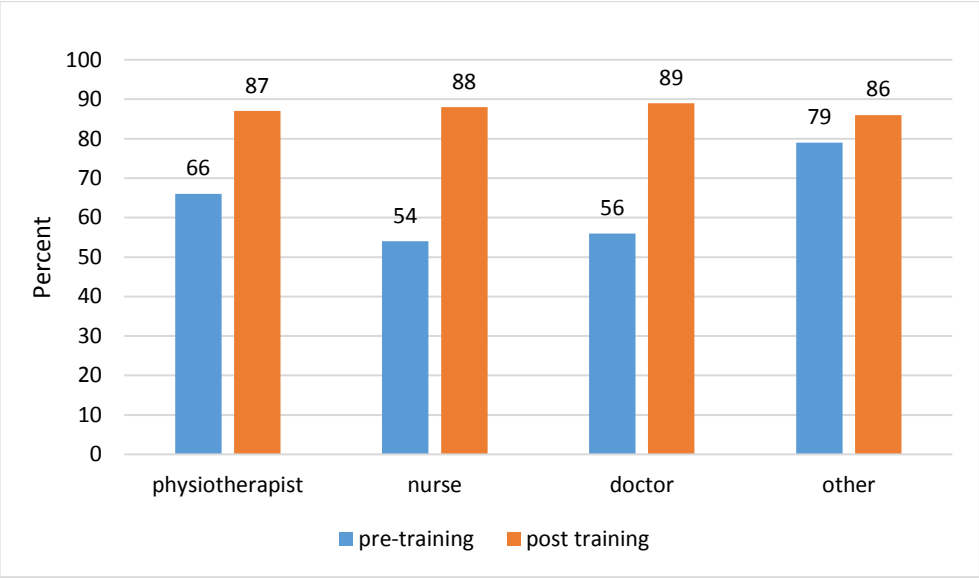

Fig 1: Pre- and post-course self-reported confidence as reported by cadre

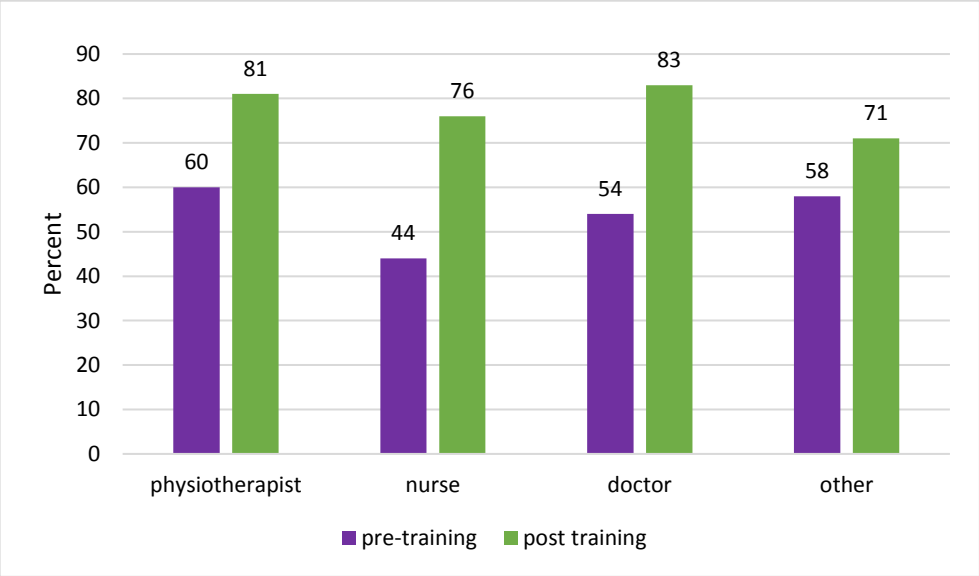

Fig 2: Pre- and post-course knowledge performance by cadre

There was no evidence for a difference between sex in confidence or knowledge when cadre was accounted for.

All participants practiced manipulation and casting on rubber models prior to the supervised treatment of children with clubfoot. The following comment reveals how a patient demonstration enabled a participant to improve their clinical reasoning skills:

> *'Live demonstration of the whole process from history taking to treatment was a good experience…has improved my professional judgement.'*

The ACT training may contribute to the successful supervised treatment of children with clubfoot (Ponseti method), even though the pilots were undertaken in a highly controlled setting.

Twenty-five participants who attended the clubfoot provider courses in Ethiopia were observed in their clinics. Over 80% (21/25) correctly assessed the clubfoot with the Pirani score and 92% (23/25) correctly manipulated the clubfoot. 22/23 health care workers met the criteria for correct application of braces. Participants met the fewest criteria in relation to discussing the Ponseti treatment with parents. Twenty participants who attended the clubfoot provider courses in Rwanda were observed in their clinics. Nineteen (95%) participants correctly assessed the clubfoot with the Pirani score and 17 (85%) correctly manipulated the clubfoot. 18 (90%) of the clubfoot providers met the criteria for correct application of braces. Supervisory notes identified one healthcare worker whom required additional support and assistance in several areas and a supervisory plan was initiated.

Discussion

This study evaluates participant, trainer and stakeholder responses to the training of clubfoot providers through Africa. Participants expressed enthusiasm and a high acceptability of the program, which they attributed to its clear purpose and the interpersonal interaction with the trainers. The role of an available trainer in direct observation and mentoring of the participants to give feedback and support cannot be overemphasized. Success is further indicated through the request of tertiary institutes in the UK to use the training materials.

The development of the training material required substantial organisation and commitment to trial the facilitated participatory training programme nine times. The combined use of quantitative data and qualitative findings produced a course design that was feasible and acceptable. Participants appreciated the final clinical content for being clear and well structured. The basic course content was distilled, with clear key messages. The advanced course introduced more complex concepts, clinical reasoning and opportunity to reflect on one’s own practice.

The development of a skills checklist allowed the identification of opportunities to strengthen and improve clubfoot treatment in participants who attended clubfoot provider training. The breakdown of specific skills within the checklist allows for targeted mentoring. For example, although the overall casting of the child’s foot was performed well, one area that consistently required improvement was casting over the

child's toes. In Rwanda, a high percentage of health care workers achieved all the criteria (>80%). Supervising individuals with the skills list allowed for the identification of one participant whom required assistance and closer mentoring.

Strengths and limitations

The methodology of this study allows a rich and deep understanding of the numerical data relating to the ACT, as well as the meaning that it had for both trainers and participants. This was a large prospective study that was situated in Anglophone and Francophone contexts. Study limitations include a short-term follow-up. The authors were closely involved in the implementation of the ACT project and in the development of the manuscript, which may lead to researcher bias. In addition, participants may have been reluctant to express their negative opinions or criticisms to the researchers since the interviewers were on the ACT development team. This study is not powered to detect a difference and there is no control group, however the figures for short-term improvement in knowledge and confidence indicate that this measure can be used in future training.

Lessons Learnt

Assessing our recruitment capability and resulting sample characteristics was important to determine if the training and future efficacy studies may be successful. Field notes and project team meeting notes were invaluable for the review of the content. Participants were eager to give feedback on the course materials and this provided an opportunity for ownership and engagement with the training course development. The selection of outcome measures was challenging and required continual assessment and re-evaluation. There was a need for clear instructions on how to run practical sessions in order to maximise their impact.

Implications and future steps

An understanding of the barriers to the success of delivering the training within individual country contexts is required for implementation, and opportunities to enhance the technology used in the training warrant further examination. Evaluation of the effectiveness of national training is required in future studies, in addition to studies that explore the retention of knowledge and confidence post-training. As countries cascade the training, the numbers of participants that complete training will increase. Recognising the growing global collaboration to decrease clubfoot as a disability by 2030 [11] and the heterogeneity of measurement and assessment, perhaps the time has come to draft globally agreed minimum standards similar to those for medical education programmes.

Conclusion

ACT has been received as a welcome addition to clubfoot treatment providers. Participants expressed high acceptability of the training, which they attributed to its clear purpose and guidance, convenience, and the interpersonal interaction with the trainers. The role of a supervisor, in direct observation of the participant, and dedicated mentoring that includes feedback and support, cannot be overemphasized. This is a particular challenge in low-resource countries due to the gap in services. The findings from this study are encouraging and merit further investigation in implementation process evaluations and larger clinical trials.

Acknowledgements:
The authors are grateful to the co-ordinators and organisers in Ethiopia, Rwanda, Kenya and the UK who facilitated training logistics. A further thanks to all of those who participated in the course, whose motivation and skills shaped the success of the project. We thank the Ethiopian Ministry of Health and Rwandan Ministry of Health and Human Resources for Health and the Ministry of Health for Kenya for their support of trainee attendance. We thank the ACT project Core Working Group, Africa Technical Advisory group, UK Technical Advisory group and the UK Clubfoot Consensus Group, without whose commitment and generosity, none of this project would have been possible. Lastly, we acknowledge and thank the multi-disciplinary ACT team involved in assembling, writing, piloting, reviewing and rewriting the ACT material.

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S1 Additional File: BPC Outline

BASIC CLUBFOOT TREATMENT PROVIDER COURSE (BPC) TIMETABLE

Start	Mins	Number	Session	Trainer
			DAY 1	
08:00	30”		Registration	
08:30	15”	1	Day 1 Opening Session (Welcome, course overview, introductions, ice-breaker, & formation of small groups)	
08:45	15”	2	Pre-course Assessment	
09:00	30”	3	Introduction to Clubfoot and the Ponseti Method	
09:30	30”	4	Anatomy and Definitions	
10:00	20”	5	The Clubfoot Deformity – CAVE	
10:20	30”		**Tea**	
10:50	30”	6	How to Assess the Severity of a Clubfoot Using the Pirani Score	
11:20	40”	7	The Ponseti Method of Manipulation	
12:00	30”	8	**Practical Session 1:** Small group hands-on manipulation of rubber models and skeleton models	
12:30	45”		**Lunch**	
13:15	30”	9	Applying Clubfoot Casts	
13:45	45"	10	**Practical Session 2:** Demonstration of Pirani score and casting (with patients)	
14:30	15”	11	Room Set up for Practical Session 3	
14:45	30”		**Tea**	
15:15	90”	12	**Practical Session 3:** Casting on rubber models (in small groups of 3)	
16:45	30”	13	**Practical Session 4:** Ponseti video	
17:15	15”	14	Day 1 Closing Session	
			DAY 2	
08:30	20”	15	Day 2 Opening Session (Review)	
08:50	30”	16	The Tenotomy and When to Do it	
09:20	40”	17	Maintenance Phase: Bracing and Relapse	
10:00	30”		**Tea**	
10:30	150”	18	**Practical Session 5:** Pirani score, manipulation, and casting with patients (in small groups of 3 or 4)	
13:00	60”		**Lunch**	
14:00	90”	19	**Practical Session 6:** (Parallel session) Group 1: Brace fitting Group 2: Tenotomy (theory & practical)	
15:30	30”		**Tea**	

16:00	30”	20	When to Stop and Rethink Treatment	
16:30	15”	21	Post-course Assessment	
16:45	15”	22	Day 2 Closing Session (Review, evaluation forms, and certificates)	

S2 Additional File: APC Outline

ADVANCED NON-SURGICAL CLUBFOOT TREATMENT PROVIDER COURSE (APC) TIMETABLE

Start	Mins	Number	Session	Trainer
			DAY 1	
08:00	30”		Registration	
08:30	30”	1	Day 1 Opening Session (Welcome, course overview, & introductions)	
09:00	15”	2	Pre-course Assessment	
09:15	45”	3	A Review of the Ponseti Method for Idiopathic Clubfoot	
10:00	30”	4	Advanced Ponseti for Idiopathic Clubfoot	
10:30	30”		**Tea**	
11:00	45”	5	Common Errors in Clubfoot Management	
11:45	45”	6	Recognition and Treatment of Atypical Clubfoot (includes video)	
12:30	60”		**Lunch**	
13:30	45”	7	Recurrent Clubfoot	
14:15	45”	8	Treating Older Children	
15:00	30”		**Tea**	
15:30	90”	9	**Practical Session 1:** 1. Refresher of casting on rubber legs 2. Demonstration & practice of new handholds & atypical casting	
17:00	10”	10	Day 1 Closing Session	
			DAY 2	
08:30	15”	11	Day 2 Opening Session	
08:45	30”	12	Secondary Clubfoot	
09:15	15”	13	Briefing on Patients	
09:30	30”		**Tea**	
10:00	180”	14	**Practical Session 2:** Case discussions (with assessment of patients, if available)	
13:00	60”		**Lunch**	
14:00	60”	15	Parent Education and Support	
15:00	60”	16	Clubfoot Clinic Set-up and Quality	
16:00	15”		**Tea**	
16:15	15”	17	Post-course Assessment	
16:30	30”	18	Day 2 Closing Session (Review, evaluation forms, and certificates)	

S3 Additional File 3: skills checklist

Training skills check	Skills demonstrated under supervision	Observed		Comments
		Yes	No	
Discuss Ponseti treatment with parents	Uses demonstration or descriptions to communicate to parents			
	Discusses the plan for treatment			
	Asks if the parents have questions			
Assessment with Pirani score	Completes entire Pirani score form including date and initials			
	Interprets and analyses findings to establish treatment plan			
Manipulation of child's foot	Good position of child and parent			
	Identifies lateral head of talus, correct handhold			
	Manipulates foot into accurate position to correct deformity			
	Uses gentle handling, 'soft hands'			
Casting of child's foot	Cast over toes			
	Moulds cast until dry			
	Lower half of cast applied first			
	Cast up to groin and gluteal fold			
	Knee at 90 degrees			
	Toes visible in casts			
	Regular talking with moulder			
Application of brace	Correct measurement of foot and selection of brace			
	Looks at heel cup to ensure heel is in brace correctly			
	Accurately conveys information regarding relapse			
	Makes appointment for review of brace			

S4 Additional File: ACT pre training confidence questionnaires for BPC

1. Enrolment

Please complete using CAPITAL letters

First Name:	
Surname/Family Name):	
Address:	
Email address:	
Mobile number including code:	
Age:	
Gender:	Male / Female
Course Location:	
Job title:	
Specialty:	
Name and address of place of work:	

4. Please place 'X' next to the 1 statement that describes you best:

1. Never seen Ponseti method before	
2. Seen but never used the basic Ponseti method before	
3. Only used the Ponseti technique under supervision	
4. Treated simple clubfoot cases with Ponseti method	
5. Competent in Ponseti method and would like to check and enhance skills	

5. **Pre-Course Confidence**

For each scenario in the table, please put a 'X' in the box that best applies to you

Scale of 1-5, where '1 = NOT confident at all' and '5 = Completely Confident'

	How confident are you to.....?	1	2	3	4	5
A	Assess a clubfoot using the Pirani score					
B	Treat a newborn child with clubfoot					
C	Treat a 6 year old child with clubfoot					
D	Manipulate a clubfoot using the Ponseti technique					
E	Talk to the parents about prognosis for clubfoot					
F	Apply a Ponseti POP cast to a clubfoot					
G	Fit a foot abduction brace and instruct parents on use					
H	Explain to someone how the bones are positioned in clubfoot					
I	Diagnose an "atypical" clubfoot					
J	Know when a tenotomy is needed					

ACT post course confidence BPC

2. Please RANK the follow aspects in the course, where '1' is MOST important, and '7' is the LEAST important

BASIC COURSE	
Manipulation using the skeletal models	
Casting on the rubber feet models	
Watching the Ponseti video	
Scoring and casting the feet of patients	
Brace-fitting and analysis	
Small group discussions	
Talking to trainers	

3. **Post-Course Confidence Matrix**

For each scenario in the table, please put a 'X' in the box that best applies to you

Scale of 1-5, where '1 = NOT confident at all' and '5 = Completely Confident'

	How confident are you to:	1	2	3	4	5
A	Assess a clubfoot using the Pirani score					
B	Treat a newborn child with clubfoot					
C	Treat a 6 year old child with clubfoot					
D	Manipulate a clubfoot using the Ponseti technique					
E	Talk to the parents about prognosis for clubfoot					
F	Apply a POP cast to a clubfoot					
G	Fit a foot abduction brace and instruct parents on use					
H	Explain to someone how the bones are positioned in clubfoot					
I	Diagnose an "atypical" clubfoot					
J	Know when a tenotomy is needed					

1. What was the most useful thing you have learned (or skill you have gained) during this course?

2. How could this course be improved? Please list 3 things.

   1.

   2.

   3.

Epilogue (Main findings and limitations)

Participants expressed high acceptability of the training, which they attributed to its clear purpose and guidance, and the interpersonal interaction with the trainers. The role of a supervisor, in direct observation of the participant and dedicated mentoring, contributed to the success of the training.

