Rare presentation: concomitant incidence of congenital idiopathic clubfoot and simple metatarsus adductus

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Background
Congenital idiopathic clubfoot and simple metatarsus adductus deformities have often been presented individually. In this series, we attempted to report a rare concomitant incidence of both deformities and explain the way they have been treated simultaneously.
Patients and methods
Four patients presented with idiopathic clubfoot on one side and simple metatarsus adductus deformity on the other side between January 2012 and October 2014. These patients were treated using Ponseti and Kite's techniques for clubfoot and metatarsus adductus, respectively.
Results
Mean follow-up period was 39 months (range, 22–56). Patients' mean precorrection Dimeglio score was 9.75 (range, 8–12), whereas the mean postcorrection score was 2.75 (range, 2–4). No relapses have been reported for both deformities. The clubfeet had a mean ankle dorsiflexion of 20°, and the contralateral feet showed normal heel bisector.
Conclusion
Concomitant incidence of clubfoot and metatarsus adductus deformities even if rare should be considered while evaluating newborns with bilateral foot deformities.

Keywords:
clubfoot, congenital incidence, metatarsus adductus

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Introduction

Congenital idiopathic clubfoot (CIC) is one of the most common pediatric orthopedic scenarios. Its incidence has been estimated to be one in every 1000 live births. Diagnosis of CIC can be easily set at birth by health professionals, and its treatment has been widely explored in the literature [1,2]. Another not uncommon congenital foot deformity is the simple metatarsus adductus (SMA) which is by definition a transverse plane deviation at the tarsometatarsal joint without a rear foot deformity; its actual incidence is not well documented and is probably comparable to clubfoot [3,4]. Treatment options for both deformities in infants are well recognized by most pediatric orthopedic surgeons, with a reported good outcome. Ponseti technique has shown remarkable success in treating virgin clubfeet while stretching exercises, and Kite's method has been repeatedly used for treatment of SMA deformity with a good outcome [5,6]. Concomitant incidence of both deformities in the same patient has never been reported in the literature. We report the unlikely simultaneous incidence of both foot deformities in four patients and the way their feet have been treated.

Patients and methods

This retrospective analysis was done on the records of four patients (eight feet, three males, and one female) who had been treated at Al-Minia University Hospitals between 2012 and 2014 for concomitant CIC and SMA deformities using Ponseti and Kite's methods, respectively, after receiving institutional review board approval. The study was approved by ethical committee of Al-Minia University. All patients were operated upon in Al-Minia University hospital after they had signed an informed consent form. Patients enrolled in the study were diagnosed to have CIC in one foot via detection of relevant deformities at the ankle, subtalar, and midtarsal joints, whereas the other foot was found to have a simple transverse plane deviation at Lisfranc joint only with a solely crease at the medial border of the foot (Fig. 1).

Syndromic nature of both deformities was excluded. All patients had undergone no previous treatment. The

average age at presentation was 3 weeks (range, 1–5) and the average age when treatment started was 3.25 weeks (range, 2–6). They were three males and one female. The clubfoot deformity was right sided in three patients. Dimeglio and Bleck's classification systems were used to evaluate the degree of clubfoot and metatarsus adductus deformities, respectively [7,8]. Pirani severity scoring system was applied to monitor progress of manipulation and casting for clubfeet, whereas a hand-held goniometer was used to measure the range of passive ankle dorsiflexion during follow-up period [9]. Plain radiographs (anteroposterior view) for the foot with SMA was requested at presentation to only confirm clinical diagnosis of the deformity and not for angles' measurement. Serial manipulation and casting were used to correct clubfeet as described by Ponseti and Smoley [10]. Kite's method of manipulation was followed to correct the feet with metatarsus adductus where the cuboid-fifth metatarsal joint was used as a fulcrum point against which the forefoot was gently and gradually drawn into abduction while supinating the forefoot to correct cavus deformity if existent. Equinus deformity in CIC was last to be corrected through a percutaneous tendo-Achilles tenotomy and then the foot was placed in a final cast for 3 weeks. After removal of the last cast, the corrected feet were placed in a locally manufactured version of Markell abduction brace with 70° and 45° of outer rotation for CIC and SMA sides, respectively (Fig. 2). Patients' information, including gestational history, characteristics, and demographic data, was recorded in a single sheet for each patient. The degree of ankle dorsiflexion after tenotomy, complications with casting, and problems with the abduction brace if any were reported.

