Treatment outcomes for standard Ponseti method in amyoplastic clubfoot

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Background
Arthrogrypotic clubfoot tends to be more rigid and severe than their idiopathic counterparts. Ponseti method has shown satisfactory results in treating clubfoot with distal arthrogryposis; however, no published work yet has investigated the role of Ponseti technique in dealing with the more rigid amyoplastic clubfoot. In the present study, we aimed to assess the outcomes of standard rather than modified Ponseti technique in the management of infants with amyoplastic clubfoot deformity.
Patients and methods
Standard Ponseti technique was used to treat 14 clubfeet (seven patients) with classic arthrogryposis (amyoplasia congenita) between July 2011 and March 2015. Our primary outcome measure was the quantitative degree of deformity correction assessed by Dimeglio and Pirani scoring systems. Secondary outcome measures included relapses with/without the need for surgical interference and the assessment of functional results.
Results
All patients were followed up for an average of 40.4 months (range, 21–69). The mean precorrection score of Dimeglio was 17.57 (range, 16–20), which significantly improved to a mean of 3.1 (range, 2–4) after correction. The average Pirani score was 5.6 (range, 5–6) and 1.2 (range, 0.5–2.5) initially and before tenotomy, respectively. Satisfactory results were reported in 71.5% of the treated feet, whereas 43% of them had a relapse.
Conclusion
The Ponseti method can be an effective first-line management for amyoplastic clubfeet, decreasing the unnecessary need for multiple surgical procedures and improving the final functional outcome.

Keywords:
amyoplasia congenita, clubfoot, Ponseti method

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Introduction

Clubfeet are the most common foot deformities in arthrogryposis that tend to be more rigid and severe than their idiopathic counterparts. Early treatment has long been recommended, to make the most of the suppleness of the newborn. Earlier articles detailed the difficulty obtaining a satisfactory result, with three to four procedures performed per foot. Much of the recent literature still highlights the high recurrence rate, describing procedures to treat failed feet [1–4]. Recently, the Ponseti method with some modifications has been used to treat clubfeet associated with distal arthrogryposis syndromes, though, Dr Ponseti himself, did not expect a comparable success when the technique used to treat classic or amyoplastic clubfeet, because these are well known to be more rigid than others with distal arthrogryposis (Fig. 1). Few reports could examine the outcome of Ponseti technique in treating clubfoot with a collective sample of amyoplasia and distal arthrogryposis [5–7]. A focused work evaluating the results of standard Ponseti technique in treating the more rigid amyoplastic clubfoot has not came out yet. In this study, we aimed to assess the outcomes of standard rather than the modified Ponseti technique in management of infants with amyoplastic clubfoot deformity.

Patients and methods

This prospective study was conducted between July 2011 and March 2015. The study was approved by the institutional ethics committee in the Orthopedic Department of Orthopaedic Surgery, Menia University, Menia, Egypt. A total of 14 clubfeet in seven consecutive children with amyoplasia congenita (classic arthrogryposis) have been treated in Minia University Hospitals. They all showed classic four

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Figure 1

(a) A patient with amyoplasia congenita showing four limb classic involvements with severe clubfoot deformity. (b) Less severe clubfoot deformity in distal arthrogryposis.