Twenty-five participants who attended the clubfoot provider courses in Ethiopia were observed in their clinics. Over 80% (21/25) correctly assessed the clubfoot with the Pirani score and 92% (23/25) correctly manipulated the clubfoot. 96% (22/23) clubfoot therapists met the criteria for correct application of braces.

Twenty participants who attended the clubfoot provider courses in Rwanda were observed in their clinics. 95% (19/20) clubfoot therapists correctly assessed the clubfoot with the Pirani score and 85% (17/20) correctly manipulated the clubfoot. 90% (18/20) clubfoot therapists met the criteria for correct application of braces.

Study limitations include (a) a short-term follow-up, (b) participants may have been reluctant to express their negative opinions or criticisms to the researchers since the interviewers were on the ACT development team, and (c) this study used a convenience sample size and was not powered to detect a difference in pre-post intervention knowledge and confidence.

SECTION C DISCUSSION

Chapter 13. Improving clubfoot services in Zimbabwe

Case study discussions in Kigali, Rwanda

This research aims to provide evidence to improve services for children with clubfoot in Africa. Synthesis of the evidence identified in the previous chapters is required to inform recommendations for improvements. However, no framework or method of evaluation is consistently used to analyse clubfoot services. Therefore in this chapter three approaches that are used to analyse health service pathways are presented. The use of these methods provides information on achievements and barriers to implementation of health services.

These approaches are used to analyse clubfoot service pathways in Zimbabwe. Limitations to each approach are considered.

Recommendations to improve clubfoot services in Africa are presented using evidence identified in the previous chapters and informed by the analysis of clubfoot service pathways and the case study of Zimbabwe.

13.1 The need

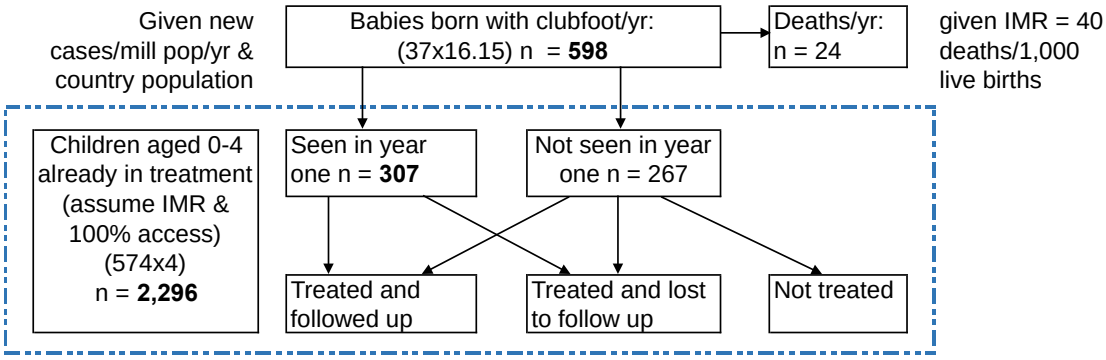
The Global Clubfoot Initiative (GCI) launched 'Run Free 2030' on 3rd June 2017, World Clubfoot Day. The strategic approach aims for 70% of children born with clubfoot in LMIC each year to access treatment, by 2030. With data provided by 55 countries in 2015, GCI estimated that 13% of children born with clubfoot in LMIC accessed treatment that year (1). Only ten (18%) of those countries (Bangladesh, Dominican Republic, El Salvador, Georgia, Honduras, Malawi, Namibia, Rwanda, Solomon Islands and Zimbabwe) provided a service that more than 50% of children born with clubfoot accessed within the year they were born. Therefore, the needs of children with clubfoot are not being fully met by health systems in LMIC.

We estimated the birth prevalence of clubfoot in Africa to be 1.11/1,000 live births (Chapter 4). Extrapolation to the population of Zimbabwe (16.15 million) with a crude birth rate of 33 live births/1,000 population (2), the number of children born per year with clubfoot in Zimbabwe is estimated to be 598. This assumes the birth prevalence of clubfoot is similar across Africa. In 2015, 307 children under the age of one year started clubfoot treatment in Zimbabwe (3). Therefore, 51% (307/598) of children in Zimbabwe with clubfoot access a

clubfoot clinic for treatment at least once within their first year. The proportion of children that start treatment after the age of one year is unknown.

Of the children that access care, we calculated the proportion with long-term success was approximately 72% (Chapter 8), as defined by an ACT score of 9 or more. Adherence to treatment was defined in Chapter 9 as children attending clinic appointments for two years or more; adherence in the cohort in Zimbabwe was 56% of children that were followed up. However, considering the infant mortality rate (IMR) of Zimbabwe is 40 deaths/1,000 live births (4), assuming the IMR of children with clubfoot is similar to the national IMR, an estimated 24 children with clubfoot will die before one year of age.

In Figure 12 below, these estimates are presented diagrammatically. The figure demonstrates the number of children with clubfoot born per year in relation to the number of children that will be in care between the ages of 0-4 years (assuming 100% access), and the need to identify follow up rates.



Legend: mill pop = million population; yr = year; IMR = infant mortality rate

Figure 12 Flow chart of access and follow up for children with clubfoot in Zimbabwe

13.2 Analysis of intervention pathways

Approaches to identify achievements and areas for improvement in clubfoot services are required but there is no framework that is consistently used to assess these services. Other health programme evaluations use a step-by-step analysis of implementation pathways to identify potential limitations to the service. Three such methods are (i) cascade analysis, (ii) bottleneck analysis, and (iii) community and systems effectiveness analysis (5).

A description of these three methods follows, with a view to adapting and using these approaches in the analysis of clubfoot services in Zimbabwe. Examples of how each analysis was previously used in programme evaluation are provided. These include evaluation of human immunodeficiency virus (HIV), malaria and child and maternal health programmes. A brief outline of the study aim, findings and recommendations is provided for each example to establish the rationale for use in clubfoot service evaluation.

Cascade analysis is a tool used to evaluate the success of the intervention delivery, acceptance, adoption and adherence. Data from both supply and demand indicators are considered. Indicators of supply relate to measuring the extent that an intervention is offered and available to the population in need. Indicators of demand of the population relate to the proportion of uptake of products, information, or procedures offered by the intervention. A loss in the proportion of the population between the cascade stages indicates where action is required.

For example, Garnett et al (2016) (6) used a cascade analysis to monitor HIV programmes in rural Zimbabwe. Study findings from 2012 – 2013 included an increase in proportion of men and women testing for HIV, whilst only a small proportion (12%) of eligible men sought to be circumcised. The application of the cascade analysis demonstrated that voluntary medical male circumcision was limited by supply in 2009 – 2011, but limited by demand in the following years. This informed recommendations for counselling services.

Bottleneck analysis is used to identify the limiting component of a system and is similar to cascade analysis. It is based on a coverage and evaluation model developed by Tanahashi et al (1978) (7) that was used to identify gaps in health service delivery. The components that are analysed are determined by the features of the intervention and may include efficacy of treatment, access of the eligible population, quality of the intervention and adherence. An indicator is defined for each component and each stage of implementation is conditional on the previous one being met. Therefore, when the proportion of success at each stage is multiplied sequentially (i.e. efficacy x access x quality x adherence), an

estimation of population level effectiveness is calculated. The ‘bottleneck’ is the component that limits the successive stages.

For example, this approach used cross sectional data to examine the effect of the EQUIP (Expanded Quality Management Using Information Power) project on maternal and new-born health in Tanzania (8). In the first district, health facility readiness was identified as the largest bottleneck. Access was identified as the limiting component in the second district. Recommendations from these findings included prioritising health service quality and provision of high-quality data to assist planning and prioritisation.

Community and systems effectiveness analysis relies on the assessment of steps in access, delivery and community effectiveness to evaluate the intervention. Indicators of the steps are determined *a priori*. The methods assume a linear process and that one step is necessary to achieve the next. Both quantitative and qualitative data from indicators may be used to identify the steps where success is achieved or improvement is required. Examples of this model of analysis include logical frameworks, logic models and theory of change. These terms are often used interchangeably. For the purpose of this PhD thesis, a logic model is defined as an illustration of programme components that depict programme activities and outcomes (9). A theory of change is defined as ‘*an approach which describes how a programme brings about specific long-term outcomes through a logical sequence of intermediate outcomes.’* (10) The theory of change therefore links outcomes with activities to explain how and why the desired change is expected to come about.

For example, Webster et al (11) aimed to explain the quantitative data from a prevention of malaria in pregnancy intervention in Mali with a theory of change approach. The study found that incorrect practices were recommended in training and supervision, and there was inconsistent management of pregnant women. The application of this method identified complex treatment guidelines and the lack of implementation guidelines. This informed the recommendations to address broad health system issues as well as specific intervention practises.

13.3 Analysis of clubfoot service pathways in Zimbabwe

The above examples demonstrate how improvements in health services were identified by three approaches that analyse intervention pathways.

The remainder of this chapter demonstrates the use of these evaluation tools for the assessment of clubfoot service pathways in Zimbabwe and then makes recommendations to improve services for children with clubfoot. Due to the similarity of approaches, the cascade analysis and bottleneck analysis are combined into one example. The second example is the community and systems effectiveness analysis.

Example (i). Cascade and bottleneck analysis approach

The first example uses the components of efficacy, access, quality and adherence in the analysis of clubfoot services in Zimbabwe. This combined cascade and bottleneck analysis approach is demonstrated in Table 6. The evidence provided in the previous chapters determines the indicators (column two and four). The assumptions and limitations of the indicators are outlined in column three. The proportion of children for whom the components are successful is outlined in column five. The population effectiveness of clubfoot services in Zimbabwe is then calculated step-by-step in the final column.

For example, efficacy of the Ponseti technique in trials is estimated as 90% (Chapter 1). If the component of '*acce*ss' is considered, defined as the proportion of the eligible population (children born with clubfoot in that year) that access treatment within the first year, then 51% of the eligible population access treatment that is 90% effective, so the population level effectiveness decreases to 46% (efficacy x access).

Table 6 Estimates of population level effectiveness of clubfoot care in Zimbabwe

Component of clubfoot service	Indicator	Assumption	Evidence	Estimated proportion of success (%)	Sequential population level effectiveness (%)
Efficacy of treatment	Ponseti treatment success in trials	Results from clinical trials are replicated in programmes	Chapter 1	90	90
Access	Number of children born/year that attend a clubfoot clinic in that year	No children access treatment after one year	GCI report and extrapolation to population	51	46 [(90 x 51)/100*]
Quality	Success of treatment measured with the ACT tool	Success in all children who access	Chapter 8	72	33 [(46 x 72)/100**]
Adherence	Clinic attendance for >2yrs of bracing	Less than 2 years bracing has no success	Chapter 8	56	18 [(33 x 56)/100***]

*Sequential calculations are indicated in figure 13 as * to ***

As demonstrated in the table, adherence to treatment for more than two years of bracing (56%) and access to treatment (51%) are identified as the two components with the lowest proportion of success (defined by the indicator). The information in this table is displayed in Figure 13. The blue columns demonstrate the proportion of success of individual components of the clubfoot service. The red line depicts total population effectiveness of clubfoot care in Zimbabwe.

The components that limit the overall effectiveness of the system (identified as absolute attrition from one stage to the next) are indicated by an arrow.

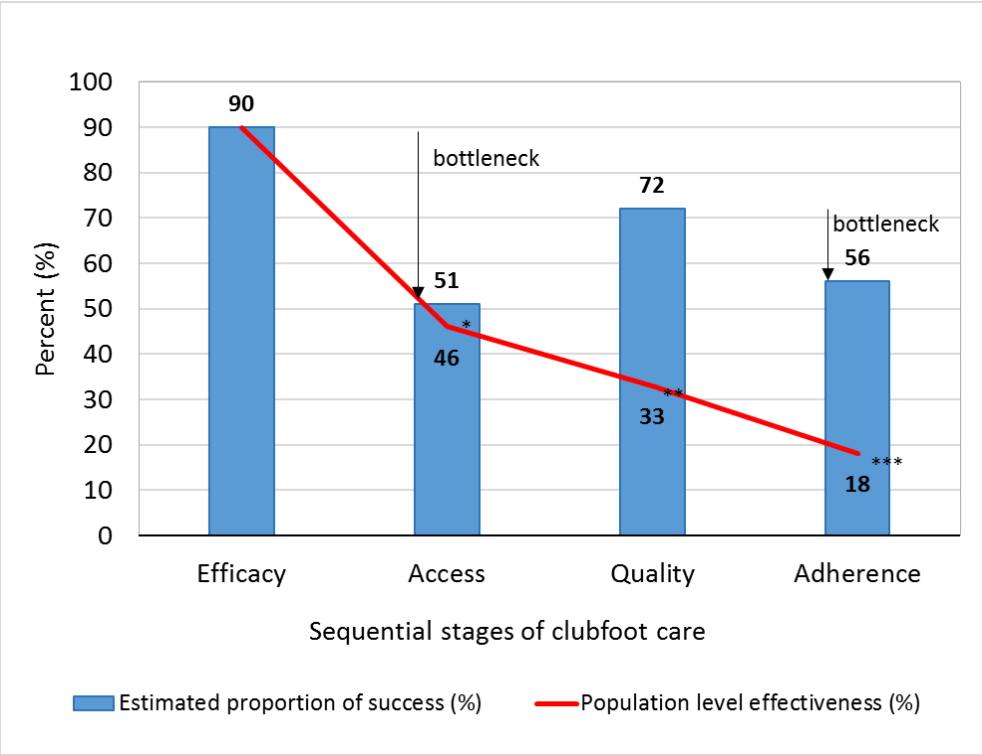
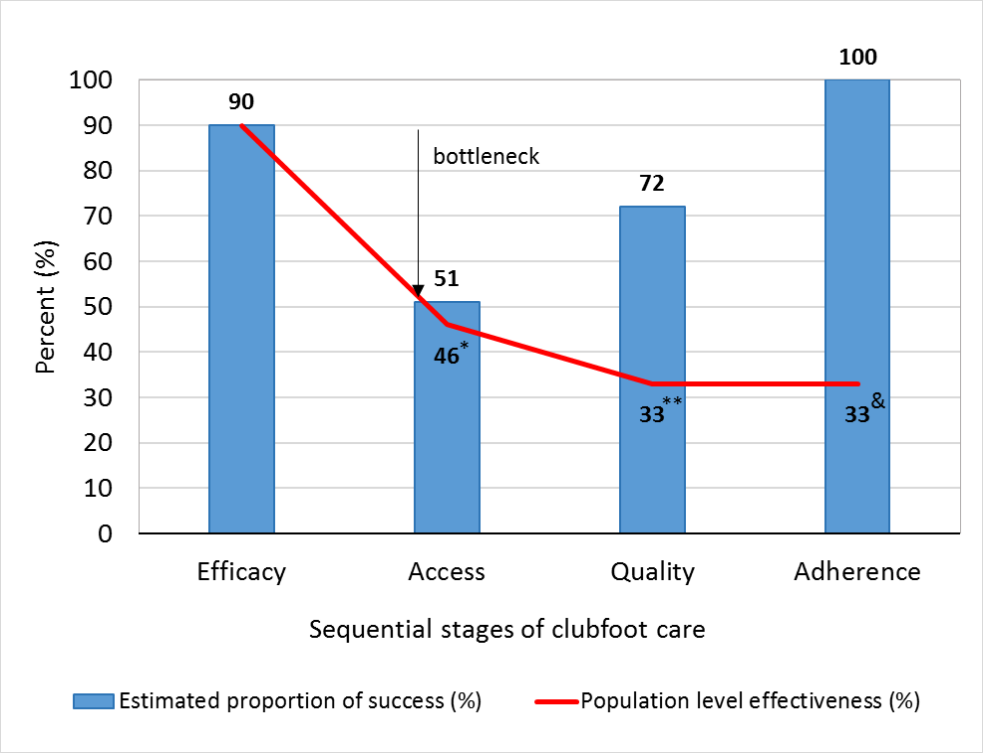


Figure 13 Cascade and bottleneck analysis

Consideration of all bottlenecks in the service is required. Figure 14 illustrates a hypothetical example. If every child adheres to treatment for two or more years of bracing (increase of adherence from 41% to 100%), the overall population effectiveness does not increase proportionately; access remains a limiting factor. Children need to attend clinic in order to adhere to either corrective treatment or bracing with a FAB of the appropriate size. Issues of access therefore also need to be addressed.



&Increase of total population effectiveness from the cascade

Figure 14 Hypothetical bottleneck analysis with increase of adherence

Limitations

Data analysed here were not collected with cascade or bottleneck analysis in mind. First, limitations arose from relying on cross-sectional and cohort data. The analysis does not consider the children that are treated after one year of age, or the treatment of children that were lost to follow up and then re-attend. Second, there are limitations in the definitions used in this example, as outlined in the assumptions column of Table 6. In this example, successful adherence is determined as those who attend treatment for two or more years, however it is recommended that children wear a FAB at night until the age of four. The cut off of 'two years or more' was used in Chapters 8 and 9 to assess the difference in outcome between children who wore the FAB for <2 years compared to those with ≥2 years of wear. This information is used for the cascade analysis, however the estimates do not conform to the recommended number of years for FAB wear. Third, whilst this analysis demonstrates that access and adherence need to be addressed, it does not explain how to improve either component.

