The impact of socio-economic factors on parental non-adherence to the Ponseti protocol for clubfoot treatment in low- and middle-income countries: A scoping review

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Summary

Background The Ponseti treatment is considered the gold standard for clubfoot globally, but requires strong engagement from parents. The aim of this review is to assess the impact of socio-economic factors on the presence of drop-out, relapse or non-compliance during Ponseti treatment in low and middle-income countries (LMICs).

Methods This scoping review includes all articles available from inception until 4.4.2022. All articles describing an association between one or more socio-economic factors and one or more adverse outcomes during the Ponseti treatment in an LMICs were considered for inclusion. Studies were identified by searching Medline/PubMed, Embase, Global Health and Global Index Medicus. Data extraction was done using Covidence extraction 2.0 by two independent reviewers.

Findings A total of 281 unique references were retrieved from the database searches, 59 abstracts were retained for full-text review, of which 19 studies were included in the final review. We grouped the identified socio-economic factors into 4 larger themes: poverty and physical accessibility of clubfoot clinics, presence of support systems, educational level of the parents, and household-level factors and cultural norms. Reduced access to care for girls was considered an important risk factor in South Asia and the Caribbean. Lack of family and community support was an issue raised more often in studies from Eastern Africa. The extreme heterogeneity among collected variables within a small sample of papers made it not possible to perform a meta-analysis.

Interpretation The identified factors are very similar to the socio-economic factors identified in studies looking at the barriers parents and children face when seeking care initially. Poverty was identified as a cross-cutting risk factor in all 4 domains and the most important socio-economic risk factor based on this review, reconfirming poverty eradication as the challenge for the 21st century.

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Introduction

Idiopathic clubfoot, or congenital talipes equinovarus (CTEV), is a congenital malformation in which the foot is twisted internally and eventually the child will walk...
on the upper part of the foot instead of the sole. It is the most common musculoskeletal congenital disease in need of intensive orthopedic treatment. About 175,000 children or 1/1000 alive born infants are born annually with clubfoot, 1–7 with a majority born in countries with weak health systems. Untreated, it can lead to lifelong impairment, inability to walk, social exclusion, and stigma.6,8,10

The Ponseti treatment protocol consists of a casting phase and a bracing phase. The casting phase includes weekly cast changes to remold the foot into a normal position and has an average duration of about 3 months. The bracing phase includes full-time brace wearing during the first 3 months after the casting cycle. Continued nighttime brace-wearing until the child reaches 4 years of age is required, or for one year after achievement of full correction for children who started wearing the braces at an older age. Children still must attend follow-up consultations regularly during the bracing phase to evaluate the child’s feet for a potential relapse of the deformity and to adapt the brace to the growing child and its growing feet. The Ponseti method has become the gold standard accepted treatment for clubfoot.4,11,12

Evidence before this study

This review aims to give an overview of the available evidence and identify potential knowledge gaps. From inception to 4/4/2022 we searched Medline/PubMed, Embase, Global Health, and Global Index Medicus for available articles. All articles describing an association between one or more socio-economic factors and one or more adverse outcomes during the Ponseti treatment in low middle income countries (LMICs) were considered for inclusion. Socio-economic factors all fell into 4 larger themes: poverty and physical accessibility of clubfoot clinics, presence of support systems, educational level of the parents, and household-level factors and cultural norms. The extreme heterogeneity among collected variables within a small sample of papers made it impossible to run a meta-analysis.

Added value of this study

To our knowledge this is the first review that aims to assess the relationship between socio-economic factors and their impact on drop-out, relapse and non-compliance with the Ponseti method throughout the entire course of the treatment. Additionally, the WHO framework on adherence to long-term therapies, has not been used to assess barriers to adherence to non-drug-based therapies like the Ponseti treatment or other musculoskeletal conditions requiring long-term treatment. Socio-economic factors are one out five factors identified in the framework that influence adherence.

Implications of all the available evidence

Our study provides novel objective evidence on poverty eradication has a great influence on parental non-adherence to Ponseti treatment protocol in LMICs. In addition to that the WHO framework for adherence to long-term therapies provides a new way to identify areas in need of research and/or policy change in clubfoot treatment. Non-adherence to treatment cannot be addressed by solely addressing socio-economic factors. All 5 aspects of the framework will need to be addressed in future programs, policies and research studies.
in current intake, follow-up and clinical surveillance programs and perhaps modified to improve adherence to treatment protocols and improve outcomes.