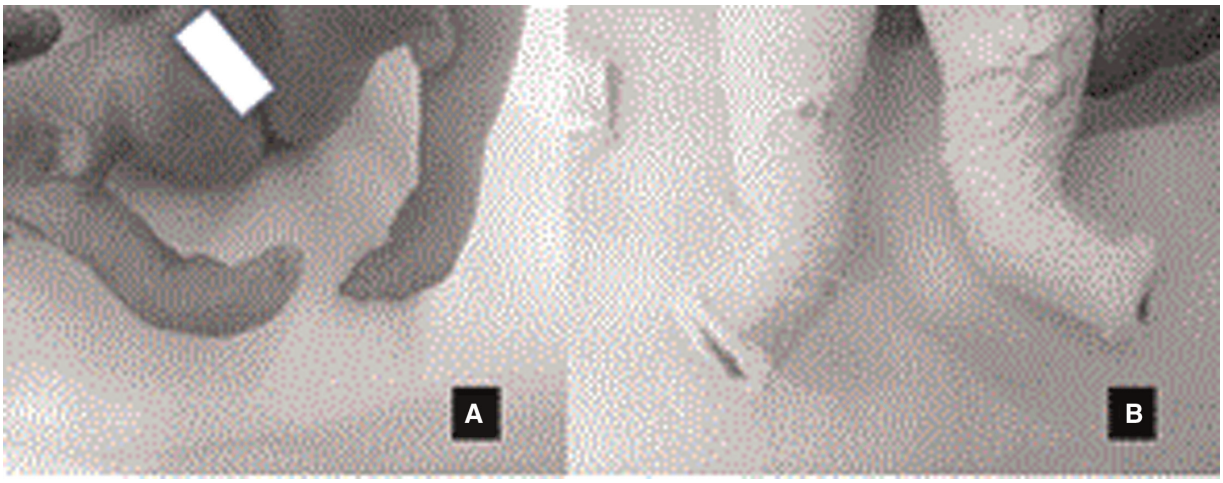
limb involvement with symmetrical knee flexion contractures, elbow extension contractures, and wrist palmar flexion and ulnar deviation. Three hips in three patients were teratologically dislocated. Patients were initially assessed by a neurologist to exclude central nervous system dysfunction and to confirm the diagnosis as amyoplasia congenita. Six children (85.7%) received no previous treatment. On initial presentation, patients' demographics and associated deformities were reported. The primary outcome measure was the quantitative degree of deformity correction assessed by Dimeglio and Pirani scoring systems [8,9]. Secondary outcome measures included relapses with/without the need for surgical interference, and the assessment of functional results was based on criteria described by Niki *et al.* [10]: a plantigrade, pain-free, and braceable foot was considered a satisfactory result, whereas a nonsatisfactory result was defined as a non-plantigrade foot with residual deformity necessitating another surgery. Steps of treatment method and potential complications were fully discussed with parents. Clinical photographs of the deformity were taken after parental consent. The classic Ponseti protocol without modification was followed in manipulation and casting (Fig. 2) [11]. Tenotomy was taken over under local topical anesthetic in the outpatient clinic. After removal of the final cast, children were placed in a locally manufactured version of Markell foot abduction brace (FAB) as originally advised by Ponseti. Children were followed up with quarter yearly clinical review in the first year then every 6 months till latest review. Complications owing to plaster and/or bracing were reported.

Results

A total of 14 clubfeet in seven infants with amyoplasia congenita were followed up for an average of 40.4 months (range, 21–69). All feet were graded as Dimeglio grade IV (very severe) at the time of presentation [8]. There were four girls and three boys, with a mean age of 5.6 weeks (range, 2–16) when treatment started. Initial correction was accomplished in all feet after treatment with a mean of 6.4 casts (range, 6–8). The patients who received initial treatment before presenting to us have had two