Qualitative approaches are needed to support a nuanced understanding of the problems that may occur with each component and a theory of change may be

useful in providing a more comprehensive understanding of steps to improve clubfoot services.

ii. Community and systems effectiveness analysis

This example includes a logic model and theory of change to identify necessary improvements in clubfoot services.

First, at the start of this research, a model of user needs and service requirements in Zimbabwe was created (Figure 15). This logic model identified key actors and activities of clubfoot services and their hypothesised interaction. The desired short-term outcome was identified as increased coverage of good quality clubfoot services and is indicated on the right hand side. The networks are linked from left to right with arrows. The community and caregivers are at the top, the clubfoot clinic in the middle, and the health system is at the base.

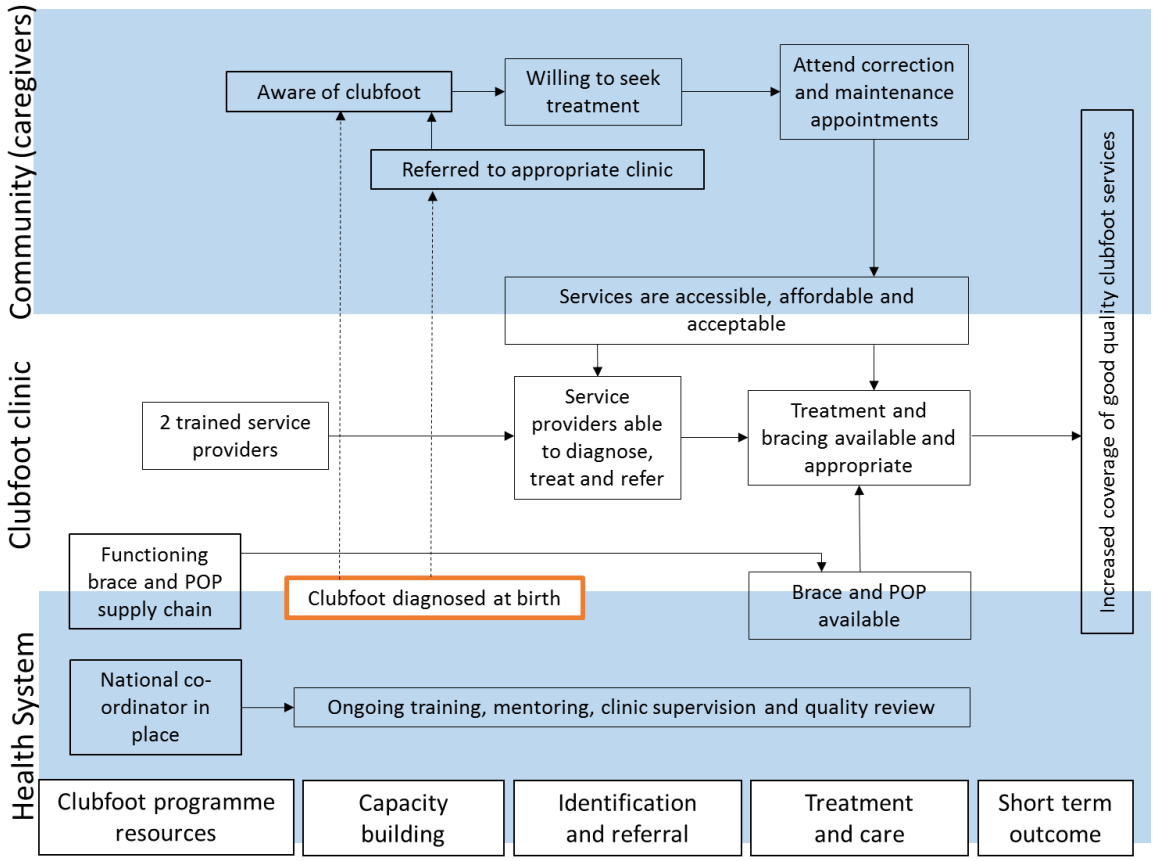


Figure 15 Initial network map of clubfoot services

Second, a theory of change for clubfoot services was developed to illustrate hypothesised causal pathways. Detail of the development of the theory of change is described in Chapter 1. This model considers the interaction of the

primary health care system, with the community and caregivers (on the right side), the clubfoot clinic (in the center) and the environment in which all of these are found, thus the enabling ecosystem of the health system (on the left side) (Figure 16). Distinct inputs are required at the level of the community, caregiver, primary health care, clubfoot clinic and health systems for good clubfoot management to be received. These activities are located at the base of the model. Assumptions of context and processes are noted in shaded boxes.

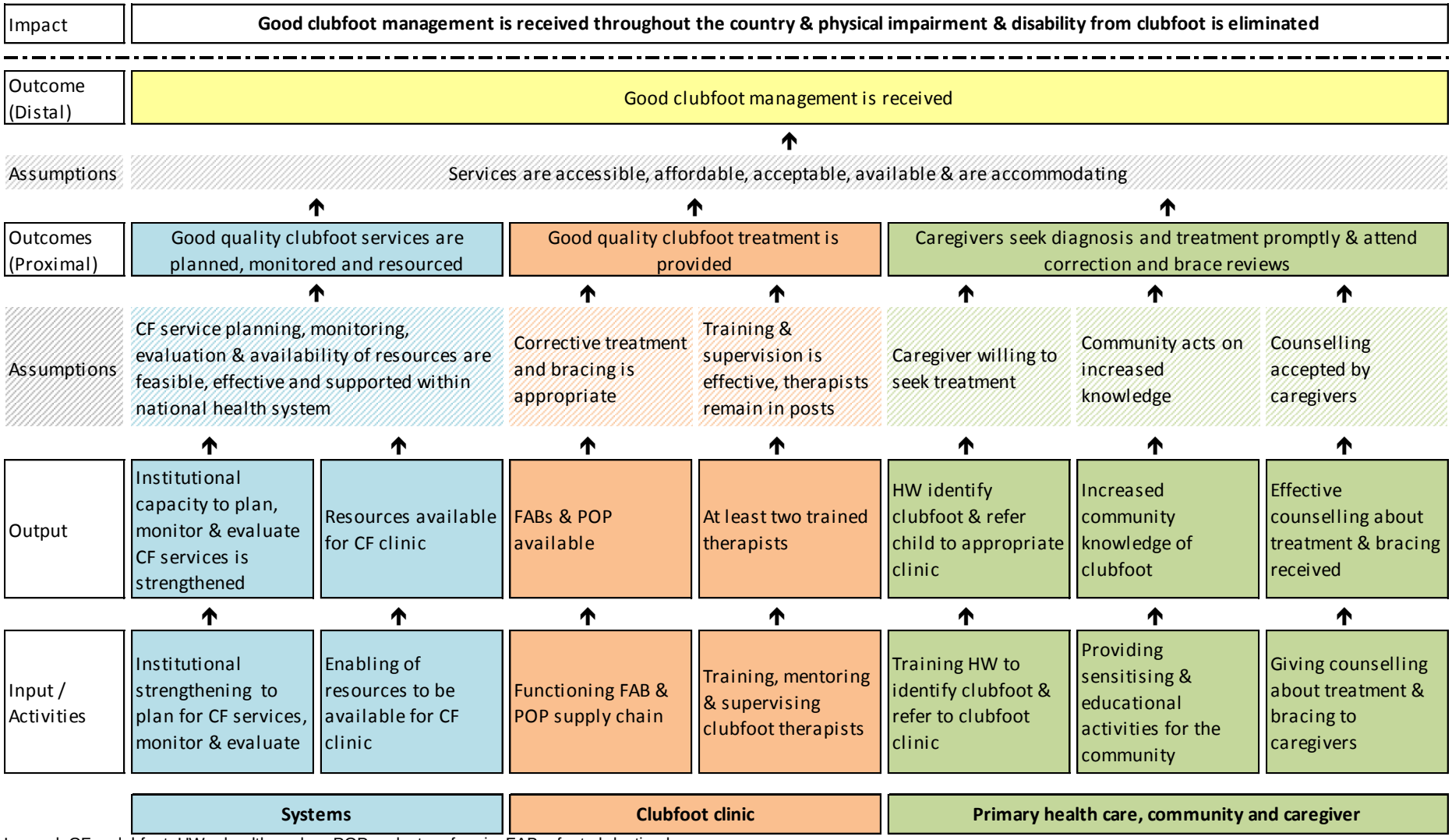
The theory of change illustrates how the planning, monitoring and resourcing of clubfoot services is needed in parallel with the capacity to provide good clubfoot treatment. However, for good clubfoot treatment to be received, these services must be matched by increasing access to care for children with clubfoot and adherence to treatment.

The previous chapters of this PhD thesis highlight the unmet need for indicators of clubfoot clinic functionality and success of long-term treatment. The two simple tools developed (FACT and ACT score) provide data to fill the information gap, and their use will contribute to assessing the proximal outcomes of ‘good quality clubfoot services are planned, monitored and resourced’ and ‘good quality clubfoot treatment is provided’ in the theory of change.

Limitations

The logic model is only conceptual. It focuses on expected outcomes and does not consider the dynamic interrelationships between resources, capacity building, identification, treatment and care. The theory of change was used to identify gaps in evidence for clubfoot service improvement. It requires further testing and may provide a framework for developing future indicators. The assumptions of the theory of change have not been tested.

The models were developed in parallel with the research and they are unable to make conclusive judgements on the impact of clubfoot services in Zimbabwe, as more information, particularly qualitative data, is required.



Legend: CF = clubfoot; HW = health worker; POP = plaster of paris; FAB = foot abduction brace

Figure 16 The theory of change for clubfoot services

13.4 Conclusions from analysis of clubfoot service pathways

Acknowledging these limitations, the use of the bottleneck and cascade approach suggests that access and adherence are two areas in which to prioritise action to improve clubfoot services in Zimbabwe. However, as demonstrated in the theory of change, increasing access to care for children with clubfoot and adherence to treatment is necessary but not sufficient for good clubfoot treatment to be received; it must be matched with improved quality of care and services that are appropriately planned, monitored and resourced. These elements of clubfoot services should be addressed in parallel.

Therefore, based on the needs identified in this body of work and guided by the analysis of clubfoot service pathways, the following recommendations to improve services of clubfoot care in Zimbabwe are made. They are embedded in a health systems-oriented approach.

13.5 Health systems-oriented approach for recommendations

The WHO defines a good health system as one that delivers *"quality services to all people, when and where they need them (12)* and is built on "*a robust financing mechanism; a well-trained and adequately paid workforce; reliable information on which to base decisions and policies; well-maintained facilities and logistics to deliver quality medicines and technologies." (12)* Health systems also include a combination of networks, such as public and private systems, local or regional systems, and social and organisational structures (13).

The analysis of clubfoot services that is co-ordinated with a health system-oriented perspective may contribute to the decrease of functional impairment and disability from clubfoot. Good quality clubfoot care cannot occur in a condition-specific silo. An example to illustrate responses to strengthen clubfoot care is as follows: Whilst programmes and health systems both use performance and results based financing, a clubfoot programme response to poorly motivated staff may be to offer financial incentives to reward delivery of priority services. The additional actions of a health system response include clarifying roles and expectations of performance, reviewing salary structures

and promotion procedures, and facilitating the implementation of performance reviews (14).

The following recommendations to improve clubfoot services in Africa are discussed with consideration of the six functions of health systems as outlined by the WHO (15):

1. Governance and leadership
2. Human resources
3. Financing
4. Essential equipment
5. Technology and health information systems
6. Service delivery

13.6 Recommendations for improving clubfoot services in Zimbabwe

Evidence from this research as identified in the previous chapters suggest the need to improve all six areas and (i) strengthen health system leadership (planning) for clubfoot services; (ii) provide human resources for clubfoot care; (iii) finance clubfoot care; (iv) make essential equipment, including POP and FAB, available; (v) provide and monitor an information system for clubfoot management; and (vi) improve service delivery for children with clubfoot.

i. Strengthen health system leadership and planning

Support for national policy, planning, and resourcing monitoring and evaluation is required by Ministry of Health. The provision of resources and implementation is required at Provincial level. Both support and provision require policies, management and plans for clubfoot services to be informed by evidence. However there is an evidence gap. Clubfoot services need to be supported to include clinical, operational, social and health system research to determine ways to improve access, adherence rates and the experiences of children and families attending clubfoot clinics. For clubfoot care to be sustainable the Ministry of Health must lead with accountability. Integration of clubfoot services with national child health and paediatric services is likely to promote sustainability.

ii. Provide human resources for clubfoot care

Task sharing approaches are recommended in resource constrained settings where health worker shortages limit the availability for specialised care (16). A clear definition of roles and agreement on responsibilities is required to optimise health worker performance without compromising patient outcomes. A training course for clubfoot therapists in Africa was found to be feasible and acceptable (Chapter 12). Performance standards should be set for the training outcomes as well as for the implementation of training. The requirements for mentoring and supervision of clubfoot therapists need to be understood. Indicators to measure activities to improve the provision of quality of care can be created and will allow informed decisions to be made to improve quality of care.

The ACT score provides a simple to use indicator to monitor the quality of clubfoot treatment (Chapter 8) and may assist clubfoot therapists to determine their long-term results, and identify which children need referral for further intervention.

Further strategies to facilitate early referral include identification of clubfoot in newborns by primary healthcare workers and during the postnatal period in the community, and the provision of appropriate counselling to parents about the next steps. The support and integration of detection and screening of clubfoot with programmes for child health may assist early referral to services. For example, the inclusion of case management for physical impairment within a child health delivery strategy, such as the Integrated Management of Childhood Illness (IMCI). The IMCI has been adopted by 102 countries as the preferred primary health care delivery strategy for ill children under 5 years (17). Training of health workers in IMCI was initiated in 1998 in Zimbabwe (18). The integrated care model focuses on management of common childhood illnesses such as acute respiratory infections, treatment of dysentery and the common causes of fever through specific case management steps and algorithms. Given the ‘survive, thrive and transform agenda’ of the WHO’s Global Strategy for Women’s, Children’s and Adolescents’ Health (19), perhaps there is need to move beyond ‘survive’ and include strategies to address ‘thrive and transform,’ which include early detection and referral for children with physical impairment, such as clubfoot.

iii. Finance clubfoot care

Health care is provided free of charge for children under the age of five years in Zimbabwe, however the costs of transport and parents' time away from work can be prohibitive to seeking treatment. No child should experience pain and disability from clubfoot because their family cannot afford treatment. Strategies to contain costs for children with clubfoot need to be considered (e.g. income generation from private patients).

Key service provision components include salaries for staff and financed equipment and consumables. Salaries for clubfoot therapists are not high when compared to specialists and consumables include POP and locally made FAB, which in Zimbabwe cost USD2-5 and USD18 respectively. Given that the number of children born with clubfoot per year is estimated to be less than 600, integrating the finance of clubfoot care within the national health budget is an achievable goal.

iv. Make essential equipment, including POP and FAB, available

National policies and regulations that support policy are required to provide an enabling environment for the functioning supply chain of essential equipment for clubfoot care (e.g. POP, FAB, and tenotomy sets). Cost containment measures such as waivers of import duty for POP will improve procurement. The local production of braces and regular quality assessment of the FAB produced can support reliable manufacturing practices and sustainability. For example, FAB are manufactured in Ruwa, 30 minutes outside of Harare in Zimbabwe and provide employment for three technicians. The monitoring and planning of orders, distribution and stock-keeping of POP, FAB, and tenotomy sets will assist to ensure availability of these essential medical equipment.