**Methods**

Search strategy — A research protocol was written by MP and approved by the rest of the team. The protocol is not available in a public repository, but a concise summary is provided here. Studies that discuss the Ponseti method in low- and middle-income countries were identified by searching Medline/PubMed (National Library of Medicine, NCBI), Embase (Elsevier, embase.com), Global Health (C.A.B. International, Ebsco), and Global Index Medicus (World Health Organization, https://www.globalindexmedicus.net/) until 4/4/2022. Controlled vocabulary terms (i.e. MeSH, Emtree) were included when available and appropriate. No publication date or languages limits were applied. The search strategies were designed and executed by a librarian (CM). The exact search terms used for each of the databases are provided in the supplementary document.

Inclusion/exclusion criteria — All articles describing an association between one or more socio-economic factors and drop-out, non-compliance or relapse during the Ponseti treatment in an LMIC were considered for inclusion. Both qualitative and quantitative papers were taken into consideration for inclusion. Grey literature and unpublished studies were not included; authors were not contacted for additional information. Abstracts and articles not available in English were translated using Google Translate. Given that a scoping review design offers more liberty to the included and excluded abstracts and full-texts as well as the extracted data from the included papers in this review.

Outcome variables - Non-compliance is defined as non-adherence to the prescribed treatment regimen, including brace-wearing, or as infrequent participation in health visits.37 Drop-out is defined as quitting completely with the proposed therapeutic regimen, and can be considered as an extreme form of non-compliance. Relapse is defined as the reappearance of deformity in a previously fully corrected foot.38 A socio-economic factor is defined as “non-medical factors that influence health outcomes.”26 An LMIC is defined as a country listed as low-, lower-middle- or upper-middle-income according to the World Bank income-level ranking.31

Presentation of results — Qualitative results are reported in a uniform way. The same description is used to describe the same theme or factor throughout the reporting, irrespective of the wording used in the respective paper. This is done to increase clarity of the reported results and to better understand patterns and importance of certain reported factors. No assessment of the strength of the association of qualitative data is made. Quantitative data is reported in the way the data was reported initially in the respective article. No summary statistics are calculated or reported because of the limited number of quantitative articles available and the high heterogeneity of the reported data and statistics. The outline of this paper follows the PRISMA guidelines.

Assessment of bias - The COREQ-checklist for appraisal of qualitative research32 was used to evaluate potential selection bias or analysis bias in the qualitative studies. For the quantitative studies, the checklists from the Joanna Briggs Institute (JBI)33 for critical appraisal of cross-sectional and cohort studies was used to evaluate for potential bias. Bias assessment was done by 2 independent reviewers (MP and AV). All studies were assessed for 3 types of bias: selection bias, information bias and analysis bias. In case of disagreement on whether a type of bias was present in a study or if the measures taken by the researchers were sufficient to address the bias, the paper was discussed between both reviewers to reach consensus.

Level of evidence — The quality and associated level of evidence of reported data was assessed using the American Academy of orthopedic Surgeons (AAOS) evidence-based guidelines for level of evidence in orthopedic surgery research.34 All qualitative studies were labeled as “level V — expert opinion”.

Role of the funding source

There was no funding source for this study.

**Results**

A total of 294 references were retrieved on 4/4/2022 from the database searches. Of these, 281 unique results
were available for screening at the title and abstract level, of which 59 were retained for full-text review. 19 studies were included in the final review (Figure 1). Of the 19 included papers, 11 were qualitative studies, 5 were quantitative studies and 3 studies were categorized as mixed-methods. There were 14 cross-sectional studies and 5 cohort studies. All qualitative studies were categorized as evidence level V, the quantitative and mixed-methods studies were considered to be level III when they were cross-sectional studies, level II for the prospective cohort studies and level I for the prospective cohort studies. A summary of the included studies and their characteristics can be found in Table 1.

The main reason for exclusion of papers during the full-text review was a focus on socio-economic factors that influenced initial access to care rather than whether children continued and successfully completed their care, or because the study examined factors other than socio-economic factors. A complete overview of excluded papers at the full-text review stage can be found in the appendix.

