Ponseti method treatment of neglected idiopathic clubfoot: Preliminary results of a multi-center study in Nigeria

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Abstract

AIM
To evaluate the effectiveness of the Ponseti method for initial correction of neglected clubfoot cases in multiple centers throughout Nigeria.

METHODS
Patient charts were reviewed through the International Clubfoot Registry for 12 different Ponseti clubfoot treatment centers and 328 clubfeet (225 patients) met inclusion criteria. All patients were treated by the method described by Ponseti including manipulation and casting with percutaneous Achilles tenotomy as needed.

RESULTS
A painless plantigrade foot was obtained in 255 feet (78%) without the need for extensive soft tissue release and/or bony procedures.

CONCLUSION
We conclude that the Ponseti method is a safe, effective and low-cost treatment for initial correction of neglected idiopathic clubfoot presenting after walking age. Long-term follow-up will be required to assess outcomes.

Key words: Ponseti method; Neglected clubfoot; Nigeria; Walking age

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Core tip: This is a retrospective study to evaluate the efficacy of the Ponseti method for initial correction of untreated, idiopathic clubfoot in patients above one year of age. The rate of initial correction to a painless, plantigrade foot without the need for soft tissue release was 78% in 255 evaluated clubfeet from 12 treatment centers in Nigeria. The Ponseti method, a non-operative treatment method involving serial manipulation and casting, is useful and effective for initial correction of clubfoot, even after walking age.

INTRODUCTION
Idiopathic clubfoot is the most common musculoskeletal congenital birth defect, affecting between 1 and 7 births in every 10000-1. Left untreated, clubfoot may lead to pain, disability, discrimination, and hardship throughout the person’s life. This is especially a problem in developing countries like Nigeria where 70% of the population lives below the poverty line and has limited access to treatment. Although Nigeria has around half the population of the United States at about 180 million, it is estimated that the prevalence of clubfoot is at least three to five times higher than in the United States. This high prevalence is mainly a result of the number of people living with neglected clubfoot, defined as clubfoot that has not been treated before walking age[2,3]. For many years in Nigeria and around the world, soft tissue release or extensive bony surgery were seen as the only options for the full correction of clubfoot. In the past two decades, the minimally invasive Ponseti method has been proven and accepted as the worldwide gold standard for treatment of clubfoot based on its success in both the short term and long term[4]. Introduced in Nigeria in 2009, the Ponseti method is a safe, effective, and economical option for many patients who cannot afford an extensive surgery. The method involves skilled serial manipulation and plaster casting as well as a percutaneous Achilles tenotomy (PAT) to completely correct the deformity. After the correction is complete, the child wears a foot abduction brace at night for a few years to prevent relapse[5-7].

There are few studies on the effectiveness of the Ponseti method for patients over age 1, but the early evidence has been promising. Studies in Brazil, Nepal, India and Ethiopia reported that use of the Ponseti method allowed for avoidance of soft tissue release in 66%-92% of cases above one year of age[8-10].

The purpose of this study was to evaluate the effectiveness of the Ponseti method for initial correction of neglected clubfoot cases in multiple centers throughout Nigeria.

MATERIALS AND METHODS
The accepted definition of neglected clubfoot is true idiopathic clubfoot that has not been treated before walking age. In this study, age one will be used as representative of walking age. Through July 2015, 1137 clubfoot cases from 2010-2015 were recorded from Ponseti treatment centers in Nigeria into the International Clubfoot Registry with patient permission. Patient charts were reviewed through the registry. Two hundred and twenty-five patients (328 clubfeet) from 12 different centers (Figure 1) met inclusion criteria and are profiled in Table 1. Patients were excluded if they were younger than 1 year of age, had non-idiopathic clubfoot (syndromic, neuromuscular, etc.) or had other treatment prior to Ponseti method.

Treatment at all 12 centers, including casting and percutaneous Achilles tenotomy as necessary, was performed by orthopaedic surgeons who were properly trained in the technique according to Ponseti[5-7]. Casts were changed at intervals of 7-10 d if possible but up to every 14 d depending on the ability of the patient to reach the clinic and pay for the treatment. All patients were treated on an outpatient basis although two of the twelve centers, including 18 total patients, performed...
tenotomies in the operating theatre for optimal sterility. Although tendon transfer can be considered part of the Ponseti protocol, one of the main objectives of this study is to show that neglected clubfoot can be corrected non-operatively as that is most beneficial in the low resource setting. Therefore in this study, tendon transfer will be considered failure in treatment along with tendoachilles lengthening and major soft tissue release.

After completion of serial casting, correction was maintained with a foot abduction brace at night or in rare cases when braces were not available, with encouragement of active play (walking, running, etc.). Abduction braces were generally crafted by each hospital’s orthotics department or local cobbbers working with the orthopaedic surgeons.