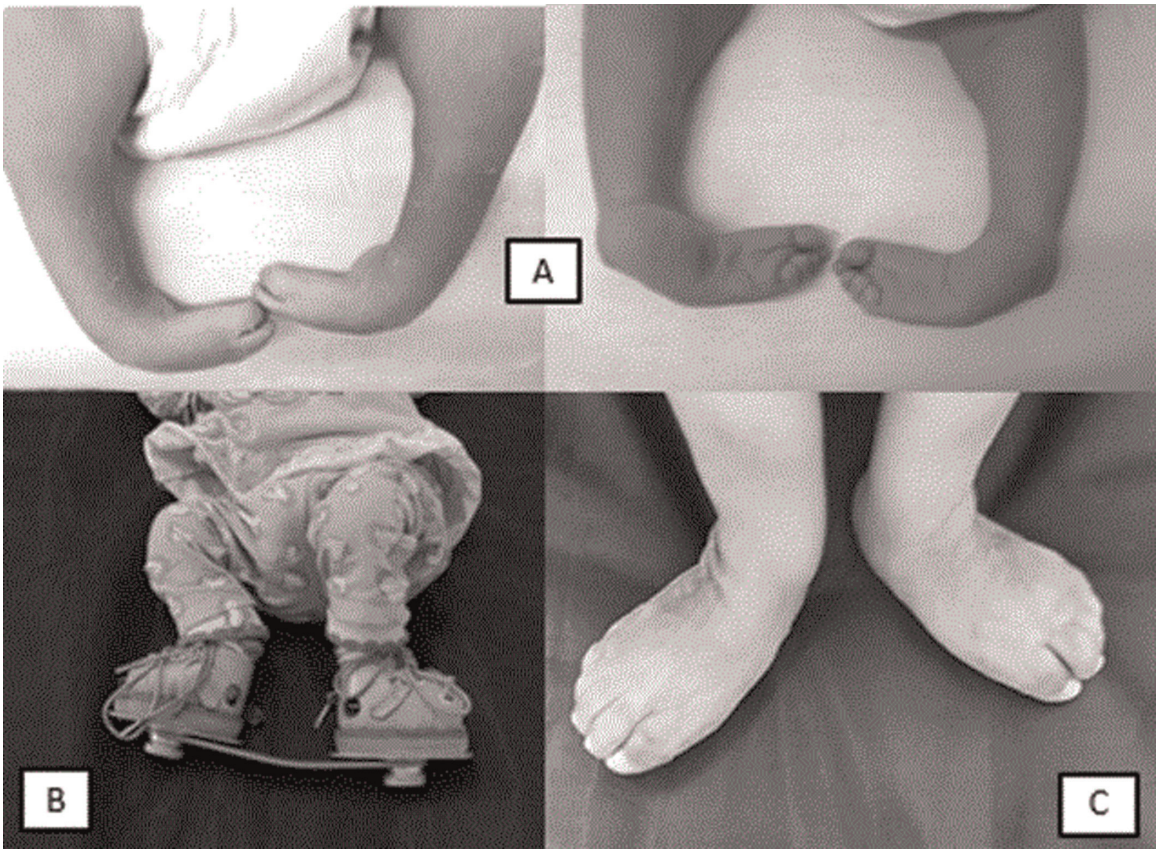
plaster casts without tenotomy; however, the precise information about the technique used was not known. The mean precorrection score of Dimeglio was 17.57 (range, 16–20) that significantly improved to a mean of 3.1 (range, 2–4) after correction ($P<0.001$) [8]. The average Pirani score was 5.6 (range, 5–6) and 1.2 (range, 0.5–2.5) initially and before tenotomy respectively (Fig. 3) [9]. Percutaneous tendo-Achilles tenotomy was done for all feet with an average of 12° of dorsiflexion (range, 10–15) obtained after tenotomy. Although satisfactory initial correction was achieved for all feet at the beginning of using the FAB, supple

Figure 2



(a) A 2-week-old patient with bilateral clubfoot (amyoplasia congenita) presented with marked equinus and cavus deformities (b) After fourth manipulation and casting session according to Ponseti method.

Figure 3



Clinical photographs for a 3-week-old female infant. (a) At presentation with bilateral, severe and symmetrical amyoplastic clubfoot. (b) The patient in FAB. (c) At latest follow-up with maintained initial correction. FAB, foot abduction brace.

adductus trend of the forefoot was noticed while in the first 6 months after bracing in 10 feet (71.4%). Three children (6/14, 43%) showed a relapse after initial successful treatment. Equinus was solely recurred in one of them who required a redo Achilles tenotomy followed by 3 weeks of immobilization in toe-to-groin cast. Combined equinus and adductus deformities were found in the remaining two children. Recurred deformities could be corrected in one of them after a second set of manipulation, casting, and tendo-Achilles tenotomy, whereas bilateral poster-medial surgical release was needed in the last one whose feet showed more resistant deformity. The average time from initial correction till diagnosis of relapse was 19.6 weeks (range, 16–23). The mean age of walking for whole group was 22 months (range, 18–26). At the latest follow-up, all feet showed satisfactory results but four (28.5%), in two patients were plantigrade but painful (Table 1). No complications related to casting or posteromedial release have been reported.