The included studies were further divided into 3 groups: qualitative (Table 2) quantitative (Table 3) and mixed methods studies (Table 4). The qualitative studies reported predominantly on factors influencing non-compliance and drop-out, while the quantitative studies focused more on relapse. All of the qualitative studies used interviews for their data collection and interviewed parents of children with clubfoot along with providers of Ponseti care. The quantitative studies included patient cohorts from the clinics in which one or more of the authors worked. Data was collected from patient files or through questionnaires handed to the parents or primary caretaker of the child. We explored the relationship between certain socio-economic factors and drop-out, relapse and non-compliance with Ponseti treatment. We were able to identify several socio-economic factors and group them together in larger themes.

Poverty and physical accessibility of clubfoot clinics — Several studies link the high cost of transport to and from the clinic, long travel time to the clinic or the risk of losing one or multiple days of income together as a major reason for non-compliance and potentially even drop-out from the program. Pinto et al. were the only ones to report migration of the family and inability to enroll in a program in the new region of settlement as a risk factor for drop-out. In most countries, clubfoot clinics are located predominantly in urban centers or the capital city. In Peru, patients living in the rural and mountainous areas have to travel very long distances to the clinic located in the capital, Lima. Often these parents decide to stay in the capital during the casting phase, but return home during the bracing phase and discontinue follow-up visits because of the long travel time and distance. Patients from rural areas in India have to travel long distances to reach clinics in urban centers. Often these clinics are packed and cannot offer patients and their parents any accommodation, adding accommodation costs onto the burden of lost wages and travel costs. Three of the quantitative and mixed-methods studies were able to confirm the findings of the qualitative studies and prove a strongly significant relationship between 1) living below the poverty line and relapse ($p = 0.001$), 2) lack of financial support and relapse ($p = 0.002$), 3) cost of transport higher than 1 USD and relapse ($p = 0.009$), 4) distance to the Ponseti clinic of more than 9 km ($p = 0.005$) and non-compliance and 5) travel time to the Ponseti clinic between 6 and 12 h ($p = 0.074$) and drop-out.

Presence of support systems — In Kenya, caregivers of children with clubfoot described a general lack of family and community support for children with clubfoot, in some cases leading to abandonment of the mother and her child by the father. Support from family is not only necessary to be able to pay for the treatment, but also to take care of the child with clubfoot at home and care for other children when the mother attends the Ponseti clinic. The importance of support from the father, the immediate family, and the wider community to avoid non-compliance and drop-out was also described by Ugandan, Indian and Peruvian researchers. According to Kazibwe et al., the risk of non-compliance is significantly higher ($p = 0.028$) in children whose caregivers lack immediate family support.

Educational level of the parents — Educational level as a risk factor was identified in one qualitative study and in one mixed-methods study, however the risk factor’s impact was not statistically significant ($p = 0.191$). Several qualitative studies, however, looked beyond the strict definition of educational level and were able to link lack of knowledge about the Ponseti treatment and lack of knowledge about clubfoot in general to drop-out and non-compliance. Kazibwe et al. however, were not able to identify a significant relationship between non-compliance and lack of knowledge about clubfoot ($p = 0.14$).

Household-level factors and cultural norms — In Nigeria, most parents desire large families; in this context, parents would postpone treatment or drop-out from treatment to prioritize an additional pregnancy. In Uganda, a similar pattern was seen, where mothers who have to care for other children in the household cannot combine this care with taking the child to the clubfoot clinic on a regular basis. However, Qudsi et al. were not able to correlate relapse cases to the order of the child in the family, and Poudel et al. only found a moderate non-significant relationship between drop-out and number of children in the household. Four studies describe being a girl as a risk factor for accessing and continuing adequate care. In India, girls have 2.6 times increased risk of dropping out compared to boys, and they are also at risk of not properly