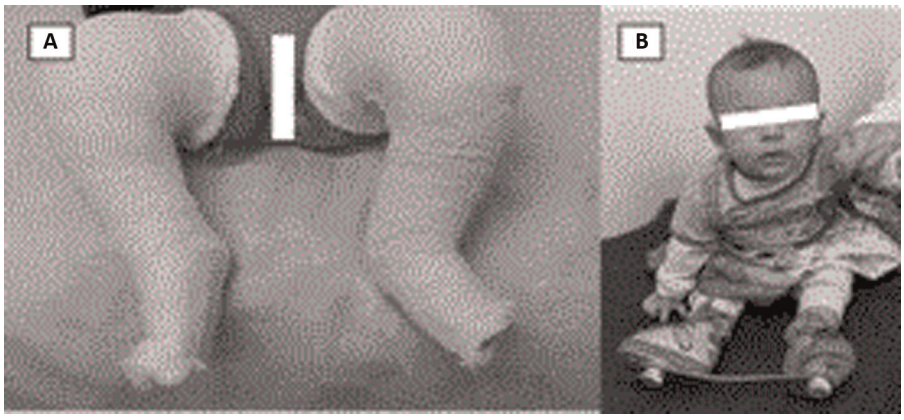
Results

On presentation, three clubfeet were classified as moderately deformed and one was severe on Dimeglio system with a mean score of 9.75 (range, 8–12), which improved to 2.75 (range, 2–4) after correction. Bleck's heel bisector line initially

Figure 1

Clinical photograph shows components of both deformities: (a) convex lateral border of the left foot with mild SMA deformity, and (b) moderate degree of right heel varus with normal heel alignment on the left side. SMA, simple metatarsus adductus.

Figure 2



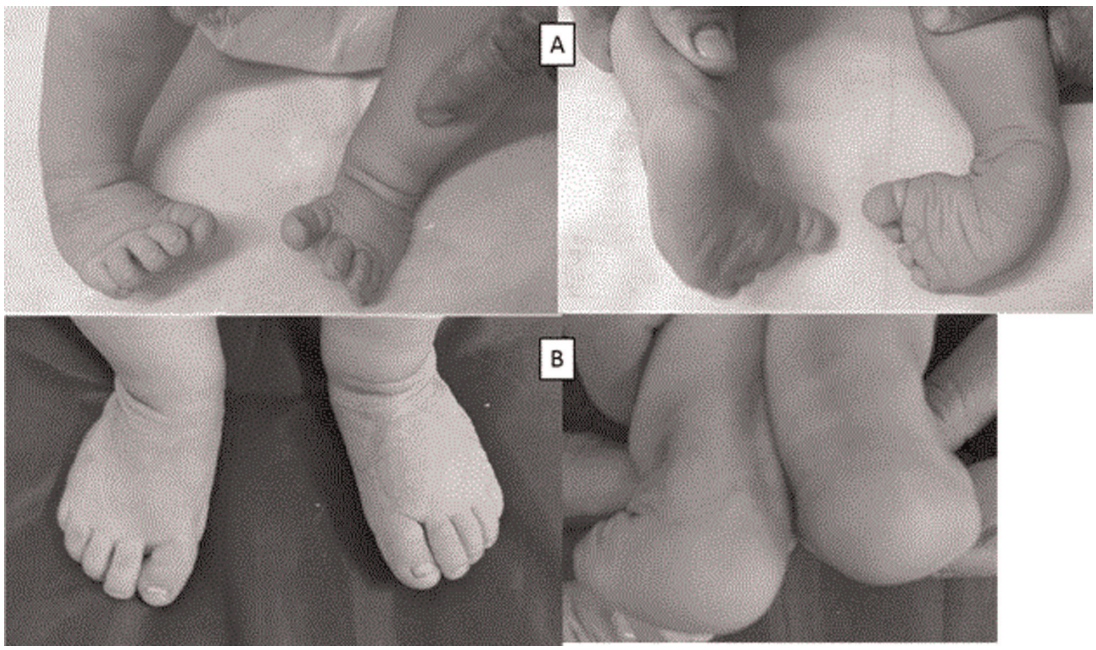
Clinical photograph shows (a) plaster cast application and (b) abduction brace with different degrees of rotation.