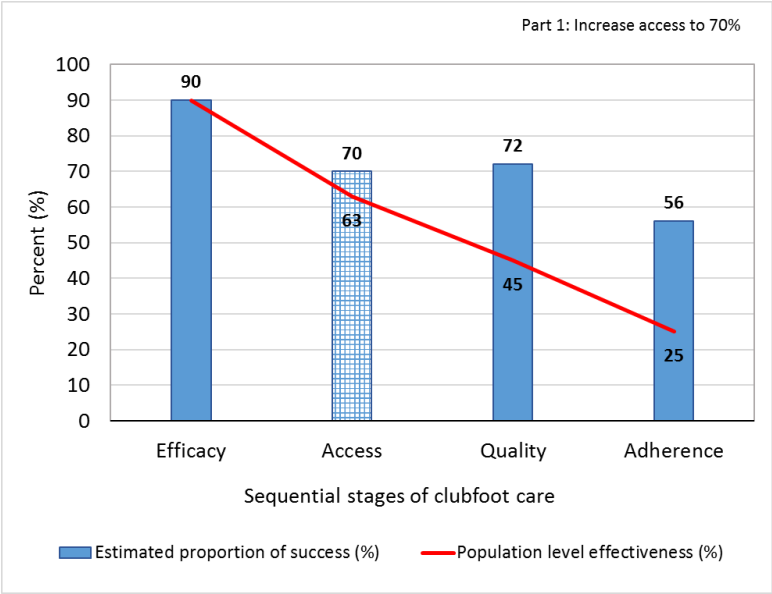
v. Provide and monitor information systems for clubfoot

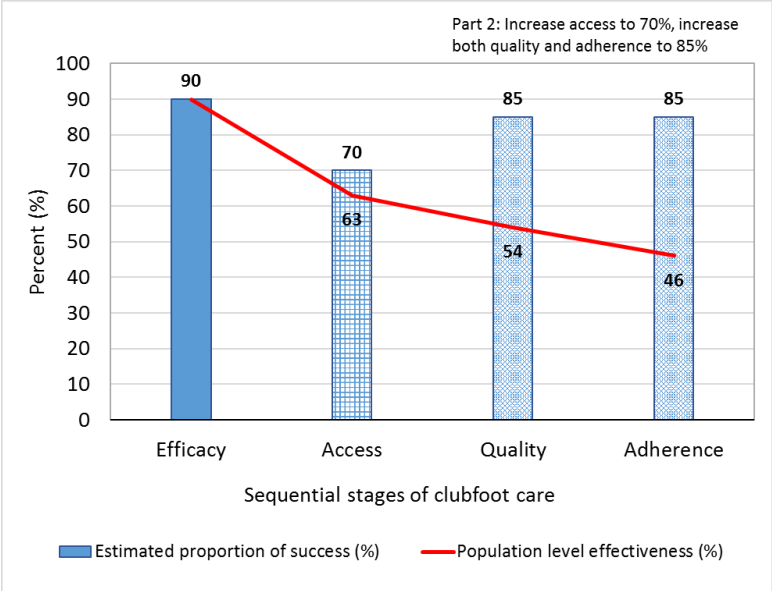
Functioning health information systems are necessary to improve services for clubfoot. A good medical records system allows patients to be tracked and trends in treatment to be monitored over time. Nationally defined care pathways contribute to a robust referral system and allow comparison between clinics. Adequate record keeping is essential to identify children that are being lost to follow-up. Analysis of care should occur for the eligible population and not

merely for the population for which complete records are available. While success of treatment can be determined through analysis of documented clinical findings, complex paperwork systems or the lack of a recording system make central analysis and identifying individual clinic results difficult. Clubfoot therapists may be motivated to record data that are seen as immediately beneficial, and identification of simple processes to collect these data is needed.

The analysis of clubfoot service pathways may identify potential stumbling blocks to implementation. For example, in Zimbabwe 51% of the population that need the service accessed it before one year of age. This parameter can be used to monitor progress over time. As the Run Free Global Strategy to end clubfoot disability aims for 70% access in 70% of LMIC by 2030 (1), these figures will be used in the following illustration.

Figure 17 includes a series of graphs that demonstrate the hypothesised improvement of 70% access (indicated with a patterned column). In part 1, an increase of access from 51% to 70% results in an increase from 19% to 25% of population level effectiveness. In part 2, activities to increase both adherence and quality to 85% raise the total population effectiveness, however the clubfoot service still does not have an effect on more than half of the eligible population and access remains the bottleneck. Strategies to increase all three components of access, quality and adherence are required.





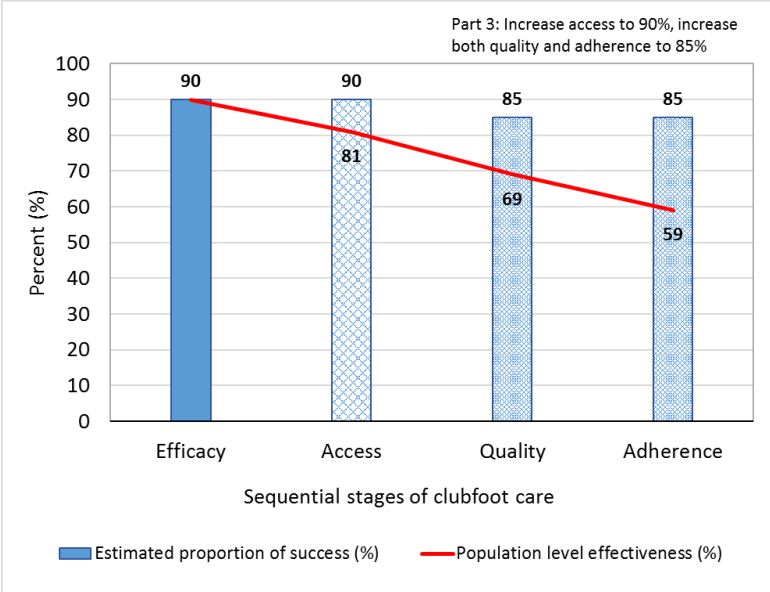


Figure 17 Models of bottleneck analysis

The goal of 70% of the eligible population accessing clubfoot services in 70% of LMIC may be a lofty benchmark given the lack of recording and collection of national level data from clubfoot services. Accurate data are needed to inform services, however measurement has been limited by lack of consistent tools and consensus on what data to collect. Data on the number of children disaggregated by age, a measure of quality of long-term treatment (as proposed with the ACT score) and data on follow-up will provide this baseline. In addition, the provision of greater access to services, with the design and implementation of demand creation activities is required to achieve the target of 70%.

vi. Improve service delivery of clubfoot management

Case volume can be used to inform the number and distribution of clubfoot clinics per population. In addition, the regular treatment of children with clubfoot may assist to maintain the skills of clubfoot therapists. The delivery of treatment for children with clubfoot, when and where it is needed, therefore requires the balance of quality of treatment and accessibility. An additional clinic may increase coverage, however in sparsely populated areas the balance between the cost of the service, the number of children that access the service, and the quality that these services can provide needs to be considered.

For example, service units of one million population have been used to estimate need in eye care programmes (20). This ‘district’ level calculation of need and resources per million population per year assists health planning and management and allows extrapolation for national requirements. District level need can be calculated for clubfoot services. The example in Table 7 uses birth rate data from Zimbabwe and the average number of casts from the case series (Chapter 6) to demonstrate estimation of need per one million population.

Table 7 Estimation of clubfoot service need per million population in Zimbabwe

Estimation of Need	Term	Actual	Comment
Birth rate (births/1,000 population)	BR	33	From demographics
Births/mill population	BR/mill	33,000	Calculation (BRx1,000)
Birth prevalence of clubfoot/1,000 births	BPCF	1.11	Chapter 4
New clubfoot cases/mill pop/yr	BPCF /mill	**37**	Calculation (BR x BPCF)
Prevalence of clubfoot age ≤4yrs (ie include all children from birth – 4 yrs)/mill/pop/yr	PCF	**148**	Assumes no mortality and no treatment of cases; calculation (BPCF/millx4)
Service delivery			
New cases needing casting/yr/mill pop	New cases	37	BPCF/mill
Average number of casts until bracing	No casts/ case	7	Chapter 6
Number of casts for new cases/yr/mill pop	No casts	259	New cases x No casts/ case
Number of casts/week/mill pop	No casts/wk	**6**	Assumes clinic works 47 wks /yr, and 100% access
Number of tenotomies/yr/mill pop	No tenoty/yr	**30**	Assumes tenotomy coverage = 80%. calculation (new cases x 0.8)
Number needing bracing from 0-4 yrs/mill pop	All bracing	148	Total 4 years of bracing
Average number of brace visits/child/yr		4	Assumes 100% adherence
Number of brace visits/yr/mill pop	Brace visits/yr	592	Assumes 4 visits/yr
Number of brace visits/week/mill pop	Brace visits/wk	13	Assumes clinic works 47 wks/yr; no mortality; no loss to follow-up; 100% access

Legend: mill = million; wk = week; yr = year; pop = population; tenoty = tenotomy

Therefore a clubfoot clinic serving one million population in Zimbabwe can expect to have about 150 children (aged 0-4) under its care at any one time, with approximately 37 new cases per year (assuming no mortality). It will need to complete an average of 6 casts and 13 brace fittings per week and a tenotomy every 2 weeks. The clubfoot clinic is likely to require 3 – 4 trained clubfoot therapists to deliver care for this estimated caseload.

As of May 2018, Zimbabwe has 13 clubfoot clinics for a population of 16.15 million, with one clubfoot clinic for each of the ten provinces and additional

clinics in the densely populated Harare and Bulawayo Provinces (two and one additional clinics respectively). The proportion of clinics that offer suitable services can be calculated with the FACT (Functionality Assessment clubfoot Clinic Tool) (Chapter 10).

13.7 Understand demand

Clubfoot is easily recognised and diagnosed and key to treatment is access to the service. Access requires caregiver knowledge about the service, finance and the decision to attend. In addition, there is a role for both caregiver 'education' and for the ability of the health worker to detect, counsel the parents and refer the child to the appropriate service. Identifying and understanding the pathways that permit people to engage with services will inform the planning of accessible services for clubfoot care (21). For example, treatment seeking behaviour and theories of causation of the clubfoot deformity have been explored in Malawi and Uganda (22-25). The studies found that lack of income, additional responsibilities, the beliefs of the caregiver and limited support from fathers and extended family affect treatment-seeking behaviour. Additional barriers to care include insufficient information about treatments and long distances to clinics. Interventions to address these challenges, such as counselling sessions, outreach clinics, brace recycling and a range of education programmes, are understudied. Examples of approaches to community sensitisation include designing education resources with caregivers. Caregivers and family members have unique insights about their challenges and situations, but are often excluded from the design process about issues that directly affect their lives.

13.8 Conclusion

Physical impairment and disability from clubfoot is preventable. It requires a health system oriented approach.

It is proposed that clubfoot therapists can effectively treat children with clubfoot (using the Ponseti method) in appropriately equipped clubfoot clinics in Zimbabwe. The case series found that the short-term outcome was successful (defined as a Pirani score of 1 or less) in 85% of feet, with a low loss to follow up (8.9% of children), when delivered by clubfoot clinic staff in Parirenyatwa

hospital, Harare. After 3.5 - 5 years, 72% of children achieved success (an ACT score of 9 or more). The cross-sectional clinic service provision study identified strengths of the clubfoot clinics in Zimbabwe; all clinics had an identifiable person in charge, two or more clubfoot therapists trained in the Ponseti method were regularly available and clinics occurred on a specific day.

The studies also demonstrated areas that require targeted intervention. Resources in clubfoot clinics need to be planned and managed appropriately; the estimate of service units of one million population provides useful information to plan treatment services. In addition, qualitative data on access and adherence is necessary to support good clubfoot management being received.

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Chapter 14. Strengths and limitations

Baby with bilateral clubfoot
(Credit: ACT project)

The strengths and limitations of the study methods and designs are examined in the individual research papers included in this PhD thesis. This chapter includes a summary table of specific limitations from individual studies. A discussion of the methodological strengths and limitations of the research study follows, and key features of the study design are considered in further detail.

14.1 Individual research paper limitations

A number of limitations of individual research papers need to be considered and are summarised on the next page (Table 8).

Table 8 Summary of limitations of individual research papers

Research Question	Method	Main Limitations
1	Systematic review and meta-analysis	• Included studies are characterised by low quality and narrow representation; • Two large databases strongly influenced the birth prevalence of clubfoot in China; • Only two papers contribute to the estimates of Europe (Turkey) and the South East Asia Region.
2	Systematic review and integrative review	• Lack of a consistent measure of success and insufficient follow up of cases restricts the conclusions; • The majority of studies were observational; • Limited external validity: many children were recruited from University and tertiary hospitals/ national centres, and not representative samples.
3	Delphi study	• The panel may not be representative of all Ponseti treatment practitioners; • The ratings of the criteria may have been influenced by the expert panel composition; • There may be some important criteria that were not considered.
4	Case series: retrospective analysis of clinic records	• The clubfoot clinic records did not include all the details for risk factor analysis and therefore limit the analysis; • There is no comparator within the case series, attribution of success to the intervention is likely but not proven; • High risk of selection bias: the children are self-selected and attended a tertiary clinic that may not be representative of the clinics that other children in Zimbabwe attend.
4	Cohort study: prospective follow-up	• Only 31% (68/218) of the original cohort of children attended for long-term follow-up; • High risk of selection bias: those followed up are likely to have better results than the whole cohort; • Results are only applicable to children with idiopathic clubfoot and not secondary clubfoot; • Likelihood of observer bias: the ACT tool and the full clinical assessment were administered by the same two people.
4	Diagnostic accuracy study	• Not all tools that were used were developed to identify need for referral for further intervention; • No distinction was made between a clubfoot that may not have been fully corrected and a clubfoot that had recurrence of the deformity; • Conclusions can only be drawn for use with idiopathic clubfoot.

Research Question	Method	Main Limitations
5	Delphi study	• The thresholds for the VAS mean and SD were chosen to include the highest ranked criteria with least variation in opinion, there was no published evidence for the chosen cut off; • The Panel was not representative of non-specialised health workers that deliver clubfoot care; • Additional data may be relevant to have a complete understanding of clubfoot clinic functionality.
6	Cross-sectional clinic service provision study	• Likelihood of observer bias: data were collected by a clinic supervisor well known to the clinics and who was internal to the research
7	Mixed methods evaluation of training development	• High risk of selection bias: survey participants were self-selected; • Absence of a pass/fail competency; • Likelihood of researcher bias: the authors were closely involved in the implementation of the ACT project and in the development of the manuscript.
7	Feasibility study of training course for clubfoot therapists	• Short-term follow-up; • There is no control group; • Likelihood of observer bias: the interviewers were on the ACT development team; participants may have been reluctant to express their negative opinions; • Not powered to detect a difference in pre-post intervention knowledge and confidence and there is no control group.

14.2 Strengths of this study

The use of both quantitative and qualitative methods, with a cohort study, a cross-sectional clinic service provision study and a pre-post evaluation of training, presented multiple perspectives of evidence to improve clubfoot services in Africa, which is a strength of is study.

This study also proposed an outcome measure (ACT score) to assist clubfoot therapists to identify successful treatment in a child of walking age, and when to refer for a second opinion or further treatment.

In addition, this study identified a tool (FACT) to enable programme managers to monitor the functionality of the national clubfoot service. The use of the tool provides information on areas for improvement and allows the comparison of clinics.

I understood the settings and the contextual factors as well as the medical implications of recommended actions, and this allowed for a comprehensive evaluation of findings.

14.3 Strengths and limitations of the study design

The study design includes a (i) case series, (ii) cohort study, (iii) cross-sectional study, and (iv) a mixed methods evaluation to provide data on treatment outcomes, clinic functionality and training feasibility.

Short-term treatment: case series

A tertiary hospital based cohort in Harare, Zimbabwe, was chosen for the study. A retrospective design is suitable for data analysis after the intervention has been implemented. Clinic records that were completed between 2011 and 2013 were analysed in 2015. The primary disadvantage of this study design is the limited control that the investigator has over data collection, and the medical records may not have all the details for a comprehensive risk factor analysis. However, well-maintained medical records for over 200 cases provided one of the largest case series reported in sub-Saharan Africa. Parirenyatwa was therefore well placed to provide the data to create a foundation for the prospective cohort study. Although the case series was self-selected and there

is no comparison group, the results are in line with the published studies in sub-Saharan Africa.

Long-term treatment: cohort study

The cohort was followed through time to evaluate long-term success of treatment. The prospective study design allowed specific data to be collected. Selection bias in prospective cohorts can occur at baseline and as a result of differential loss to follow-up. To address possible selection bias between those followed up and those not followed up, efforts were made to find as many children in the case series as possible from phone numbers and physical addresses. In the pilot phase, efforts to minimise loss to follow-up included study team members visiting all home addresses provided in clinic notes for participants that could not be reached by phone. In cases where the family had moved, the study team met with neighbours and visited door-to-door in an attempt to identify the new residence of the participant. Reminder text messages were sent to caregivers on the day before treatment. Despite these efforts, only 68 children of the cohort of 218 children (31.2%) attended. This selection bias affects papers in Chapter 8 and Chapter 9. The implication of this bias is that the children that were followed up (n=68) are likely to have better results than the entire cohort (n=218) as length of time in treatment is a predictor for good outcome.

In addition, the clubfoot services offered by clubfoot therapists in the 13 clinics may be acceptable, but this study did not assess the results of the Ponseti method of clubfoot treatment by clubfoot therapists in all the clinics in Zimbabwe; it evaluated a tertiary clinic in Zimbabwe and the ACT score was assessed with expert physiotherapists.