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Figure 1. Flow chart of search results (image created by Covidence 2.0).
| Reference      | Country                | Type of publication | Research design | Study design          | Follow-up period | Period of data collection/participant enrollment | Level of evidence |
|----------------|------------------------|---------------------|-----------------|-----------------------|------------------|--------------------------------------------------|-------------------|
| Akintayo 2012  | Nigeria                | Journal article     | Qualitative     | Cross-sectional       | --               | December 2009 – 2010                              | V                 |
| Behera 2021    | India                  | Journal article     | Quantitative    | Retrospective cohort study | 5 years         | January 2009 – December 2012                     | II                |
| Boardman 2011  | Chile, Peru, Guatemala | Journal article     | Qualitative     | Cross-sectional       | --               | Not available                                    | V                 |
| Evans 2021     | Bangladesh             | Journal article     | Qualitative     | Cross-sectional       | --               | July-October 2019                                 | V                 |
| Gadokh 2012    | India                  | Journal article     | Qualitative     | Cross-sectional       | --               | Not available                                    | V                 |
| Kazibwe 2009   | Uganda                 | Journal article     | Mixed-methods   | Cross-sectional       | --               | Not available                                    | III               |
| Kingau 2015    | Kenya                  | Journal article     | Qualitative     | Cross-sectional       | --               | Not available                                    | V                 |
| Limpaphayom 2019 | Thailand               | Journal article     | Quantitative    | Retrospective cohort study | 6 months | 2011 – 2016                                       | II                |
| Lu 2010        | China                  | Journal article     | Qualitative     | Cross-sectional       | --               | Not available                                    | V                 |
| McElroy 2007   | Uganda                 | Journal article     | Qualitative     | Cross-sectional       | --               | Not available                                    | V                 |
| Mootha 2011    | India                  | Journal article     | Quantitative    | Prospective cohort study | 2 – 7 years   | June 2003 – January 2007                         | I                 |
| Muzzammit 2021 | Pakistan               | Journal article     | Quantitative    | Cross-sectional       | --               | January – December 2018                          | III               |
| Nogueira 2013  | Brazil                 | Journal article     | Qualitative     | Cross-sectional       | --               | Not available                                    | V                 |
| Palma 2013     | Peru                   | Journal article     | Qualitative     | Cross-sectional       | --               | Not available                                    | V                 |
| Pinto 2021     | India                  | Journal article     | Mixed-methods   | Prospective cohort study | 6 years | August 2011 – July 2017                        | I                 |
| Pletch 2015    | Peru                   | Journal article     | Qualitative     | Cross-sectional       | --               | 2013                                             | V                 |
| Poudel 2019    | India                  | Journal article     | Mixed-methods   | Cross-sectional       | --               | January – March 2014                             | III               |
| Qudsi 2019     | Haiti                  | Journal article     | Quantitative    | Retrospective cohort study | 4 years     | November 2011 – October 2015                     | II                |
| Shayo 2015     | Tanzania               | Conference abstract | Qualitative     | Cross-sectional       | --               | June – August 2012                               | V                 |

Table 1: Included studies and characteristics.
| Reference    | Number and type of participants | Reported association between socio-economic risk factors and non-adherence | Risk of bias                                                                                     |
|--------------|---------------------------------|------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------|
| Akintayo 2012 | 42 parents of children between 0 and 5 years of age | non-compliance: inability to pay for transport, inability to pay for medical materials for treatment, losing day of income, higher number of children in the household, parents practicing polygamy, lack of knowledge about clubfoot drop-out: inability to pay for transport, inability to pay for medical materials for treatment, losing day of income, additional children in the household, parents practicing polygamy | Selection bias: not reported Information bias: Interviews conducted in English, relationship between interviewer-participant not described Data interpretation bias: not reported |
| Boardman 2011 | 28 physicians providing Ponseti care | non-compliance: stigma associated with clubfoot, linguistic differences with provider, financial difficulties drop-out: linguistic differences with provider, distance to the clinic, lack of knowledge about the Ponseti method | Selection bias: Not reported Information bias: Interviews conducted in Spanish, interviews conducted by bilingual medical student; relationship between interviewer-participant not described Data interpretation bias: not reported |
| Evans 2021    | 309 parents of children between 7 months and 11 years of age | drop-out: increased parental load/family issues, financial difficulties, distance to the clinic, lack of knowledge about Ponseti method | Selection bias: Adequately addressed. Children included from 8 different clinics across rural and urban settings Information bias: Adequately addressed. Interviews conducted by specifically trained community facilitators unrelated to study team Data interpretation bias: not reported |
| Gadkho 2012   | 38 orthopedic surgeons providing Ponseti care and 19 parents of children with clubfoot | non-compliance: distance to the clinic drop-out: distance to the clinic, inability to pay for medical materials for treatment, female sex, inability to pay for transport, losing day of income relapse: distance to the clinic | Selection bias: Surgeons from hospital and clinics included. No information on representativeness of study population. Information bias: Interviews conducted in Hindi and English, interviews conducted by bilingual medical student; relationship between interviewer-participant not described Data interpretation bias: not reported |
| Kingau 2015   | 10 parents of children with clubfoot and care givers providing Ponseti care | non-compliance: distance to the clinic, inability to pay for medical materials for treatment, lack of community/family support drop-out: inability to pay for medical materials for treatment, lack of community/family support | Selection bias: Representative sample based on background and areas of residence of parents, inclusion of different care providers Information bias: Interviews conducted in Swahili and English; interviewer was main researcher with prior knowledge desired answers Data interpretation bias: not reported |
| Lu 2010       | 39 physicians providing Ponseti care and 8 parents of children with clubfoot | non-compliance: education level of the parents, distrust in western medicine drop-out: inability to pay for medical materials for treatment, inability to pay for transport | Selection bias: not reported Information bias: Interviews conducted in Mandarin, interviews conducted by bilingual medical student; relationship between interviewer-participant not described Data interpretation bias: Adequately addressed. Use of triangulation to assess validity of data collected |

