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Club Foot with Contralateral Congenital Amniotic Band Syndrome Amputation Successfully Treated with Ponseti Method: Case Report

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Learning Point of the Article:

The Ponseti method of serial casting remains the gold standard for clubfoot, even complicated by CABS and contralateral amputation. A custom fitted prosthetic allows for the use of a traditional bilateral AFO to allow for the best maintenance of the correction.

Abstract

Introduction: Constriction amniotic band syndrome (CABS) is a rare condition associated with the fibrous amniotic bands that restrict and ensnare the fetus in utero resulting in malformations at birth in one per around 15,000 live births. CABS associated with clubfoot, historically required extensive soft-tissue release due to its propensity for relapse.

Case Report: A 2-day-old Caucasian male infant born at 27 weeks gestation through emergency cesarean section due to concern for placental abruption and premature rupture of membranes in the setting of a prenatal history significant for oligohydramnios. The patient presented with non-viable tissue in the right leg requiring amputation with a left-sided clubfoot deformity. Following amputation of the right leg, the clubfoot was corrected with Ponseti method of serial casting and Achilles tenotomy. Three-week post-tenotomy and 6 months of age, a custom fit prosthesis of the right leg allowed for the application of a left abduction foot orthosis which maintained the correction.

Conclusion: This case study supports recent literature that CABS-associated clubfoot can be corrected with the Ponseti method. While a contralateral amputation can prevent the use of a traditional bilateral abduction foot orthosis, a custom fitted prosthesis can allow for its use and prevention of a relapse of deformity.

Keywords: Clubfoot, amniotic band syndrome, amputation, Ponseti casting, custom prosthesis.

Introduction

Constriction amniotic band syndrome (CABS) is a rare condition associated with the fibrous amniotic bands that restrict and endanger the limb and vasculature of the fetus in utero resulting in malformations at birth in one per around 15,000 live births [1]. The resulting malformations are associated with distal deformity of a limb, fusion of distal portions, or intrauterine amputation, cleft lip and palate, scoliosis, and limb length discrepancy [2]. The prevalence of CABS associated clubfoot has been observed in about 12–56% of patients presenting with CABS [2]. Clubfoot of any kind is typically identified through ultrasound, but rarely detected before the 16th week of gestation [1]. Clubfoot associated with CABS has historically been reported as treatment resistant because of a tendency for relapse of the deformity and reported rigidity [3, 4]. Treatment of CABS associated clubfoot often requires extensive soft tissue releases [1, 3].

Idiopathic club foot is treated by the Ponseti method of serial casting to correct the foot deformity. This has been the standard of care for idiopathic club foot, but favorable outcomes recently show effectiveness in treating non-idiopathic pathologies as well [3, 5]. The patient is serially casted and monitored carefully to

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Author's Photo Gallery


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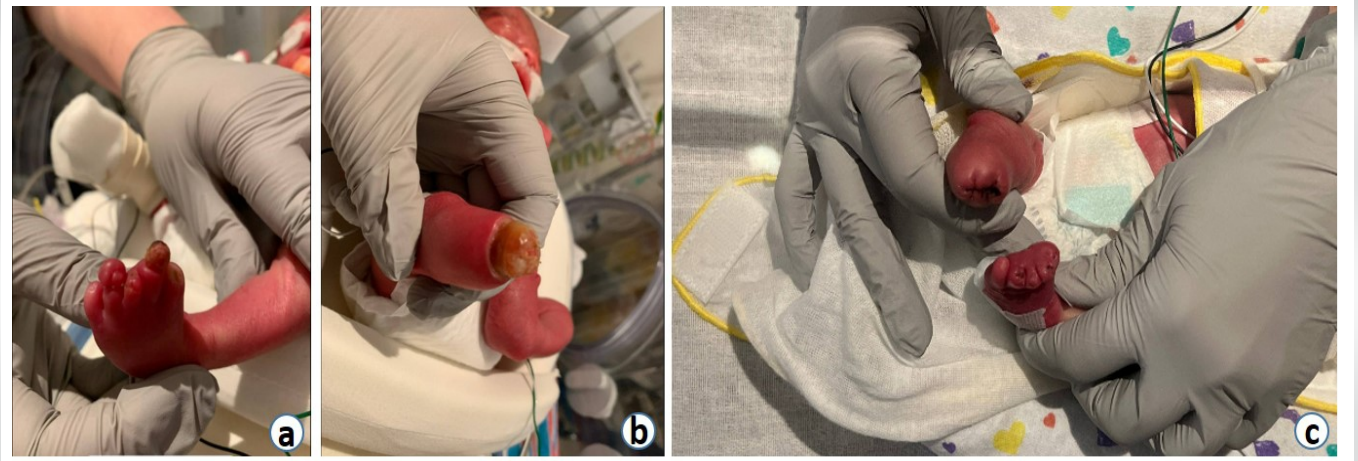

Figure 1: (a) Initial left foot presentation. (b) Initial right foot presentation. (c) Post-operative photo of patient following foot and toe amputation.

sequentially correct the clubfoot deformities: Cavus, adductus, varus, and finally equinus [3]. Following cast correction, the patient is placed in a foot abduction brace to prevent the recurrence of the clubfoot after casting. The initial bracing protocols begins with 23 h of use each day which tapers down to 12 h a day in the first few months to only worn at night after walking age [1]. In addition, Achilles tenotomy is often required if the equinus defect is persistent after casting and bracing [5].