Clinic analysis: cross-sectional study

Cross-sectional designs are suitable to estimate the prevalence of a characteristic in a population. A strength of this cross-sectional service provision study is that all clinics participated and data were collected by the same investigator over six months, which minimised the potential for non-response bias. As the study design allows the examination of data at one

particular time point, there is no loss to follow-up. However, only an association, and not causation, can be inferred from the study.

Training evaluation: prospective mixed methods

The mixed methods evaluation of the Africa Clubfoot Training project was a before-after design that involved both qualitative and quantitative methods. It measured the occurrence of outcomes before and again after the training was implemented. Limitation of this design is a lack of control of external factors. The study does not control for elements that may change at the same time that the intervention was implemented (e.g. an increase in the availability and supply of POP). Attribution of the change in knowledge and practice to the training is plausible. However, it is not proven.

14.4 Generalisability of results

The findings and recommendations of this research are likely to be applicable to clubfoot services in contexts in sub-Saharan Africa where the challenges faced by most clubfoot therapists and their patients are similar, such as limited human resources and specialised equipment. However, the cohort was based in a large tertiary hospital and may limit the generalisability of results to small clinics. In addition, the application of results is only to idiopathic clubfoot and not secondary clubfoot. The inclusion of all ages in the cohort that attended the clinic between 2011 and 2013 reflects the likely range of age groups that are treated in clubfoot clinics. This improves the generalisability of the results. This analysis of ‘a real life situation’ is a strength of the study, however in this case it is also a weakness as the study cannot be perfectly designed.

14.5 Ongoing research

Ongoing research questions were framed by the gaps in evidence identified through this body of work. Current research includes providing evidence for the development of clinical guidelines and protocols to manage older children with clubfoot in Zimbabwe. This need was identified through the prospective cohort study, in which some children that had been lost to follow-up were reviewed. These children are older and typically have an element of recurrence of the deformity. A young baby can be treated with manipulation and casting,

tenotomy and bracing; an older child may require modified casting techniques in close consultation with surgical teams. There are currently no national guidelines in Zimbabwe for this treatment pathway and further research is needed to inform the best management guidelines for older children.

Chapter 15. Conclusions and recommendations

Training in Tanzania, April 2018

This final chapter draws upon the entire PhD thesis to address the aim of the thesis and the research undertaken to achieve this aim. The findings and recommendations for each research question are summarised. Suggestions for future research and the implications for policy and clubfoot programmes conclude this chapter.

15.1 Review of the aim of this research

The aim of this PhD thesis was to determine how services for children with clubfoot in Africa can be improved using Zimbabwe as a case study.

Research methods to achieve this aim included:

1. A systematic review and meta-analysis, to investigate the birth prevalence of clubfoot in LMIC. These estimates informed the actual need, so that the resources required to develop services for children born with clubfoot in Africa can be planned and made available.

2. A second systematic review, to determine and evaluate how results of clubfoot management in sub-Saharan Africa are reported. This review identified that clinical assessment was commonly used to report short-term outcome, with the Pirani score the most frequent outcome measure used. It identified gaps in how the long-term results of clubfoot treatment are reported.

3. A Delphi process, which established key criteria for measuring success of treatment in children of walking age.

4. A retrospective case series of 218 children with idiopathic clubfoot, treated in one clinic in Harare, Zimbabwe. The short-term outcomes of clubfoot treatment were documented and factors that influence the outcome were identified.

5. A cohort study, to document long-term outcomes of clubfoot treatment at 3.5 - 5 years follow up. Factors that influence the outcome were identified and results of five outcomes measures were compared.

6. A second Delphi study, that identified the components required for a good clubfoot service and clinic in Africa. The indicators were piloted in a tool to evaluate the Zimbabwe clubfoot clinics.

7. A prospective mixed methods (both quantitative and qualitative) evaluation, to assess the feasibility of a training programme for clubfoot therapists in Africa.

15.2 Main findings and conclusions

Research Question 1: How many children are born with clubfoot per million population in African countries each year?

We estimate that approximately 43 children/million population are born with clubfoot in sub-Saharan Africa each year, based on a birth prevalence of 1.11 (95%CI: 0.96 – 1.26)/1,000 live births and a crude birth rate of 38.3/1,000 population. These estimates will be useful for planning of services and to estimate areas of need for country programmes.

Research Question 2: What are the reported results of clubfoot treatment (Ponseti technique) in Africa?

The short-term results of clubfoot treatment (Ponseti technique) in Africa are commonly reported through clinical assessment. Different outcome measures are used and report success in 68% - 98% of cases. Long-term results of treatment are inconsistently reported in the literature. There is need for the definition of success after Ponseti management to be standardised. This will assist the monitoring and evaluation of service delivery, allow comparison of results and identify reasons for poor outcome.

Research Question 3: How can the results of Ponseti treatment be monitored and assessed by clubfoot therapists in Africa?

The results of Ponseti treatment can be monitored and assessed using one question about the plantigrade position of the foot answered by physical examination, and three questions answered by the caregiver regarding the child's pain, the child's ability to wear shoes and caregiver satisfaction. It is likely that clubfoot therapists in Africa can use these questions delivered through the ACT tool to assess the success of treatment. The total ACT score

has a high sensitivity and specificity in identifying children who need additional intervention compared to full clinical assessment. Further work is required to evaluate the ACT tool in other clinics and countries, and with other cadres of clubfoot therapists.

Research Question 4: What are the short and long-term results of Ponseti treatment in a cohort of children with clubfoot in Zimbabwe and what factors influence the outcome of treatment?

A case series of children with clubfoot treated in one tertiary hospital in Harare found that 85% of feet achieved a successful short-term result, when success is defined as a Pirani score of 1 or less at the end of the correction phase. After 3.5 - 5 years, 72% of children achieved success (defined as an ACT score of 9 or more). Female sex was the only factor associated with success in short-term outcome, however it was not a factor that influenced long-term outcome. Factors associated with the success of long-term outcome included (a) the child completed casting with a Pirani score of ≤ 1, (b) the child completed casting and was fitted with a FAB, and (c) the child used the FAB for two years or more.

Research Question 5: What indicators are required to assess the functionality of clubfoot clinics in Africa?

The indicators that are required to assess the functionality of clubfoot clinics in Africa were identified as (1) Plaster of paris is available; (2) Essential equipment is available; (3) A completed record for each visit; (4) A functional referral system for tenotomy; (5) The clinic occurs on a specific day; (6) There is a standard treatment protocol available; (7) There is someone in charge (for continuity); (8) There is a process for surgical referrals; (9) There is a process to monitor drop out; and (10) Two or more trained therapists regularly attend the clinic. The FACT was developed to measure these indicators. Further work in other settings in Africa is required to assess the validity of the FACT. The tool also needs to be used in long-term monitoring of clinic services to see if it assists with decision-making and improvement of services.

Research Question 6: What are the strengths and weaknesses of current clubfoot clinics in Zimbabwe; how can the services be improved?
All clubfoot clinics in Zimbabwe had an identifiable person in charge and two or more trained clubfoot therapists in clinics that occurred on a specific day. The most common needs identified related to service provision; they were (a) a standard treatment protocol, (b) a process for surgical referrals, and (c) a process to monitor drop out of patients. The services can be improved through an understanding of individual clinic contexts and a health system-oriented approach to supporting the development of the above protocols and processes.

Research Question 7: What is the feasibility of a training programme for clubfoot therapists in Africa?
A training programme that includes a basic and an advanced course with standardised elements and opportunity for close supervision and mentorship is feasible for clubfoot therapists in Africa. Next steps should include exploring the retention of knowledge and confidence post-training and investigating the short-term and long-term outcomes of children that are treated by the trained clubfoot therapists.

15.3 Implications for future research

Whilst the findings of this research contribute to evidence for improving clubfoot services in Africa, they also demonstrate that gaps remain and present opportunities for future research.

The key questions are:
1. What further adaptions to the theory of change model are required, and which assumptions in the theory of change are realised?

2. Can the ACT tool be used by clubfoot therapists, in other countries and what is the reliability, validity and effectiveness of the tool?

3. Can the FACT be used in other countries and what is the reliability, validity and effectiveness of the tool?

4. What factors are associated with access to clubfoot treatment in Zimbabwe?

5. What factors are associated with adherence to clubfoot treatment in Zimbabwe?

6. To what extent does family and community support continue following the correction phase, and what is the role of this support in adherence to bracing?

15.4 Implications for policy

The National Health Strategy for Zimbabwe (2016 – 2020) 'Equity and quality in health: leaving no one behind' (1) has an objective to 'reduce disability and dependence by 50%.' The strategy laid out plans to improve availability of services, however both baseline (2014) and target (2020) data for the measurement of disability remain to be determined. There is little detail on the resources that will be provided to support services that will reduce disability. Policies that include clubfoot care with programmes for child health may improve awareness and access, and contribute to realisation of these plans. In addition, policies that define roles and expectations of performance for health care workers involved in the task-shifting of clubfoot services may improve the supportive environment for these services. Collaboration within government structures and between NGOs is necessary to identify which part of a clubfoot service works for whom and how, to share learning and to allow extrapolation to other countries.

15.5 Implications for clubfoot programmes

The evidence provided by this research has important implications for the implementation of clubfoot programmes in Africa: clubfoot therapists can be trained to provide treatment for children with clubfoot, they are likely to be able to assess the long-term outcomes of their treatment with the ACT tool and score, and clinic supervisors can use the FACT to assess clubfoot clinic functionality (capability and suitability).

Increasing access to care for children with clubfoot is necessary but not sufficient to ensure disability from clubfoot is eliminated; it must be matched with adherence to treatment and improved quality of care. Continued supervision and mentoring of clubfoot therapists and appropriate counselling of caregivers to promote access and adherence to treatment should be addressed

in parallel. The following recommendations are formulated on the basis of the PhD thesis findings:

1. Use the number of children with clubfoot per million population to estimate need for clubfoot care and to plan and resource services; one clubfoot clinic per million population provides estimates of feasible service delivery at 'district' level and allows extrapolation for national requirements.

2. Trial the use of the ACT score with clubfoot therapists as a monitoring and decision-making tool. Evaluation of the tool for acceptability in different contexts is required.

3. Assess the use and effectiveness of the FACT in long-term monitoring of clinic services for decision-making and improvement of services. Evaluation of acceptability in different countries is required.

4. Given the evidence that only half of expected children with clubfoot access services in the first year of life, it is important to give health education to community health workers about clubfoot treatment, in particular how to counsel parents on the importance of going for treatment early.

5. Factors that influence the success of long-term outcome of treatment include (a) the child completed casting with a Pirani score ≤ 1, (b) the child completed casting and was fitted with a FAB, and (c) the child used a FAB for two years or more. These factors offer direction for strategies to improve long-term outcome.

6. Provide training with the ACT programme and follow up mentorship to improve the skills of clubfoot therapists.

7. Liaise closely with national policy regulators to advocate for clubfoot clinics to be well equipped.

8. Analyse routinely collected data (e.g. input and output data) and provide timely feedback to clubfoot therapists and clinic supervisors.

9. Analyse data to determine which components in the health system-oriented approach to clubfoot care, and in what quantity, are essential for effectiveness. For example, identify the pathways that contribute much of the effect (such as strategies to increase access, quality community mobilisation, adherence support, trained therapists or functioning supply chain) and prioritise these components.

Conclusion

Clubfoot services can be improved in Zimbabwe and probably the wider Africa region. It requires a health system-oriented approach. The evidence presented in this PhD thesis indicates that children with clubfoot can be effectively treated by trained clubfoot therapists (using the Ponseti method). To enable this there is a need to ensure that clubfoot clinics are appropriately equipped and clubfoot therapists are appropriately trained.

The work presented in this PhD thesis sought to address gaps in evidence to improve services for children with clubfoot in Africa. This study highlighted the fundamental unmet need for indicators of success of long-term treatment and clubfoot clinic functionality within a health system. We propose two simple tools to provide data to fill the information gaps for clubfoot services. This research has provided evidence for where to prioritise action. Strengthening multiple pathways of the health system will ensure that all children born with clubfoot can ‘Run Free’.

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SECTION D APPENDICES

Appendix 1 Ethics approval

Appendix one includes the ethics approval for the cohort study and cross-sectional survey in Zimbabwe, and the evaluation of training with the Africa Clubfoot Training project.

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APPROVAL

Ref: MRCZ/B/789 **11 February, 2015**

Tracy Smythe
London School of Hygiene and Tropical Medicine
Keppel Street, London, WC1E7HT
London
C/O Parirenyatwa Hospital, Harare, Zimbabwe

RE:- Zimbabwe's first Ponseti Clubfoot Clinic based at Parirenyatwa Hospital, Harare: a review of the initial two years of the programme.

Thank you for the above titled proposal that you submitted to the Medical Research Council of Zimbabwe (MRCZ) for review. Please be advised that the Medical Research Council of Zimbabwe has **reviewed** and **approved** your application to conduct the above titled study. This is based on the following documents (among others) that were submitted to the MRCZ for review:

a) Research Protocol
b) Informed Consent forms (English and Shona)

- **APPROVAL NUMBER** **: MRCZ/B/789**

This number should be used on all correspondence, consent forms and documents as appropriate.

- **TYPE OF REVIEW** **: EXPEDITED**
- **EFFECTIVE APPROVAL DATE** **: 11 February, 2015**
- **EXPIRATION DATE** **: 10 February, 2016**

After this date, this project may only continue upon renewal. For purposes of renewal, a progress report on a standard form obtainable from the MRCZ Website should be submitted three months before the expiration date for continuing review.

- **SERIOUS ADVERSE EVENT REPORTING**: All serious problems having to do with subject safety must be reported to the Institutional Ethical Review Committee (IERC) as well as the MRCZ within 3 working days using standard forms obtainable from the MRCZ Website.
- **MODIFICATIONS**: Prior MRCZ and IERC approval using standard forms obtainable from the MRCZ Website is required before implementing any changes in the Protocol (including changes in the consent documents).
- **TERMINATION OF STUDY**: On termination of a study, a report has to be submitted to the MRCZ using standard forms obtainable from the MRCZ Website.
- **QUESTIONS**: Please contact the MRCZ on Telephone No. (04) 791792, 791193 or by e-mail on mrcz@mrcz.org.zw
- **Other**
- Please be reminded to send in copies of your research results for our records as well as for Health Research Database.
- You're also encouraged to submit electronic copies of your publications in peer-reviewed journals that may emanate from this study.

Yours Faithfully

[redacted]

.....

MRCZ SECRETARIAT
FOR CHAIRPERSON
MEDICAL RESEARCH COUNCIL OF ZIMBABWE

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PROMOTING THE ETHICAL CONDUCT OF HEALTH RESEARCH

MRCZ – continuing approval

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CONTINUING APPROVAL LETTER

REF: MRCZ/B/789 **11 February, 2016**

Tracey Heather Smythe
London School of Hygiene & Tropical Medicine
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RE: Results of the Ponseti Method for Clubfoot Treatment in Parirenyatwa Hospital, Harare.

Thank you for the application for review of Research Activity that you submitted to the Medical Research Council of Zimbabwe (MRCZ). Please be advised that the Medical Research Council of Zimbabwe has **reviewed**and **approved**your application to continue conducting the above titled study.

This approval is based on the review and approval of the following documents that were submitted to MRCZ for review:-

a) Annual Progress report
b) Study Protocol

- **APPROVAL NUMBER** **: MRCZ/B/789**

This number should be used on all correspondence, consent forms and documents as appropriate.

- **TYPE OF MEETING** **: Expedited**
- **EFFECTIVE APPROVAL DATE** **: 11 February 2016**
- **EXPIRATION DATE** **: 10 February, 2017**

After this date, this project may only continue upon renewal. For purposes of renewal, a progress report on a standard form obtainable from the MRCZ Offices should be submitted three months before the expiration date for continuing review.

- **SERIOUS ADVERSE EVENT REPORTING**: All serious problems having to do with subject safety must be reported to the Institutional Ethical Review Committee (IERC) as well as the MRCZ within 3 working days using standard forms obtainable from the MRCZ Offices or website.
- **MODIFICATIONS**: Prior MRCZ and IERC approval using standard forms obtainable from the MRCZ Offices is required before implementing any changes in the Protocol (including changes in the consent documents).
- **TERMINATION OF STUDY**: On termination of a study, a report has to be submitted to the MRCZ using standard forms obtainable from the MRCZ Offices or website.
- **QUESTIONS**: Please contact the MRCZ on Telephone No. (04) 791792, 791193 or by e-mail on mrcz@mrcz.org.zw

Other

- Please be reminded to send in copies of your research results for our records as well as for Health Research Database.
- You're also encouraged to submit electronic copies of your publications in peer-reviewed journals that may emanate from this study.