Table 2 (Continued)
Reference | Number and type of participants | Reported association between socio-economic risk factors and non-adherence | Risk of bias
--- | --- | --- | ---
McElroy 2007 | 42 parents of children with clubfoot, 2 adults living with clubfoot, 40 community leaders, 39 traditional healers and 38 biomedical practitioners treating clubfoot | non-compliance: inability to pay for medical materials for treatment, inability to pay for transport, inability to pay for treatment visits, additional children in the household, support/approval from father for seeking care, household workload of primary caretaker/mother drop-out: inability to pay for medical materials for treatment, inability to pay for transport, inability to pay for treatment visits, additional children in the household, support/approval from father for seeking care, household workload of primary caretaker/mother | Selection bias: Representative sample based on background and areas of residence of parents and prior usage of clubfoot treatment Information bias: Adequately addressed. Interviews conducted by specifically trained graduate students, speaking 12 different local languages, coming from diverse backgrounds and who are independent to the study Data interpretation bias: Adequately addressed. Use of triangulation to assess validity of data collected
Nogueira 2013 | 29 orthopaedic surgeons and 16 residents providing Ponseti care | non-compliance: lack of knowledge about the Ponseti method, financial difficulties, stigma associated with clubfoot drop-out: lack of knowledge about the Ponseti method, distance to the clinic, travel time to the clinic | Selection bias: not reported Information bias: not reported Data interpretation bias: not reported
Palma 2013 | 32 physicians providing Ponseti care | drop-out: lack of knowledge about the Ponseti method, distance to the clinic, travel time to the clinic | Selection bias: Representative sample of providers from different types of hospitals across the capital Information bias: Interviews conducted in Spanish, interviews conducted by bilingual medical student, relationship between interviewer-participant not described Data interpretation bias: not reported
Pletch 2015 | 5 parents of children with clubfoot | non-compliance: lack of family support, lack of sharing responsibilities among caregivers, lack of knowledge about the Ponseti method, financial difficulties | Selection bias: not reported Information bias: Interviews conducted in Spanish, interviews conducted by bilingual medical student, relationship between interviewer-participant not described Data interpretation bias: triangulation through focus groups and follow-up interviews
Shayo 2015 | 84 physiotherapists providing Ponseti care | non-compliance: inability to pay for treatment visits, inability to pay for transport, lack of knowledge about the Ponseti method drop-out: inability to pay for treatment visits, inability to pay for transport, lack of knowledge about the Ponseti method relapse: inability to pay for treatment visits, inability to pay for transport, lack of knowledge about the Ponseti method | Selection bias: not reported in abstract Information bias: not reported in abstract Data interpretation bias: not reported in abstract

Table 2: Reported associations between socio-economic factors and non-adherence in qualitative research.
receiving full treatment until they are at a marriable age. Behera et al. who specifically looked at drop-out during the casting phase in an urban setting in India, surprisingly found that more boys than girls drop out during this phase. Pinto et al. looked at another Indian urban setting, found more girls than boys dropping out during the casting phase, but also found more boys dropping out over the entire course of the treatment. It is important to note, that neither the results generated by Behera et al., nor those from Pinto et al. were statistically significant. Qudsi et al. found more girls than boys experiencing relapse in Haiti (p = 0.04) while Limpaphayom et al. were not able to reproduce the same results in Thailand.