In the newborn population, Pirani and Dimeglio scoring systems are generally used to assess the severity and characteristics of a clubfoot. These systems have been validated and are useful for comparing the quality of correction\textsuperscript{[17,18]}. However, these have not been validated for the neglected population and no system exists yet for the qualification of neglected clubfoot\textsuperscript{[19]}.

### RESULTS

The study population consisted of 225 patients, 131 (58.2%) were male, and the clubfoot was bilateral in 103 cases (46%). One hundred and forty-seven patients were age 1 or 2, 59 were between the ages of 3 and 9, and 19 patients were age 10 to 16.

Of 328 neglected clubfeet studied, 255 feet (78%) achieved initial correction, a completely plantigrade foot, without soft tissue release or other surgical intervention besides PAT. Of the 73 other feet, 47 were lost to follow up mid-treatment and 26 of the feet were converted to Achilles tendon lengthening, tendon transfer procedures, or major soft tissue release.

A profile of the total number of casts required for complete correction and the PAT rates based on age group is shown in Table 2. A distribution of the number

### Table 1  Phenotypic characteristics $n$ (\%) 

| Cases | 
|-------|
| Female | 94 (42) |
| Male | 131 (58) |
| Bilateral | 103 (46) |
| Right | 66 (29) |
| Left | 56 (25) |
| Age $\leq$ 2 yr | 147 (65) |
| Age $>$ 2 yr | 59 (26) |
| Age $\geq$ 10 yr | 19 (9) |

Figure 1  Map of clubfoot clinic sites. A: LASUCOM; B: FMC Abeokuta; C: University of Ilorin; D: Obafemi Awolowo University; E: Wesley Guild Hospital Ilesa; F: FMC Ido-Ekiti; G: Ekiti St Teaching Hospital; H: FMC Owo; I: Gwarinpa GH; J: FMC Keffi; K: NOH Enugu; L: NKST-Mkar, Benue.
of casts required across all age groups is shown in Figure 2. The average number of casts necessary for correction was 6.84, ranging from 6.71 casts/foot for 1 and 2 year olds up to 7.43 casts/foot for those above age 10. Analysis of variance between the groups shows no statistically significant difference ($P = 0.78$). The overall rate of percutaneous Achilles tenotomy was 51% and also tended to increase with patient age from 42% in the youngest age group to 60% in the oldest. There were no failures of PAT's reported.

**DISCUSSION**

Neglected clubfoot is a serious problem in developing nations where a significant number of children born with the deformity do not receive immediate treatment. It is common for parents to delay treatment due to cost, lack of transportation or limited availability of proper care. Frequently, children do not present until around the start of school age at 4 or 5 because of increased interaction with peers and social pressures to fit in. Neglected clubfoot can cause significant physical, psychological and financial burden to the child and the family. Adults with untreated clubfoot may experience significant pain and disability, be unable to work, and have difficulty performing daily activities of living.

Historically, neglected clubfoot has been treated with bony surgery and extensive soft tissue release. These techniques are difficult, costly, have substantial complication rates, and they are rarely feasible in the healthcare settings of developing nations$^{[20-23]}$.

Our study has shown that the Ponseti method is effective in the initial correction of neglected clubfoot and reduction of the need for surgical treatment beyond percutaneous tenotomy. Its suitability at the clinical level is imperative for use in developing countries like Nigeria, where operating room access comes at a premium. Our results are similar to multiple recent studies on Ponseti treatment of neglected clubfoot cases, shown in Table 3.

Ponseti et al$^{[5]}$ described successful results from Brazil after Ponseti treatment in 17 children (24 feet) with an average age at presentation of 3.9 (1.2-9.0). A full correction was achieved in 16 feet (67%) without the need for extensive surgery and it was found that those who eventually needed surgery ended up with a lesser surgical intervention. Six of the patients had recurrence, and the best results were achieved in the youngest group.

In Nepal, Ponseti et al$^{[6]}$ reported the use of Ponseti treatment in neglected cases between the age of 1 and 6. In 260 feet, 83% were able to avoid any surgery besides PAT. Similar to our study, with a broader range of ages and larger study group, they found a wider range of casts were necessary to achieve full correction.

Spiegel et al$^{[9]}$ in India had 37 neglected clubfoot patients (55 clubfeet) with a mean age of presentation of 2 (1-3) years. After a Ponseti trial, 4 patients (6 feet) were considered unfit for Ponseti treatment and had a posteromedial release, 4 patients (4 feet) relapsed and had tibialis tendon transfer and 3 patients (4 feet) required tendoachilles lengthening to obtain adequate dorsiflexion.