Yours Faithfully

[redacted]

MRCZ SECRETARIAT
FOR CHAIRPERSON
MEDICAL RESEARCH COUNCIL OF ZIMBABWE

MEDICAL RESEARCH COUNCIL OF ZIMBABWE
2016 -02- 11
APPROVED
P.O. BOX CY 573 CAUSEWAY, HARARE

PROMOTING THE ETHICAL CONDUCT OF HEALTH RESEARCH

Joint Research Ethics Committee, for the University of Zimbabwe, College of Health Sciences and Parirenyatwa Group of Hospitals (JREC)

Joint Research Ethics Committee For The University of Zimbabwe, College of Health Sciences and Parirenyatwa Group of Hospitals

Parirenyatwa Group of Hospitals

JREC Office No.4, 5th Floor College of Health Sciences Building
Telephone: +263 4 708140/ 791631 Exts 2241/2242
Email: jrec.office@gmail.com/ jrec@medsch.uz.ac.zw,website:www.jrec.uz.ac.zw

University of Zimbabwe College of Health Sciences

APPROVAL LETTER

Date: 20th July 2014 **JREC Ref: 143/14**

Names of Researchers: Miss Tracey Smythe
Address: c/o Parirenyatwa Hospital, Department of Physiotherapy.

Re: Zimbabwe's First Ponseti Clubfoot Clinic: A Retrospective Review of The Initial Two Years.

Thank you for your application for ethical review of the above mentioned research to the Joint Research Ethics Committee. Please be advised that the Joint Research Ethics Committee has reviewed and approved your application to conduct the above named study. You are still required to obtain MRCZ approval and if required by the nature of your study, RCZ approval as well, before you commence the study.

- **APPROVAL NUMBER:** JREC/143/14
- **APPROVAL DATE:** 29th September 2014
- **EXPIRY DATE:** 28th September 2015

This approval is based on the review and approval of the following documents that were submitted to the Joint Ethics Committee:

a) Completed application form
b) Full Study Protocol
c) Informed Consent in English and/or appropriate local language
d) Data collection tool version:

After this date the study may only continue upon renewal. For purposes of renewal please submit a completed renewal form (obtainable from the JREC office) and the following documents before the expiry date:

a. A Progress report
b. A Summary of adverse events.
c. A DSMB report

- **MODIFICATIONS:**

  Prior approval is required before implementing any changes in the protocol including changes in the informed consent.

- **TERMINATION OF STUDY:**

  On termination of the study you are required to submit a completed request for termination form and a summary of the research findings/ results.

Yours faithfully,

[redacted]

Dr C.E Ndhlovu
For JREC Chairman

Telephone: +263-4-722187
Telegraphic Address:
"MEDICUS", Harare
Fax: +263-4-794734
(702293 FHP)
Telex: MEDICUS 22211ZW

ZIMBABWE

Reference:

Ministry of Health and Child Care
P. O. Box CY1122
Causeway
HARARE

13 May 2016

Attention: Tracey Smythe
16 Pendennis Rd
Mt Pleasant
Harare

Re: Request for permission to carry out research to provide evidence for Clubfoot Management

The above subject matter refers:

Your letter of request seeking permission and approval to carry out research on "Club foot Management" in some of our institutions in Zimbabwe is acknowledged.

We authorize you to pursue your project, with the understanding that all clearances with relevant authorities have been effected. We however need to be appraised of the proposed study sites in order to inform the relevant authorities.

The Rehabilitation Unit in the Ministry will be your contact point during your research time. You may feel free to contact them to discuss practical implementation of the research

[redacted]

SECRETARY FOR HEALTH & CHILD CARE
(01) SECRETARY OFFICE (01)
13 MAY 2016
P.O. BOx CY 1122, CAUSEWAY
ZIMBABWE

Brigadier General (Dr) G. Gwinji
Secretary for Health and Child Care

London School of Hygiene & Tropical Medicine – Zimbabwe study

Miss Tracey Smythe
LSHTM

24 May 2016

Dear Tracey

Study Title: Evidence to improve clubfoot services in Africa with emphasis on Zimbabwe as a pilot programme

LSHTM Ethics Ref: 11132

Thank you for responding to the Observational Committee's request for further information on the above research and submitting revised documentation. The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

Approval is dependent on local ethical approval having been received, where relevant.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document Type	File Name	Date	Version
Covering Letter	Response to clarification questions 16th May 2016		
Protocol / Proposal	1. Study proposal Zimbabwe Clubfoot research v1	29/03/2016	1
Protocol / Proposal	2. Interview Guide for clubfoot health care worker v1	29/03/2016	1
Protocol / Proposal	2b.Health System Assessment Questionnaire v1	29/03/2016	1
Protocol / Proposal	3a.Proxy or child PedsQLgeneral wellbeing questions	29/03/2016	1
Protocol / Proposal	3b. Proxy or child PedsQL satisfaction with health system treatment	29/03/2016	1
Investigator CV	Tracey Smythe CV 2016	29/03/2016	1
Information Sheet	5a.Information Sheet and Consent form for Trainees	29/03/2016	1
Information Sheet	5b.Information Sheet and Consent form for hospital interviewee	29/03/2016	1
Information Sheet	5c. Information for Consent to Parents	29/03/2016	1
Local Approval	Zimbabwe programme consent	29/03/2016	1
Information Sheet	5c. Information for Consent to Parents v2 ammended 16 May 2016	16/05/2016	2

After ethical review

The Chief Investigator (CI) or delegate is responsible for informing the ethics committee of any subsequent changes to the application. These must be submitted to the Committee for review using an Amendment form. Amendments must not be initiated before receipt of written favourable opinion from the committee.

The CI or delegate is also required to notify the ethics committee of any protocol violations and/or Suspected Unexpected Serious Adverse Reactions (SUSARs) which occur during the project by submitting a Serious Adverse Event form.

At the end of the study, the CI or delegate must notify the committee using an End of Study form.

All aforementioned forms are available on the ethics online applications website and can only be submitted to the committee via the website at: http://leo.lshtm.ac.uk Additional information is available at: www.lshtm.ac.uk/ethics

Yours sincerely,

[redacted]

Zimbabwe Sustainable Clubfoot Programme

23rd February, 2016

Ms Tracey Smythe
London School of Hygiene and Tropical Medicine
Keppel Street,
London,
WC1E7HT

Dear Tracey,

Re: Evaluation of the Clubfoot Programme in Zimbabwe

Thank you very much for your enthusiasm to do evaluation and research in Zimbabwe with regard to the existing clubfoot programme during 2016/2017.

On behalf of the Zimbabwe Sustainable Clubfoot Programme, we grant permission for you to perform your research and evaluation in all the clinics that have been established by the ZSCP in Zimbabwe as part of your PhD study entitled "Does the introduction of a standardised training programme improve quality of services for children with clubfoot?"

You have our consent to include monitoring and evaluation data, (current, historical and future), collected from the 13 established clinics in Zimbabwe

The ZSCP is funded by Miraclefeet, CURE and ZANE, and is supported by the MOHCC, It would be appreciated if their contribution to the programme can be acknowledged in your study.

Therefore I would be very grateful if you would confirm in writing your acceptance of these terms and conditions, specifically that:

- You will notify the ZSCP and partners of public presentation or publication of work that includes ZSCP programme data, and provide opportunity for review prior to release or publication.
- You will acknowledge the programme funders and partners in publication or dissemination of this work

With many thanks,

Yours sincerely,

[redacted]

Ryan Bathurst
Director: ZSCP

CC: Chipo Harper, Programme Manager, ZSCP
Mrs C Nleya, Deputy Director Rehabilitation Services, MOHCC

London School of Hygiene & Tropical Medicine – ACT project

Miss Tracey Smythe
LSHTM
24 November 2015

Dear Tracey

Study Title: The introduction of a standardised training programme for the management of clubfoot in children: a pilot evaluation of the Africa Clubfoot Training Project at the Cure Hospital, Addis Ababa

LSHTM Ethics Ref: 10412

Thank you for responding to the Observational Committee's request for further information on the above research and submitting revised documentation. The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

Approval is dependent on local ethical approval having been received, where relevant.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document Type	File Name	Date	Version
Protocol / Proposal	1.Study protocol for pilot in Ethiopia Jan 2016	29/10/2015	1
Protocol / Proposal	2. Interview Guide for CURE clubfoot health care worker	29/10/2015	1
Protocol / Proposal	3.ACT PRE COURSE assessment answer form	29/10/2015	1
Protocol / Proposal	4.ACT POST-course form	29/10/2015	1
Protocol / Proposal	5.MCQ-questions for PRE and POST course	29/10/2015	1
Protocol / Proposal	6.Pilot Health System Assessment Questionnaire for clubfoot clinics coordinator	29/10/2015	1
Investigator CV	Tracey Smythe CV 2015	29/10/2015	1
Information Sheet	7.Information Sheet for ACT trainee	29/10/2015	1
Information Sheet	8.Participant Consent Form ACT trainee	29/10/2015	1
Information Sheet	9.Information Sheet for CURE hospital interviewee	29/10/2015	1
Information Sheet	10.Participant Consent Form CURE hospital	29/10/2015	1
Covering Letter	LSHTM Ethics Committee clarification 151120	20/11/2015	1

After ethical review

The Chief Investigator (CI) or delegate is responsible for informing the ethics committee of any subsequent changes to the application. These must be submitted to the Committee for review using an Amendment form. Amendments must not be initiated before receipt of written favourable opinion from the committee.

The CI or delegate is also required to notify the ethics committee of any protocol violations and/or Suspected Unexpected Serious Adverse Reactions (SUSARs) which occur during the project by submitting a Serious Adverse Event form.

At the end of the study, the CI or delegate must notify the committee using an End of Study form.

All aforementioned forms are available on the ethics online applications website and can only be submitted to the committee via the website at: http://leo.lshtm.ac.uk

information is available at: www.lshtm.ac.uk/ethics

Yours sincerely,

[redacted]

Linda Hansen
CURE Clubfoot
Regional Manager - Africa

Beit CURE Hospital of Zambia
Great North Road
PO Box 36961
Lusaka, Zambia
C +260 (0)977 740 164
E linda.hansen@cure.org
cure.org

18th January, 2016

Ms Tracey Smythe
London School of Hygiene and Tropical Medicine
Keppel Street, London, WC1E7HT

Dear Tracey,

Re: Evaluation of Clubfoot Training in Ethiopia

Thank you very much for your assistance with the design and implementation of monitoring and evaluation of the clubfoot training activities related to the Africa Clubfoot Training (ACT) project (March 2015 – February 2017).

On behalf of the ACT partners (University of Oxford, CURE Ethiopia Children's Hospital, CURE Clubfoot programme and Global Clubfoot Initiative), we grant permission for you to include anonymised ACT project monitoring and evaluation data collected in accordance to agreed ACT project protocols as part of your PhD study entitled "Does the introduction of a standardised training programme improve quality of services for children with clubfoot?"

You may include monitoring and evaluation data collected with consent from course participants during clubfoot training courses held in Ethiopia during the ACT Project, and as part of follow-up monitoring and evaluation activities such as follow-up questionnaires and interviews with course participants up to one year after the training event.

The Africa Clubfoot Training project is funded by the UK Department for International Development and, as such, the use or publication of any data collected during the project is subject to the terms and conditions of the funding contract. Therefore I would be very grateful if you would confirm in writing your acceptance of these terms and conditions, specifically that:

- Any of your participation in monitoring and evaluation data collection on behalf of the ACT project will be undertaken in partnership with the ACT team, according to the ACT monitoring and evaluation plan approved by our funders
- You will notify the ACT project partners of public presentation or publication of work that includes ACT project data, and provide opportunity for review prior to submission or presentation.
- You will acknowledge the project funders and partners in publication or dissemination of work that includes ACT project data as per the funders' guidelines on acknowledgements in the main funding contract

With many thanks,

Yours sincerely,

[redacted]

Linda Hansen
CURE Clubfoot – Regional Director, Africa

CC:
Grace Le, ACT Project Manager, University of Oxford
Rosalind Owen, Global Clubfoot Initiative Programme Manager
Richard Gardner, Consultant Orthopaedic Surgeon, CURE Ethiopia Children's Hospital

Appendix 2 Information and consent documents

Appendix two contains the research information sheet and consent documents in Shona and English in the format that is required by the Medical Research Council of Zimbabwe.

Information form and consent – Shona

Page 1 [of 2] IRB No._____

INFORMED CONSENT FORM FOR ADULT CONSENT

London School of Hygiene & Tropical Medicine
Keppel Street, London WC1E 7HT
United Kingdom
Switchboard: +44 (0)20 7636 8636
www.lshtm.ac.uk

LONDON SCHOOL of HYGIENE &TROPICAL MEDICINE

Evidence to Improve Clubfoot Services in Africa with Zimbabwe as a Case Study

Principal Investigator: Tracey Smythe
Supervisor: Allen Foster
Phone number 0785 939 322

Tinoda kukukokai kuti muve nhengo ye avo vanopinda muchidzidzo chiri maererano nemashandiro e clubfoot. Musati maita sarudzo, zvakakosha kuti munzwisise kuti sei tsvakiridzo iyi ichiitwa uye kuti chii chinodiwa. Torai nguva yenyu munyatsoverenga zvinyorwa izvi muchitauriranawo nevamwe kana zvichiita. Mutibvunze kana paine chimwe chisina kunyatsojeka kana kuti paine zvamunoda kutsanangurirwa.

Musoro wenyaya: humboo hwekuvandudza mashandiro e Clubfoot mu Africa.

Ndini ani
Ndinonzi Tracey Smythe, mudzidzi pachikoro chinonzi 'London School of Hygiene and Tropical Medicine'.

Zvatirikuita
Tinoda kunzwisisa kuti kurapwa kwe clubfoot kunoshanda sei mu Zimbabwe. Pakuita izvi, tinodawo kunzwa kuti munoti chii nemarapiro e clubfoot.
Kana mukabvuma, muchakumbirwa kudaira mibvunzo inoenderana nezvamakasangana nazvo kuchipatara che clubfoot uye zvamunofunga pamusoro pegumbo remwana wenyu. Zvehupenyu hwenyu zvinosanganisira zera, kuti muri munhurume kana munhukadzi nekwamunoshanda zvichabvunzwawo. Zita renyu richapiwa imwe nhamba yekudaidzwa nayo. Musati matanga kubvunzwa, muchanyora rugwaro runoratidza kuti mabvuma kupinda muchirongwa ichi. Izvi zvinoitirwa tsvakiridzo yezvidzidzo. Zvinozoratidza kuti munonzwisisa kuti chidzidzo chacho chiri maererano nei uye kuti mungapindawo sei muchidzidzo chacho. Kupinda muchirongwa ichi isarudzo yenyu, uye munogona kungobuda machiri chero nguva zvayo musina kutarisirwa kutaura zvikonzero zvekubuda. Kana mukabuda hamuzobhadhariswi kana kurasikirwa nechimwe chinhu. Kubvunzurudza chichava awa. Muchapandukirwa $3- $5 nokuda kwenyu kufamba mari.

Kuvanzika
Mhinduro dzenyu dzichachengetedzwa mumichina panzvimbo yakachengetedzwa dzoshandiswa izvozvi kana muneramangwana uye zvisingaburitswe kuti dzabva kunani. Kunogona kutanga kwaitwa dzimwe tsvakiridzo uye kupiwa mvumo neve Research Ethics Committee kuti zvinyorwa zvakachengetedzwa zvishandiswe munguva inotevera. Zvinyorwa zvese zvine mazita zvichagara zvakavharirwa munzvimbo yakachengetedzeka, hazvizoonekwi nevamwe vanhu uye zvichagara zvakavanzika sezvinotarisirwa nemutemo. Zvinyorwa zvese zvinobva mumibvunzo yamunobvunzwa zvingangotarisiswa zvakare neavo vane kodzero yekuona kuti tsvakiridzo inoitwa nemazvo. Vanhu vese ava vanotarisirwa kuchengetedza zita renyu rakavanzika.
Kana muine mibvunzo munogona kubvunza chero nguva ipi zvayo.

Zvakaipirei
Parizvino, hationi chakaipa chingaitike nekuda kwekupinda kwenyu muchirongwa ichi. Zvibingamupinyi zvingangosanganikwa nazvo mushure mekupinda kwenyu muchirongwa ichi ndezvinongosanganikwawo nazvo mazuva ose muhupenyu.

Zvakanakirei
Tinovimba kuti chidzidzo ichi chichabatsira mukukurudzira kunzwisisa kurapwa kwe clubfoot mu Zimbabwe.

Tinotenda kutipa nguva yenyu.

 IRB No. _____

Usati kusaina fomu ichi, tapota kubvunza chero mibvunzo iri chero chinhu chine chokuita kudzidza iri kujeka kwauri. Unogona kutora nguva yakawanda sezvinobvira zvakafanira kufunga pamusoro.