Nine studies described efforts taken to address selection bias, but only six included information about the representativeness of their included population. The majority (14/16) of studies described measures taken to address information bias: language of the questionnaire adapted to population, description of patient file, representative sample of clubfoot population of the larger population around the capital, usage of standardized international Clubfoot Registry forms for data collection, and usage of adjusted relative risk ratios and confidence intervals to adjust for confounding. Nine studies described efforts taken to address selection bias, but only six included information about the representativeness of their included population. The majority (14/16) of studies described measures taken to address information bias: language of the questionnaire adapted to population, description of patient file, representative sample of clubfoot population of the larger population around the capital, usage of standardized international Clubfoot Registry forms for data collection, and usage of adjusted relative risk ratios and confidence intervals to adjust for confounding.

| Reference       | Number and age of participants | Reported association between socio-economic risk factors and non-adherence | Risk of bias                                                                 |
|-----------------|--------------------------------|--------------------------------------------------------------------------|-------------------------------------------------------------------------------|
| Behera 2021     | 384 children with a median age of 3 months | Drop-out: male sex (p > 0.005), child was born at home instead of a hospital (p > 0.005), distance to the clinic > 50 km (p > 0.005) | Selection bias: all children enrolled at the clinic within the selected timeframe were included in the study. Information bias: not reported. Data interpretation bias: not reported. |
| Limpaphayom 2019 | 34 children with club-foot between 0 and 58 weeks of age | relapse: female sex (p = 0.61), living outside of the capital metropolitan area (p = 0.23), not living with biological parents (p = 0.53) | Selection bias: not reported. Information bias: Adequately addressed. Sample only includes children operated and treated by the main researcher. Data interpretation bias: only crude odds ratio and confidence intervals reported. |
| Mootha 2011     | 86 children with club-foot between 0 and 1 year of age | relapse: living below the poverty line (p = 0.00) | Selection bias: not reported. Information bias: not reported. Data interpretation bias: not reported. |
| Muzzammil 2020  | 153 children with club-foot between 0 and 3 years of age | non-compliance during bracing: malnutrition 18.19% vs no malnutrition 3.36%, relapse: malnutrition 16.02% vs no malnutrition 10.22% | Selection bias: representative sample based on background and social class of parents. Information bias: data assessor not reported. Data interpretation bias: not reported. |
| Qudsi 2019      | 168 children with club-foot between 0 and 4.4 years of age | Relapse: female sex RR 1.54 (p = 0.04), child is the first-born child RR 1.02, child is a native of the capital metropolitan area RR 1.27, child was born at home instead of a hospital RR 1.14 | Selection bias: inclusion of all children with foot abnormalities to avoid accidental exclusion based on wrong classification of patient file. Representative sample of clubfoot population of the larger population around the capital. Information bias: usage of standardized international Clubfoot Registry forms for data collection. Data interpretation bias: usage of adjusted relative risk ratios and confidence interval to adjust for confounding. |

Table 3: Reported associations between socio-economic factors and non-adherence in quantitative research.

Discussion
We were able to identify four categories of socio-economic factors that increase the risk for drop-out, relapse or non-compliance with the Ponseti treatment: poverty and physical accessibility of clubfoot clinics, presence of support systems, educational level of the parents, and household-level factors and cultural norms. These factors were found to be significant across different studies and in different countries.
factors are very similar to the socio-economic factors identified in studies looking at the barriers parents and children face when seeking care initially. To our knowledge, this is the first time a study looks into the barriers patients face when being enrolled in Ponseti treatment and trying to complete the prescribed treatment regimen. This understanding is necessary to inform and maintain sustainable clubfoot care in LMICs.