Discussion

Ponseti technique could successfully deal with clubfeet in patients with distal arthrogryposis on few occasions. Using the concept of manipulation and casting in treating those patients saved them the dilemma of unnecessary multiple surgical procedures with reported unsatisfactory outcomes [12,13]. Only a few studies have been published. Matar *et al.* [14] reported 64.7% satisfactory results with Ponseti method in treating 10 patients (17 feet) with arthrogryposis. In their short-term study (average 2 years follow-up), Boehm *et al.* [5] used the technique to successfully treat 12 patients with 24 clubfeet with distal arthrogryposis. Six feet had relapsed but were successfully treated by repeat casting, with an overall reported satisfactory outcome in 11 (92%) patients, with an average child age at final follow-up of 32.3 months. In another short-term study, Van Bosse *et al.* [6] reported satisfactory outcomes in 15/19 (78.9%) arthrogrypotic clubfeet in 10 patients using a modified Ponseti method with initial percutaneous Achilles tenotomy, followed by serial casting and a second tenotomy in 53%, with an average follow-up of 3 years. However, treatment results of Ponseti technique in treating the more rigid clubfeet with classic or amyoplastic form are still questionable, as a pure sample of amyoplastic clubfooted patients has not been examined before this work.

Kowalczyk and Lejman [15] evaluated the results of Ponseti technique in five patients with bilateral arthrogrypotic clubfeet with a similar average follow-up period (35.8 months) and same age group. Three of them had classic arthrogryposis. Posteromedial surgical release was needed to correct recurrent deformities in nine (90%) feet compared with 14.2% in our study. At the final follow-up, satisfactory results according to Nikki criteria were 71.5 and 70% in our and their work, respectively. They needed a higher average number of casts (8.4) to initially correct their patients' feet. Despite their final results being comparable to ours, we could not point out the exact reasons beyond their obviously higher rate of recurrence and the need for surgical release in almost all of their children but one. The higher number of casts in their study may be explained as they have repeated manipulation for three of their children immediately after removal of the cast applied when tenotomy was done aiming to correct any remaining equinus deformity. Although we appreciate their trials of remanipulation proposed to improve dorsiflexion a couple of weeks after tenotomy, it is logical that the degree of dorsiflexion gained immediately after tenotomy cannot be improved by further manipulation and casting especially in arthrogrypotic feet as it would be more attributed to posterior capsular contracture. Moreover, prolonged times of immobilization in arthrogrypotic patients

Table 1 Demographics, results, and outcome measures

Patients	Age at treatment (weeks)	Sex	Number of casts	Number of tenotomy	Dimeglio		Pirani		Relapse	Functional outcome	Follow-up (months)
					Pre	Post	Pre	Post			
SK	3	F	6	1	16	3	5	1	No	Satisfactory	69
FF	4	F	6	1	19	3	6	1	No	Satisfactory	54
MB	2	M	6 (+1)	2	18	4	6	1.5	Yes	Unsatisfactory	44
BA	3	F	6	1	17	3	5	1	No	Satisfactory	36
SA	2	F	6	1	17	2	6	0.5	No	Satisfactory	34
MH	9	M	8	1	20	4	6	2.5	Yes (PMR)	Unsatisfactory	23
AA	16	M	7 (+3)	2	16	3	5	1	Yes	Satisfactory	21

F, female; M, male; PMR, posteromedial release.

may increase other contractures and disturb other ongoing treatment for other joints. Kowalczyk and Lejman [15] had to use an ankle foot orthosis instead of FAB in three of their patients (6 feet) due to concomitant severe external rotation deformity of the hip. Ankle foot orthosis cannot efficiently maintain the corrected forefoot deformity in the required external rotation position with resultant adductus deformity, and secondarily, the heel may migrate upward and tilt inward recalling the original equinovarus position that may explain the higher recurrence rate reported in their study and frequent number of surgical releases needed.

In the present study, satisfactory outcome was achieved, that is, a plantigrade, braceable, and pain-free foot, in 71.5% of our children, with an average follow-up of 40.4 months (range, 21–69). Although initial correction was achieved in all patients, maintaining the correction is mainly dependent on brace compliance and a multidisciplinary team approach to meet their rehabilitation needs.

Conclusion

In our experience, the Ponseti method is an effective first-line treatment for amyoplastic clubfeet that can at least save many unnecessary surgical procedures for those feet, although higher risk of relapse is a potential complication of the disease itself whatever the treatment modality received.

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Conflicts of interest
There are no conflicts of interest.

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