1. Ndawana mukana wekufunga nezvakanyorwamo,ndikabvunza mibvunzo uye ndikapindurwa mibvunzo yangu ☐
2. Ndinonzwisisa kuti kupinda muchidzidzo ichi isarudzo yangu uye ndinogona kungobuda chero nguva ipi zvayo ndisina kupa chikonzero. ☐
3. Ndinonzwisisa kuti zvinyorwa zvaunganidzwa kubva muchidzidzo ichi zvingangotariswa nevakuru varipamutemo uye vanemvumo. Ndinopa mvumo kuvanhu ava kuti vapiwe zvinyorwa.. ☐
4. Ndinobvuma kuti mibvunzo yandabvunzwa pamwe nemhinduro dzacho zviiswe mumuchina wekutapa mazwi. ☐
5. Ndinobvuma kuti dzimwe mhinduro dzangu dzishandiswe pakuita dzimwe tsvakiridzo nezvimwewo zvinyorwa zvingada kuitwa. ☐
6. Ndinobvuma kuva nhengo yechidzidzo ichi. ☐

_____ _____

Zita reari kubvunzwa ne Signature Date and Time

_____

Signature reuchapupu

_____ _____

Zita reari kubvunza ne Signature Date and Time

Iwe achapiwa kopi chimiro ichi
Kana muine mibvunzo pamusoro kudzidza ichi muine uye vangada kutaura munhu asiri nhengo tsvakurudzo chikwata, tapota inzwa wakasununguka kuonana Medical Research Council of Zimbabwe on runhare 791792 kana 791193 uye cellphone nhamba 0784 956 128

Information form and consent – English

 IRB No._____

INFORMED CONSENT FORM FOR ADULT CONSENT

London School of Hygiene & Tropical Medicine
Keppel Street, London WC1E 7HT
United Kingdom
Switchboard: +44 (0)20 7636 8636
www.lshtm.ac.uk

LONDON SCHOOL of HYGIENE &TROPICAL MEDICINE

Evidence to Improve Clubfoot Services in Africa with Zimbabwe as a Case Study

Principal Investigator: Tracey Smythe
Supervisor: Allen Foster
Phone number 0785 939 322

We would like to invite you to take part in a study about clubfoot services. Before you decide, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with other people if you wish. Ask us if there is anything that is not clear or if you would like more information.

Title of the study: Evidence to improve clubfoot services in Africa

Who I am
I am Tracey Smythe, a student at the London School of Hygiene and Tropical Medicine.
What we are doing
We would like to understand how treatment of clubfoot in Zimbabwe works. To do this, we would like to hear what you think about clubfoot treatment.
If you agree, you will be asked to answer some questions about your experience at the clubfoot clinic and what you think about your child's foot. Some personal information such as age, gender and employment will also be asked. Your name will be linked with a made up code number. Before the interview starts I will ask you to sign a consent form to show that you have agreed to take part. This is always done for a research study. It will show that you understand what the study is about and how you can take part. It is up to you to decide to join the study. Taking part is completely voluntary and you are free to withdraw at any time without giving a reason. If you do this there will be no penalties and you will not be prejudiced in any way. The interview will be an hour. You will be reimbursed $3-$5 for your travel costs.

Confidentiality
Your answers will be stored electronically in a secure environment and used for research now or at a later date and will not reveal who you are. All future use of the stored data will be subject to further Research Ethics Committee review and approval. All identifying information will be kept in a locked file cabinet, will not be available to others and will be kept confidential to the extent possible by law. The records from your participation may be reviewed by people responsible for making sure that research is done properly. All of these people are required to keep your identification confidential.
If you have any questions you can ask them at any time. You can refuse to answer a question you do not want to answer.

Disadvantages
At the present time, we do not see any risk of harm from your participation. The risks associated with participation in this study are no greater than those encountered in daily life.
Advantages
We hope that this study will be helpful in promoting understanding of clubfoot treatment in Zimbabwe.

Thank you for your time.

 IRB No. _____

Before you sign this form, please ask any questions on any aspect of this study that is unclear to you. You may take as much time as necessary to think it over.

AUTHORIZATION

YOU ARE MAKING A DECISION WHETHER OR NOT TO ALLOW YOUR CHILD TO PARTICIPATE IN THIS STUDY. YOUR SIGNATURE INDICATES THAT YOU HAVE READ AND UNDERSTOOD THE INFORMATION PROVIDED ABOVE, HAVE HAD ALL YOUR QUESTIONS ANSWERED, AND HAVE DECIDED TO ALLOW YOUR CHILD TO PARTICIPATE.

The date you sign this document to enroll your child in this study, that is, today's date, MUST fall between the dates indicated on the approval stamp affixed to each page. These dates indicate that this form is valid when you enroll your child in the study but do not reflect how long your child may participate in the study. Each page of this Informed Consent Form is stamped to indicate the form's validity as approved by the MRCZ.

_____
Name of Child (please print)

_____ _____
Name of Parent (please print) Date and Time

_____
Signature of Parent or legally authorized representative

_____
Relationship to the Participant

_____
Signature of Witness (Optional)

_____ _____
Name (Print) and Signature of Research Staff Date and Time

YOU WILL BE GIVEN A COPY OF THIS CONSENT FORM TO KEEP.

If you have any questions concerning this study or consent form beyond those answered by the investigator, including questions about the research, your rights as a research Participant or research-related injuries; or if you feel that you have been treated unfairly and would like to talk to someone other than a member of the research team, please feel free to contact the Medical Research Council of Zimbabwe on telephone 791792 or 791193 and cellphone number 0784 956 128

Shona caregiver consent - cohort

Page 1 [of 2]

IRB No._____

INFORMED CONSENT FORM FOR PARENTAL CONSENT

London School of Hygiene & Tropical Medicine
Keppel Street, London WC1E 7HT
United Kingdom
Switchboard: +44 (0)20 7636 8636
www.lshtm.ac.uk

LONDON SCHOOL of HYGIENE &TROPICAL MEDICINE

Evidence to Improve Clubfoot Services in Africa with Zimbabwe as a Case Study

Principal Investigator: Tracey Smythe
Supervisor: Allen Foster
Phone number 0785 939 322

Rugwaro rwevabareki rwekutsvaka ruzivo pamusoro penhaurirano

Tinoda kukukokai kuti muve nhengo ye avo vanopinda muchidzidzo chiri maererano nemashandiro e clubfoot. Musati maita sarudzo, zvakakosha kuti munzwisise kuti sei tsvakiridzo iyi ichiitwa uye kuti chii inodiwa. Torai nguva yenyu munyatsoverenga zvinyorwa izvi muchitauriranawo nevamwe kana zvichiita. Mutibvunze kana paine chimwe chisina kunyatsojeka kana kuti paine zvamunoda kutsanangurirwa.

Musoro wenyaya: humboo hwekuvandudza mashandiro e Clubfoot mu Africa.

Ndini ani
Ndinonzi Tracey Smythe, mudzidzi pachikoro chinonzi 'London School of Hygiene and Tropical Medicine'.

Zvatirikuita

Tinoda kunzwisisa kuti kurapwa kwe clubfoot kunoshanda sei mu Zimbabwe. Pakuita izvi, tinodawo kunzwa kuti munoti chii nemarapiro e clubfoot.

Kana mukabvuma, muchakumbirwa kudaira mibvunzo inoenderana nezvamakasangana nazvo kuchipatara che clubfoot uye zvamunofunga pamusoro pegumbo remwana wenyu. Zvehupenyu hwenyu vinosanganisira zera, kuti muri munhurume kana munhukadzi nekwamunoshanda zvichabvunzwawo. Zita renyu richapiwa imwe nhamba yekudaidzwa nayo. Musati matanga kubvunzwa, muchanyora rugwaro runoratidza kuti mabvuma kupinda muchirongwa ichi. Izvi zvinoitirwa tsvakiridzo yezvidzidzo. Zvinozoratidza kuti munonzwisisa kuti chidzidzo chacho chiri maererano nei uye kuti mungapindawo sei muchidzidzo chacho. Kupinda muchirongwa ichi isarudzo yenyu, uye munogona kungobuda machiri chero nguva zvayo musina kutarisirwa kutaura zvikonzero zvekubuda. Kana mukabuda hamuzobhadhariswi kana kurasikirwa nechimwe chinhu.

Kuvanzika
Mhinduro dzenyu dzichachengetedzwa mumichina panzvimbo yakachengetedzwa dzoshandiswa izvozvi kana muneramangwana uye zvisingaburitswe kuti dzabva kunani. Kunogona kutanga kwaitwa dzimwe tsvakiridzo uye kupiwa mvumo neve Research Ethics Committee kuti zvinyorwa zvakachengetedzwa zvishandiswe munguva inotevera. Zvinyorwa zvese zvine mazita zvichagara zvakavharirwa munzvimbo yakachengetedzeka, hazvizoonekwi nevamwe vanhu uye zvichagara zvakavanzika sezvinotarisirwa nemutemo. Zvinyorwa zvese zvinobva mumibvunzo yamunobvunzwa zvingangotarisiswa zvakare neavo vane kodzero yekuona kuti tsvakiridzo inoitwa nemazvo. Vanhu vese ava vanotarisirwa kuchengetedza zita renyu rakavanzika.

Kana muine mibvunzo munogona kubvunza chero nguva ipi zvayo.

Zvakaipirei
Parizvino, hationi chakaipa chingaitike nekuda kwekupinda kwenyu muchirongwa ichi. Zvibingamupinyi zvingangosanganikwa nazvo mushure mekupinda kwenyu muchirongwa ichi ndezvinongosanganikwawo nazvo mazuva ose muhupenyu.

Zvakanakirei
Hapana chouviri chamunowana pakupinda muchirongwa ichi. Zvichakadaro, tinovimba kuti chidzidzo ichi chichabatsira mukukurudzira kunzwisisa kurapwa kwe clubfoot mu Zimbabwe.

Tinotenda kutipa nguva yenyu.

English caregiver consent – cohort

 IRB No._____

INFORMED CONSENT FORM FOR PARENTAL CONSENT

London School of Hygiene & Tropical Medicine
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LONDON SCHOOL of HYGIENE &TROPICAL MEDICINE

Evidence to Improve Clubfoot Services in Africa with Zimbabwe as a Case Study

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Supervisor: Allen Foster
Phone number 0785 939 322

What you should know about this research study:

- We give you this consent so that you may read about the purpose, risks, and benefits of this research study.
- The main goal of research studies is to gain knowledge that may help future patients.
- You have the right to refuse to allow your child to take part, or agree for your child to take part now and change your mind later.
- Whatever you decide, it will not affect your child's regular care.
- Please review this consent form carefully. Ask any questions before you make a decision.
- Your choice to allow your child to participate is voluntary.

PURPOSE

You are being asked to allow your child to participate in a research study of clubfoot management. The purpose of the study is to understand how treatment of clubfoot in Zimbabwe works. Your child was selected as a possible participant in this study because your child has had clubfoot treatment.

PROCEDURES AND DURATION

If you decide to allow your child to participate, your child will undergo one assessment of their feet for approximately one hour. It will be similar to the assessments your child had in the clubfoot clinic.

RISKS AND DISCOMFORTS

At the present time, we do not see any risk of harm from participation. The risks associated with participation in this study are no greater than those encountered in daily life.

BENEFITS AND/OR COMPENSATION

We hope that this study will be helpful in promoting understanding of clubfoot treatment in Zimbabwe. Reimbursement for your transport to and from the clinic with your child will be provided ($3-$5 in total).

CONFIDENTIALITY

If you indicate your willingness for your child to participate in this study by signing this form, we plan to let the Ministry of Health and Child Care and the Zimbabwe Sustainable Clubfoot Programme know the results. Any information that can be identified with your child will remain confidential and will be disclosed only with your, and when appropriate, your child's permission. Under some circumstances, the MRCZ and the local Institutional Review Board may need to review patient records for compliance audits. Identification will be kept confidential.

IF YOU HAVE QUESTIONS

If you have questions, contact Tracey Smythe on 0785 939 322

VOLUNTARY PARTICIPATION

Participation in this study is voluntary. If you decide not to allow your child to participate in this study, your decision will not affect your or your child's future relations with the clinic. If you decide to allow your child to participate, you and your child are free to withdraw your consent and assent and discontinue participation at any time without penalty.

 IRB No. _____

Before you sign this form, please ask any questions on any aspect of this study that is unclear to you. You may take as much time as necessary to think it over.

AUTHORIZATION

YOU ARE MAKING A DECISION WHETHER OR NOT TO ALLOW YOUR CHILD TO PARTICIPATE IN THIS STUDY. YOUR SIGNATURE INDICATES THAT YOU HAVE READ AND UNDERSTOOD THE INFORMATION PROVIDED ABOVE, HAVE HAD ALL YOUR QUESTIONS ANSWERED, AND HAVE DECIDED TO ALLOW YOUR CHILD TO PARTICIPATE.

The date you sign this document to enroll your child in this study, that is, today's date, MUST fall between the dates indicated on the approval stamp affixed to each page. These dates indicate that this form is valid when you enroll your child in the study but do not reflect how long your child may participate in the study. Each page of this Informed Consent Form is stamped to indicate the form's validity as approved by the MRCZ.

_____
Name of Child (please print)

_____ _____
Name of Parent (please print) Date and Time

_____
Signature of Parent or legally authorized representative

_____
Relationship to the Participant

_____
Signature of Witness (Optional)

_____ _____
Name (Print) and Signature of Research Staff Date and Time

YOU WILL BE GIVEN A COPY OF THIS CONSENT FORM TO KEEP.

If you have any questions concerning this study or consent form beyond those answered by the investigator, including questions about the research, your rights as a research Participant or research-related injuries; or if you feel that you have been treated unfairly and would like to talk to someone other than a member of the research team, please feel free to contact the Medical Research Council of Zimbabwe on telephone 791792 or 791193 and cellphone number 0784 956 128

Participant consent – ACT project

ACT BASIC PRE-COURSE Participant Form

Welcome to the ACT Basic Clubfoot Treatment Provider Course.

1. Enrolment

Please complete using CAPITAL letters

First Name:	
Surname/Family Name):	
Address:	
Email address:	
Mobile number including code:	
Age:	
Gender:	Male / Female
Course Location:	
Job title:	
Specialty:	
Name and address of place of work:	

2. Consent

We hope you enjoy this course and that what you learn will be of use to you in the future wherever you work.

In order to assess how relevant the course material is for your situation, at the start and end of the course we will ask you to answer a few questions and to complete a short assessment. Your answers will remain confidential and will not affect your being given a course attendance certificate.

Your name, address, telephone and email details will be kept by ACT only for the purposes of this course follow up. They will not be given to any third party without your consent.

We will take photos during the course and follow up visits which maybe used in an end of course report, or on our websites and other publications to promote the clubfoot training.

If you are happy to take part in the course and the follow up, please signbelow.

I give my consent to participate in the course and the follow up described above. I understand that I can withdraw my consent at any time without giving a reason, and still attend the course.

Signature:_____Date: _____

Appendix 3 Data collection forms

Appendix three contains the forms that were used to collect data in the cohort, and the cross-sectional study. Data were collected on paper forms.

Pilot Clubfoot assessment form

Pilot Clubfoot Assessment and Parent reported outcomes

Tick when attached:
Heathcare satisfaction ☐
Quality of Life Questionnaire ☐
Roye Score ☐
Bangla Score ☐
Completed:
Physical assessment ☐
Photograph ☐

DATE:_____

Patient Name:_____ PID:_____

DOB: _____ Age:_____

Parent/Guardian_____ Phone _____

Address_____

Date of last attendance at clubfoot clinic:_____

Reason for appointment: _____

Ever Fitted for braces: Yes ☐ No ☐
Number of years braces worn: <1 year ☐ 1-2 years ☐ 2-3years ☐
3-4 years ☐ 4-5 years ☐

General Questions
Where could improvements in your experience of clubfoot management be made?
Referral to the clubfoot clinic Yes ☐ No ☐
The first clinic appointment Yes ☐ No ☐
Information to take home Yes ☐ No ☐
Time to ask questions of clubfoot clinic staff Yes ☐ No ☐
Schedule of appointments (day/hour) Yes ☐ No ☐
Information about tenotomy Yes ☐ No ☐
The tenotomy procedure Yes ☐ No ☐
Follow up after tenotomy Yes ☐ No ☐
Brace fitting Yes ☐ No ☐
Brace reviews Yes ☐ No ☐

Other_____

1

Pilot Clubfoot Assessment and Parent reported outcomes

Many parents and children have difficulties with bracing. Please could you tell us what problems you found, or what was difficult for you. What was easy?