In Ethiopia, Khan et al$^{[11]}$ found that in 22 neglected clubfoot patients (32 feet), an open tendoachilles lengthening was required for 5 patients (7 feet) plus posterior capsulotomy in 3 additional patients (4 feet). Correction was achieved for the remaining 14 patients (21 feet) with the Ponseti method and PAT alone.

In our study, 78% of 328 feet achieved correction without major surgery. Some factors, such as time between casts, were variable between the 12 different treatment centers because of the socioeconomic differences between the regions of Nigeria. In a survey of
the providers at each center, reportedly patients who were wealthier or in more urban areas were more likely to have casts changed slightly more frequently, 7-8 d as opposed to 13-14. PAT’s were performed under local anesthesia in clinics at 10 sites and in the operating theatre at 2 sites (18 study patients) for better infection control and sterile instruments.

If the number of casts required for correction can serve as a proxy for the severity of a clubfoot, neglected clubfoot can be described simply as a more severe form of idiopathic clubfoot in a newborn. The average number of casts required in newborns is around 5 with a correction rate around 90%.[20] In our study and review of the literature, casts required for neglected cases is between 7 and 10 with successful correction rates from 66%-83%. The Ponseti method is nearly as successful on neglected clubfoot as in newborns at preventing the need for surgical intervention.

The phenotypic characteristics for cases in our study were similar to the reported rates of incidence of congenital idiopathic clubfoot in the literature.[24-27]. Fifty-eight percent of patients in our study population were male, consistent with the evidence that males are twice as likely to be affected. Also, in concordance with the evidence that half of cases occur bilaterally, 46% of our cases were bilateral and 54% were unilateral. This is significant because there has not been profound gender or laterality bias in presentation of neglected cases. Therefore, these factors do not seem to affect parents’ willingness to seek treatment for their children before age 1.

One intriguing finding in our study is that the percutaneous Achilles tenotomy rate seems to be lower than in much of the literature.[3][11]. There are a few unique factors that could be causing this finding. The first is that there was considerable variability in rate of tenotomy between different centers. Costs for sterile equipment, local anesthetic and time required may be causing some providers to increase the number of casts to achieve necessary dorsiflexion rather than performing a PAT. It is also possible that some of the cases treated were actually positional equinovarus deformity rather than true talipes equinovarus, and did not require tenotomy. General guidelines for the Ponseti method recommend that a clubfoot achieve 15° of dorsiflexion to be considered corrected.[22] More research into the tenotomy rate for neglected clubfoot worldwide and for all idiopathic clubfoot in Nigeria would be encouraged.

Our study has some potential limitations. During the course of treatment, 47 patients were lost to follow up. In the setting of a developing country, patients are often lost to follow-up because of socioeconomic factors such as costs, transportation, and lack of social support and behavioral factors such as feeling improved function mid-treatment and knowledge deficit about duration of treatment.[24] Data collection was variable between centers so 32 neglected clubfoot patients had to be excluded from the initial study population because there was not enough treatment data. If more data could be recorded, research into the feasibility of Pirani and Dimeglio scoring for neglected clubfoot would be very helpful and could aid providers in guiding their manipulation and casting to each individual. The current algorithm for Ponseti treatment of neglected clubfoot is provided in Figure 3.

In conclusion, this study has shown that the Ponseti method can be effectively used to correct neglected clubfoot in children older than walking age. If the method is performed properly, it will significantly reduce the need for bony surgery or major soft tissue release.

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COMMENTS
Background
Clubfoot is the most common musculoskeletal birth defect and left untreated, can result in lifelong disability and hardship. The Ponseti method involving serial manipulation and casting has become the gold standard in treatment of clubfoot. In low resource and developing countries, neglected clubfoot, or clubfoot that is untreated by one year of age, is a significant problem because of limited access to healthcare. In this study, the authors evaluated the efficacy of the Ponseti method in treating neglected clubfoot in 12 centers in Nigeria.

Research frontiers
Currently, there is very little literature on the effectiveness of the Ponseti method in clubfoot patients over age one. The current studies are on small populations.
but have shown significant success.

**Innovations and breakthroughs**

In this study, the Ponseti method was successful in the initial correction of 78% of neglected clubfeet. These results are in concurrence with previous, smaller studies which showed similar correction rates.

**Applications**

This study suggests that providers throughout the world who are treating neglected clubfoot should begin with the Ponseti method as it can be performed at low cost and is highly effective.

**Terminology**

**PAT**: Percutaneous achilles tenotomy - minimally invasive procedure involving the nicking of the heel cord in order to achieve maximal stretching in the final phase of treatment.

**Peer-review**

The paper is interesting and emphasizing the statement that Ponseti method can be effectively used to correct neglected clubfoot in children older than walking age, and significantly reduce the need for bony surgery or major soft tissue release.

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