demonstrated a moderate deformity in three feet and mild in one with SMA deformity, and at latest follow-up, normal bisector course was restored in all feet (Fig. 3). The mean follow-up was 39 months (range, 22–56). No patients were lost to follow-up. Correction was obtained with a mean of five casts (range, 4–6) for CIC and three casts for all patients with SMA. The cast for the side with SMA was left for longer time to catch up with the other foot on final removal. Before treatment, the mean Pirani scoring system was 4.5 (range, 4–5.5), which was improved to a mean of 1.1 (range, 0.5–1.5) before doing the tenotomy. The positive final Pirani scoring recorded in all patients was attributed to hind foot clinical signs only. A mean of 20° (range, 15–25) of passive ankle dorsiflexion could be obtained by the clubfooted side after tenotomy. Results for all patients are given (Table 1). Neither complication related to casting or bracing nor recurrence for both deformities were reported during follow-up. All patients but one was born via normal delivery. Everyone had vertex presentation and was full term without a history of oligohydramnios or amniocentesis.

Discussion

Clinical examination of a newborn with clubfoot is enough to reveal the essential components of the deformity. The hind foot will be held in equinus with varus malalignment of the heel, whereas the forefoot is in adduction and supination but still less supinated than hind foot producing cavus deformity [7]. SMA shares clubfoot same adductus and to some extent cavus deformity of the forefoot though, missing the hind foot and subtalar deformities [11]. Considering both congenital foot deformities have

Figure 3



A 3-week-old infant: (a) both deformities before correction and (b) at final follow-up.

Table 1 Demographics and results

Patients	I	II	III	IV
Sex	M	F	M	M
Side	LT	RT	RT	RT
Age at treatment (weeks)	2	6	2	3
Dimeglio score (initial)[a]	10	8	9	12
Dimeglio score (final)[a]	3	2	2	4
Pirani score (initial)[a]	4.5	4	4	5.5
Pirani score (final)[a]	1.5	1	0.5	1.5
Number of casts[a]	5	4	5	6
Ankle dorsiflexion [a]	20°	20°	25°	15°
Follow-up (months)	56	46	32	22

[a]Clubfooted side. F, female; LT, left; M, male; RT, right.

similar incidence, it may be little confusing for junior pediatric orthopedic surgeons to differentiate, especially when they happen concomitantly with mild deformity of clubfooted side.Proposals of etiology for both deformities are still controversial. We wish if we could support the validity of intrauterine compression theory as a causative factor for concomitant incidence of both deformities [12], but unfortunately, negative history of oligohydramnios or amniocentesis throughout pregnancy could not make it possible.

Treatment of either deformity with manipulation and casting has already been proven effective in literature, and the outcome of our results was comparable to others. The foot abduction orthosis has been widely used to maintain correction of clubfoot, not for SMA deformity. Reporting no recurrence in both sides encouraged us to consider using it in the future to maintain correction after treatment of isolated moderate metatarsus adductus deformity.

Our Medline search could not reveal any previous work that reported coincidence of both deformities.

We are aware about the limitations of this study. First, the treatment options offered for both deformities have already been explored on a larger scale. However, it is worth reporting such a rare presentation for the sake of diagnosis even if the proposed treatment's outcome was evaluated on different occasions before. Second, the sample size of the series is very small; the rarity of such presentation would take us very long time before collecting the proper sample. Third, no radiographic follow-up was done for both deformities. As Ponseti, we found that the foot's shape and dorsiflexion improved after few months; therefore, it was not necessary to obtain radiographs, particularly when interpretation of foot's radiographs in such a young age group would be difficult and more prone to intraobserver and interobserver reliability issues. Finally, the average duration of follow-up was 39 months only. Because most relapses in both deformities would happen in the first 2–3 years of life, and also correction of deformities was maintained for few months after finishing cast, it is unlikely for a full-blown deformity to recur in the future.

Conclusion

The potential coincidence of CIC and SMA can still happen, and each deformity should be manipulated differently. This in turn will need careful clinical examination of the ankle, subtalar, and midtarsal joints in newborns presented with foot deformities.

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Conflicts of interest

There are no conflicts of interest.

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