_____

_____

_____

B. Functional Activities	Yes	Not fully/with assistance	No	Comment on quality
6. Squatting				
7. Walking				
8. Running				
9. Up/down steps				
C. Clinical examination	Valgus	Straight	Varus	
10. Heel position - L				
Heel position - R				
	Fixed	Mobile	None	
11. Supination/Cavus - L				
Supination/ Cavus - R				
	> 0 dorsiflexion	0 / 90 degrees	< 0 dorsiflexion	
12. Ankle range - L				
Ankle range - R				

Further comments on quality of function/ROM/ Clinical measurements

_____

_____

_____

_____

_____

_____

Clubfoot assessment form

Clubfoot Assessment PID: DATE:

Tick when attached:
Heathcare satisfaction ☐
Quality of Life Questionnaire ☐
Completed:
Physical assessment ☐
Photograph ☐

DATE:_____

PID:_____

DOB: _____ Age:_____

Parent/Guardian_____ Phone _____

Address_____

Date of last attendance at clubfoot clinic:_____

Reason for appointment: _____

Ever Fitted for braces: Yes ☐ No ☐

Number of years braces worn: <1 year ☐ 1-2 years ☐ 2-3years ☐
3-4 years ☐ 4-5 years ☐

Many parents and children have difficulties with bracing. Please could you tell us what problems you found, or what was difficult for you. What was easy?

_____
_____
_____
_____

Clubfoot Assessment PID: DATE:

Roye Score

1 **How satisfied are you with the status of your child's foot?**

Very satisfied	Somewhat satisfied	Somewhat dissatisfied	Very dissatisfied

2 **How satisfied are you with the appearance of your child's foot?**

Very satisfied	Somewhat satisfied	Somewhat dissatisfied	Very dissatisfied

3 **How often is your child teased because of his or her clubfoot?**

Never	Sometimes	Usually	Always

4 **How often does your child have problems finding shoes that fit?**

Never	Sometimes	Usually	Always

5 **How often does your child have problems finding shoes he or she likes?**

Never	Sometimes	Usually	Always

6 **Does your child complain of pain in his or her affected foot?**

Yes	No

7 **How limited is your child in his or her ability to walk?**

Not at all limited	Somewhat limited	Moderately limited	Very limited

8 **How limited is your child in his or her ability to run?**

Not at all limited	Somewhat limited	Moderately limited	Very limited

9 **How much does your child complain of pain during heavy exercise?**

Never	Sometimes	Usually	Always

10 **How much does your child complain of pain during moderate exercise?**

Never	Sometimes	Usually	Always

Bangla Score

A. Parental rating	Yes	Don't know	No
1. Are you happy with your child's feet?			
2. Would you recommend this program to others?			
3. Does your child play with other children?			
4. Does your child wear shoes of your/their choice?			
5. Does your child have painful feet?			

2

Clubfoot Assessment PID: DATE:

B. Functional Activities	Yes	Not fully/with assistance	No	Comment on quality
6. Squatting				
7. Walking				
8. Running				
9. Up/down steps				
C. Clinical examination	Valgus	Straight	Varus	
10. Heel position – L				
Heel position – R				
	> 0 dorsiflexion	0 / 90 degrees	< 0 dorsiflexion	
11. Ankle range – L				
Ankle range – R				

Relapse

Description (Grade)	L	R
Reduced ankle DF (<15 to 0) (1A)		
Fixed ankle equinus <0 (2A)		
Dynamic supination, flexible (1B)		
Fixed adduction of forefoot (2B)		
Two or more deformities (3)		

Clubfoot score

Score	The foot is plantigrade	Does your child complain of pain in their affected foot?	Can your child wear shoes of your/their choice?	How satisfied are you with your child's foot?
1	Less than plantigrade with add,cavus, varus	Yes and it **often** limits their activity	Never	Very dissatisfied
2	Less than plantigrade	Yes and it **sometimes** limits their activity	Sometimes	Somewhat dissatisfied
3	Plantigrade	Yes but it **does not** limit their activity	Usually	Somewhat satisfied
4	More than plantigrade	No	Always	Very satisfied

Shona Peds QL

*In the past **ONE month**, how much of a **problem** has your child had with …*

Pamusoro pemuviri wake *(problems with…)*	**Haana**	**dzimwe nguva**	**Kazhinji**	**Dzaka-wanda**	**Nguva dzose**
1. zvakandiomera kufamba chinhambwe chakareba	0	1	2	3	4
2. kumhanya	0	1	2	3	4
3. kutamba mitambo	0	1	2	3	4
4. kusimudza zvinhu zvinorema	0	1	2	3	4
5. kuita mabasa emumba	0	1	2	3	4

Pamusoro pekunzwa kwake *(problems with…)*	**Haana**	**dzimwe nguva**	**Kazhinji**	**Dzaka-wanda**	**Nguva dzose**
1.kunzwa kutya	0	1	2	3	4
2. kunzwa kutsamwa	0	1	2	3	4
3. kunzwa hasha	0	1	2	3	4
4. kutya zvinhu zvichaitika kwaari	0	1	2	3	4

Pamusoro pekuva navamwe *(problems with…)*	**Haana**	**dzimwe nguva**	**Kazhinji**	**Dzaka-wanda**	**Nguva dzose**
1. kutamba nevamwe zvakanaka	0	1	2	3	4
2. vamwe vana vanosarudza kusatamba naye	0	1	2	3	4
3. kutukwa navamwe	0	1	2	3	4

Pamusoro pechikoro *(problems with…)*	**Haana**	**dzimwe nguva**	**Kazhinji**	**Dzaka-wanda**	**Nguva dzose**
1. kunzwisisa zvirikudzidziswa	0	1	2	3	4
2. kukanganwa zvinhu zvaadzidza	0	1	2	3	4
3. kurongeka pabasa rake rechikoro	0	1	2	3	4

English Peds QL

In the past ONE month, how much of a problem has your child had with …

PHYSICAL FUNCTIONING *(problems with…)*	Never	Almost never	Some-times	Often	Almost always
1. Walking 100meters	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activites or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Doing chores, like picking up his or her toys	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with…)*	Never	Almost never	Some-times	Often	Almost always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost never	Some-times	Often	Almost always
1. Getting on with other children	0	1	2	3	4
2. Other children not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other children	0	1	2	3	4

SCHOOL FUNCTIONING *(problems with…)*	Never	Almost never	Some-times	Often	Almost always
1.Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3.Keeping up with school activities	0	1	2	3	4

Munofadzwa zvakadii neizvi…

Ruzivo	**Haana**	**dzimwe nguva**	**Kazhinji**	**Dzaka-wanda nguva**	**Nguva dzose**	**Hazvi-goneke**
1.Makapiwa ruzivo rwakawanda sei maringe nechirwere chakawanikwa chine mwana wenyu	0	1	2	3	4	N/A
2. Makapiwa ruzivo rwakawanda sei maringe nekurapwa nemamiriro ehutano hwemwana wenyu	0	1	2	3	4	N/A
3.Munopiwa ruzivo rwakadii maringe nezvingakanganiswe nemishonga yekurapa mwana wenyu	0	1	2	3	4	N/A
4.Mukawana nekukasika zvakadii zvakabuda mukuongororwa kwakaitwa mwana wenyu	0	1	2	3	4	N/A
5.Munoudzwa nezve hutano hwemwana wenyu mushure menguva yakareba sei	0	1	2	3	4	N/A

Kusanganisira vemhuri	**Haana**	**dzimwe nguva**	**Kazhinji**	**Dzaka-wanda nguva**	**Nguva dzose**	**Hazvi-goneke**
1.Mwoyo murefu wamakaratidzwa imi pamwe nevemhuri yenyu pairapwa mwana wenyu.	0	1	2	3	4	N/A
2.Chido chekupindura mibvunzo yamuinayo imi nevemhuri yenyu	0	1	2	3	4	N/A
3.Kurudziro yekuti imi nevemhuri yenyu mupinde munhaurirano pamusoro pemachengeterwo emwana wenyu, uye kupiwa ruzivo maringe nemamiriro ehutano hwemwana wenyu	0	1	2	3	4	N/A
4.Makapiwa mukana wakareba sei wekubvunza mibvunzo maererano nehurwere hwemwana wenyu pamwe nemarapirwo acho.	0	1	2	3	4	N/A

Healthcare satisfaction

PID: DATE:

KUTAURIRANA	Haana	dzimwe nguva	Kazhinji	Dzaka-wanda nguva	Nguva dzose	Hazvi-goneke
1.Vashandi vakatsanangura zvakanaka sei mamiriro ehutano nekurapwa kwemwana wenyu nenzira yaakanzwisisa	0	1	2	3	4	N/A
2.Nguva yakatorwa kutsanangurwa mamiriro ehutano hwemwana wenyu nemarapirwo acho **kwamuri** nenzira yamakanzwisisa	0	1	2	3	4	N/A
3.Vashandi vaiterera nekunzwisisa zvichemo zvenyu here	0	1	2	3	4	N/A
4.Gadziriro yemakapiwa nezvekutarisira pazvinhanho zvese zvekurapwa kwemwana	0	1	2	3	4	N/A
5.Gadziriro yakapiwa **mwana wenyu** maringe nezvinhanho zvese zvekurapwa kwake.	0	1	2	3	4	N/A

MASHANDIRO AKANAKA	Haana	dzimwe nguva	Kazhinji	Dzaka-wanda nguva	Nguva dzose	Hazvi-goneke
1.Vashandi vanobata mwana zvakanaka sei	0	1	2	3	4	N/A
2.kuyedza kwavo kuti mwana wenyu agadzikana uye marwadzo ake aderere	0	1	2	3	4	N/A
3.vakatora nguva yakadii kubatsira pakudzoka kwemwana wenyu kumba	0	1	2	3	4	N/A

ZVITSVAKO ZVEMWOYO	Haana	dzimwe nguva	Kazhinji	Dzaka-wanda nguva	Nguva dzose	Hazvi-goneke
1.Nguva inopiwa mwana yekutamba,kutaura manzwiro ake uye nemibvunzo yaangangova nayo	0	1	2	3	4	N/A
2.Nguva inotorwa kubatsira mwana wenyu kudzokera kuchikoro	0	1	2	3	4	N/A
3.Nguva inotorwa kuchitariswa zvitsvago **zvemwoyo wemwana wenyu**	0	1	2	3	4	N/A
4.Nguva inotorwa kuchitariswa zvitsvago zvemwoyo **wenyu**	0	1	2	3	4	N/A

KUGUTSIKANA PANE ZVESE	Haana	dzimwe nguva	Kazhinji	Dzaka-wanda nguva	Nguva dzose	Hazvi-goneke
1.Zvese zvirikuitwa pakuchengeta mwana wenyu	0	1	2	3	4	N/A
2.Vashandi vakasununguka uye vanobatsira zvakadii	0	1	2	3	4	N/A
3.Nzira mwana wenyu yaanobatwa nayo kuchipatara	0	1	2	3	4	N/A

English Healthcare satisfaction PedsQL

PedsQL 2

How happy are you with...

INFORMATION	Never	Some-times	Often	Almost Always	Always	Not Appli
1. How much information was provided to you about your child's diagnosis	0	1	2	3	4	N/A
2. How much information was provided to you about the treatment and course of your child's health condition	0	1	2	3	4	N/A
3. How much information was provided to you about the side effects of your child's treatment	0	1	2	3	4	N/A
4. How soon information was given to you about your child's test results	0	1	2	3	4	N/A
5. How often you are updated about your child's health	0	1	2	3	4	N/A

INCLUSION OF FAMILY	Never	Some-times	Often	Almost Always	Always	Not Appli
1. The sensitivity shown to you and your family during your child's treatment	0	1	2	3	4	N/A
2. The willingness to answer questions that you and your family may have	0	1	2	3	4	N/A
3. The effort to include your family in discussion of your child's care and other information about your child's health condition	0	1	2	3	4	N/A
4. How much time the staff gave you to ask any questions you may have had about your child's health condition and treatment	0	1	2	3	4	N/A

COMMUNICATION	Never	Some-times	Often	Almost Always	Always	Not Appli
1. How well the staff explained your child's health condition and treatment to **your child** in a way that she/he could understand	0	1	2	3	4	N/A
2. The time taken to explain your child's health condition and treatment to **you** in a way you could understand	0	1	2	3	4	N/A
3. How well the staff listens to you and your concerns	0	1	2	3	4	N/A
4. The preparation provided for **you** about what to expect during tests and procedures	0	1	2	3	4	N/A
5. The preparation provided for **your child** about what to expect during tests and procedures	0	1	2	3	4	N/A

How happy are you with...

TECHNICAL SKILLS	Never	Some-times	Often	Almost Always	Always	Not Appli
1. How well the staff responds to your child's needs	0	1	2	3	4	N/A
2. Efforts to keep your child comfortable and as pain-free as possible	0	1	2	3	4	N/A
3. How much time the staff took to help you with your child coming back home	0	1	2	3	4	N/A

EMOTIONAL NEEDS	Never	Some-times	Often	Almost Always	Always	Not Appli
1. The amount of time given to your child to play, talk about her/his feelings, and any questions she/he may have	0	1	2	3	4	N/A
2. The amount of time spent helping your child with going back to school	0	1	2	3	4	N/A
3. The amount of time spent attending to **your child's** emotional needs	0	1	2	3	4	N/A
4. The amount of time spent attending to **your** emotional needs	0	1	2	3	4	N/A

OVERALL SATISFACTION	Never	Some-times	Often	Almost Always	Always	Not Appli
1. The overall care your child is receiving	0	1	2	3	4	N/A
2. How friendly and helpful the staff is	0	1	2	3	4	N/A
3. The way your child is treated at the hospital	0	1	2	3	4	N/A

Cross-sectional study

Domain	Criteria	Rating				Score
		0	1	2	3	
Leadership	There is an identifiable person in charge of the clinic	In the past 8 weeks:				
		There was no identifiable person in charge of the clinics	There was an identifiable person in charge of 5 or fewer clinics	There was an identifiable person in charge of 6-7 clinics	There was an identifieable person in charge of every clinic	
Human resources	There are regularly two or more Ponseti trained health care workers (HCW) available at each clinic	In the past 8 weeks:				
		There were fewer than 2 trained HCWs available at every clinic	There were 2 or more trained HCWs available in 5 or fewer clinics	There were 2 of more trained HCWs available in 6 -7 clinics	There were 2 or more trained HCWs available at every clinic	
Essential medical equipment	There is always plaster of paris available	In the past 8 weeks:				
		Plaster of paris was not available for 3 or more clinics	Plaster of paris was not available for 2 clinics	Plaster of paris was not available for one clinic	There was always plaster of paris available	
	There is always equipment (such as bucket and scissors/blade, tenotomy set, correct size braces) available	In the past 8 weeks:				
		A bucket, scissors/blade, tenotomy set or correct size braces were not available for 3 or more clinics	A bucket, scissors/blade, tenotomy set or correct size braces were not available for 2 clinics	A bucket, scissors/blade, tenotomy set or correct size braces were not available for one clinic	There was always a bucket and scissors/blade, tenotomy set, correct size braces available	
Service delivery	There is a standard treatment protocol	There is no standard protocol for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot or non-idiopathic clubfoot	There is a standard verbal treatment protocol but nothing in writing for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot and non-idiopathic clubfoot	There is a written protocol for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot and non-idiopathic clubfoot but it is not consistently used	There is a standard written protocol for treatment of idiopathic clubfoot in children under 2 years, the older child with clubfoot and non-idiopathic clubfoot and it is followed consistently	
	There is a functioning referral system for tenotomy	In the 2 most recent tenotomy cases:				
		There was no clinician to whom children requiring a tenotomy could be referred	The children were referred for tenotomy and there is no record of the outcome	The children were referred but the tenotomy was not completed as anticipated	The children were referred for tenotomy and it was completed as anticipated	
	There is a process for surgical referrals	In the 2 most recent cases who required surgery:				
		There was no clinician to whom children requiring a surgical review could be referred	The children were referred and there is no record of the outcome	The children were referred but the surgery was not completed as anticipated	The children were referred for surgery and it was completed as anticipated	
	There is a process to monitor drop out of patients	There is no process to monitor drop out of patients	There is a verbal process to monitor drop out of patients but nothing in writing	There is a written protocol to monitor drop out of patients but it is not consistently followed	There is a written protocol to monitor dop out of patients and it is consistently followed	
	The clinic occurs on a specific day, at minimum weekly	In the past 8 weeks:				
		The clinic did not have an identified day for clubfoot treatment	The clinic has been held on the identified day/s for less than 6 of the weeks	The clinic has been held on the identified day/s for 6-7 of the weeks	The clinic has been held on the identified day/s every week	
Health information system	There is a completed clinic record for each patient visit	In the last 10 clinic records:				
		5 or less of the clinic records were filled in completely	6-7 of the records were filled in completely	8-9 of the clinic records were filled in completely	All of the clinic records were filled in completely	
Total score						