Poverty-related factors and lack of physical accessibility to Ponseti treatment were reported in every qualitative study, while other factors were more regional in nature. Reduced access to care for girls was identified as an important and statistically significant risk factor in two studies from South Asia and in one study from the Caribbean. Women and girls are known to have reduced access to health care compared to their male counterparts around the world and especially when living in poverty. Additionally, disabled women and girls lack access to adequate services around the globe and are at increased risk for discrimination and abuse. Given the small number of studies included in this scoping review, it is difficult to determine whether decreased access to care for girls is indeed a larger problem in certain parts of the world, or rather if these researchers were more aware of the global injustices women and girls face when seeking care and actively decided to research this topic.

Lack of family and community support seems to be an issue raised more often in studies from the African region. Malagelada et al. confirm this issue in their South African study as well. They observed a higher reliance of parents on their families and wider communities as well as a higher use of coping strategies during the treatment of their children. One of the explanations given for this disparity is the underlying poverty that parents in South Africa face which already places an increased strain on families and households irrespective of having a child with clubfoot.

McElroy et al. stated eloquently that “human behavior is complex and behavioral factors are difficult to separate from each other”. This statement can, in the context of this review, be expanded to physiological factors beyond behavior as well. In Muzzammil et al., the context of this review, be expanded to physiological factors beyond behavior as well. In Muzzammil et al., the authors show that malnourished children are at higher risk of relapse compared to their well-nourished peers. However, what remains unclear is how the impact of poverty on parents’ behavior intersects with the

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### Table 4: Reported associations between socio-economic factors and non-adherence in mixed-methods research.

| Reference | Number, type and age of participants | Reported association between socio-economic risk factors and non-adherence | Risk of bias |
|-----------|-------------------------------------|------------------------------------------------------------------------|-------------|
| Kazibwe 2009 | 167 parents of children between 0 and 7 months of age | non-compliance: lack of knowledge about clubfoot (p = 0.14), lack of support from clinicians (p = 0.022), lack of family support (p = 0.028), lack of financial support (p = 0.092), transport cost > 1 USD (p = 0.009), distance to clinic > 9 miles (p = 0.005), longer waiting time at clinic (p = 0.52) | Selection bias: not reported |
| Pinto 2021 | 965 children between 0 and 99 months of age | Drop-out: female sex (p = 0.061), inability to pay for medical materials for treatment, losing day of income, migration to new place, lack of family support, distance to clinic, non-availability of transport, lack of knowledge about Ponseti treatment, superstitious beliefs about clubfoot | Selection bias: not reported |
| Poudel 2019 | 238 children with club-foot between 0 and 18 years of age | non-compliance: lack of knowledge about clubfoot (p = 0.14), lack of support from clinicians (p = 0.022), lack of family support (p = 0.028), lack of financial support (p = 0.092), transport cost > 1 USD (p = 0.009), distance to clinic > 9 miles (p = 0.005), longer waiting time at clinic (p = 0.52) | Selection bias: not reported |

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physiological impact of malnutrition on the remodeling in the children’s feet. This cross-cutting effect of poverty, across almost all the identified risk factors, should be kept in mind when identifying and analyzing people’s intentions and behavior. This is especially the case in qualitative studies where providers and not the caretakers of the children were interviewed. One might question to what extent these providers were able to dissect the complex interactions between poverty and other risk factors in patients and how it eventually influenced their behavior.

This cross-cutting effect of poverty in disadvantaged populations is not unique to the LMIC setting and has also been studied in high-income countries. Zionts et al. evaluated the impact of socio-economic factors on the population they treat in Los Angeles, California, USA.57 None of the evaluated socio-economic factors returned statistically significant. Only income-level of the parents rendered a borderline significant result \( (p = 0.07) \).57 However, Avilucea et al. went a step further and did a sub-population analysis of the population attending their clubfoot clinic in Albuquerque, New Mexico, USA.58 Patients attending the clinic from rural parts of the state (used as a proxy for distance and travel time to the clinic), especially Native American patients, faced similar issues as patients in LMICs. The socio-economic factors that increased the risk of relapse in patients living in rural areas were: Native American ethnicity, single parenthood, living below the poverty line and having no or public insurance.58 These results strongly confirm the cross-cutting nature of poverty and the aforementioned complex interactions between poverty and behavior.

While it would be ideal to alleviate the socio-economic factors identified in our work as inhibitors of adequate treatment of clubfoot, it is more practical in the short-term to acknowledge them and attempt to design programs that decrease their influence on treatment. The aforementioned WHO adherence to long-term therapies framework can be of use here. Even though the framework focuses heavily on increasing adherence for therapies for infectious diseases and non-communicable diseases, the 5 identified dimensions (Figure 2) would also be of value to take into consideration when drafting clubfoot programs around the world.

The importance of the health system and the health care team are also clear in the organization of Ponseti care. The lack of formal recognition of the Ponseti treatment by the Ministry of Health in Peru, linguistic differences between patients and caregivers in Guatemala,40 difficulties acquiring braces in Brazil, which can take up to 6 months,45 and the lack of hands-on Ponseti training in China57 are barriers that can only be overcome through systems strengthening approaches. These approaches include physician training, evidence-based planning of health care services, and stronger relationships with providers of medical materials locally and abroad.

Condition-related factors include the severity of the disability and its curability, while patient-related factors include parents’ and patients’ knowledge, attitudes and beliefs.57 Educating parents and caregivers about the physical, social, and economical importance of treating a child’s clubfoot must be a cornerstone in any effort to increase adherence. Factors such as devaluing girls’ medical care can also be addressed through education. The significant economic effects of a family member living with a lifelong disability on the wider community could be used to show the importance of Ponseti treatment. Some efforts have been undertaken to alleviate the therapy-related barriers. Trials which examine less invasive techniques to perform Achilles’ tendon

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**Figure 2.** The five dimensions influencing and impacting adherence to long-term therapies. Reproduced from “Eduardo Sabaté. Adherence to Long-Term Therapies: Evidence for action. Geneva; 2003” (Ref. 27).
tenotomies, or to speed up the casting phase, are promising. Studies looking into making the bracing period less straining physically and in duration are currently still lacking.

This study has several limitations. The majority of studies included were conducted with a broader or different aim than establishing a correlation between socio-economic factors and non-compliance, relapse or drop-out. Therefore, the wording used to describe socio-economic factors was often vague and lacked further clarification. As such, it is unclear whether our interpretation of terms such as “lack of family support” or “financial constraints” is in line with the meaning initially intended by the interviewees and the authors. Almost half of the qualitative data stems from interviews with Ponseti care providers which relies on their interpretation of obstacles faced by patients and their families. This makes the collected data less valid and reliable than when it is collected directly from parents and patients living with these experiences. The data collected from the mixed-methods and quantitative studies are more reliable. However, unfortunately, these studies focused predominantly on relapse and less on risk factors associated with drop-out and non-compliance. Many of the socio-economic factors analyzed in the qualitative papers, or the qualitative sections of the mixed-methods papers, were unique to that specific paper, or were analyzed in relation to a different outcome across different papers. This extreme heterogeneity among collected variables within a small sample of papers made it impossible to run a meta-analysis on the quantitative data available in this review.

Poverty can be identified as the most important socio-economic risk factor based on this review, reinforcing poverty eradication as the challenge for the 21st century. Future quantitative studies are needed to help identify socio-economic factors affected by poverty such as malnutrition and inability to pay for treatment and transport, where targeted policies and programs can have a meaningful impact. Additional regionally-focused studies are needed to better understand how certain cultural factors affect specific regions in the world differently than others, such as decreased access to treatment for girls and lack of family and community support during treatment. Additionally, the discrepancy between girls’ access to care in rural and urban setting should be further explored to better understand how poverty eradication can potentially benefit girls’ access to care irrespective of where they live. However, the absence of these locally generated studies should not inhibit clubfoot programs, and their providers should be aware of these disparities and provide support where they can. The WHO adherence to long-term therapies framework can serve as a guideline to initiate future studies and programs and put the role of socio-economic factors into better perspective. Non-compliance and drop-out will only be eliminated when all dimensions of the framework are adequately covered through meaningful collaborations between the surgical-clinical realm and the public health sphere.

Contributors
MP, AV and BCA developed the search strategy and CM conducted the database search. MP and AV conducted the literature review and data analysis. MP, AV, HAD and CM contributed to the writing of the article. All authors contributed to the review and editing process of the article. All authors reviewed and approved the final version of the manuscript.

Data sharing statement
The full study protocol and a complete overview of included and excluded abstract and full-texts are available upon request by emailing the corresponding author.

Declaration of interests
We declare no competing interests.

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