Case Report

A 2-day-old Caucasian male infant born at 27 weeks gestation through emergency cesarean section due to concern for placental abruption and premature rupture of membranes in the setting of a prenatal history significant for oligohydramnios. On examination, the neonate exhibited evidence of congenital amniotic band syndrome and a constellation of orthopedic anomalies, including a congenital below-knee near amputation of the right leg with approximately 1 cm of nonviable residual limb tissue with exposed tibia and fibula. The left foot demonstrated complex syndactyly, with a nonviable appearance of the second and fourth toes, and a congenital talipes equinovarus deformity (Fig. 1a-c).

On physical examination, the left foot displayed syndactyly involving the second and fourth toes. These digits exhibited non-viability along with a talipes equinovarus deformity resulting in plantarflexion and inversion. The exposed bone and approximately 1 cm of nonviable tissue on the right lower extremity presented a significant risk for osteomyelitis development, particularly concerning given the neonate's prematurity. Delaying surgery for a few days would lower the risk of interventricular hemorrhage associated with general anesthesia. However, this delay could significantly elevate the

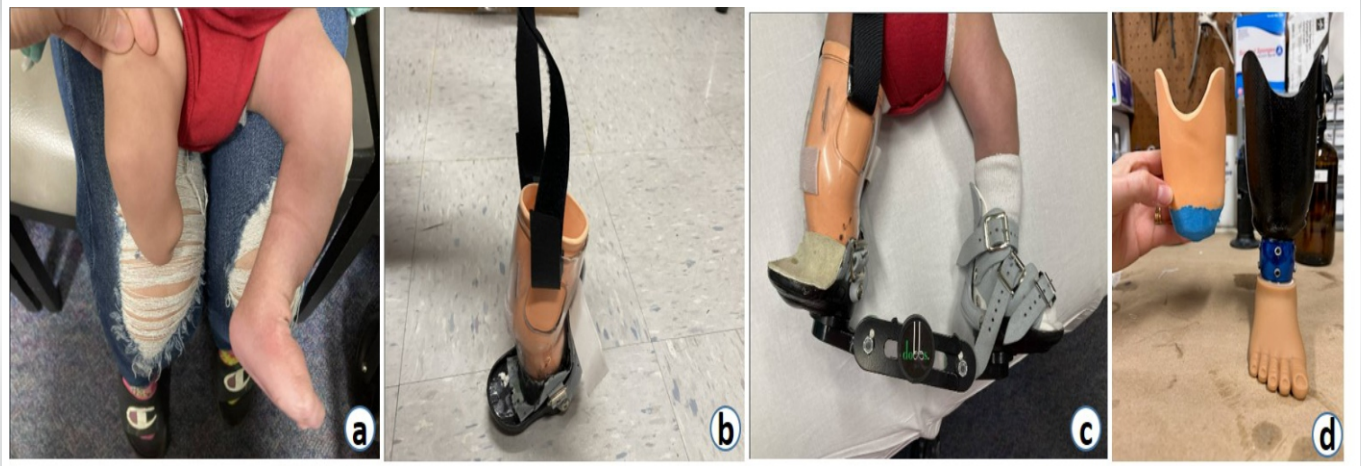

Figure 2: (a) Patient presentation in clinic at 8 months post operatively. (b) Initial right foot prosthesis for abduction boot and bar. (c) Patient with abduction boot and bar applied. (d) Custom fitted prosthesis for use without abduction boot.

Figure 3: (a and b) Patient presentation at the 9 month follow up visit in clinic.

risk of osteomyelitis development.

Following a comprehensive discussion with the neonatology team and the infant's mother, we elected to proceed with the immediate below-knee amputation (BKA) and debridement. The priority was minimized the risk of osteomyelitis and provided the optimal foundation for future prosthetic fitting due to the high likelihood of further surgery to address potential tibial and fibular overgrowth, a common occurrence in this age group [6].

Treatment

Amputation

Left foot (amputation of toes)

On the left foot, a gentle Esmarch tourniquet facilitated hemostasis while the nonviable 2nd and 4th toes were sharply excised with a scalpel. This approach prioritized both cosmetic outcome and adequate skin flap preservation for closure. Brisk bleeding from the amputation sites confirmed viable tissue, followed by copious irrigation, the wounds were closed with 6–0 chromic sutures.

Right BKA

Attention then shifted to the right BKA. An Esmarch tourniquet was again utilized for hemostasis. We identified 1 cm of nonviable tissue with exposed tibia and fibula and chose to amputate at the level of remaining viable soft tissue, approximately 1 cm proximal to the exposed bone ends. Nonviable tissue was meticulously excised using a rongeur, with careful sharp excision and neurovascular dissection. The tibia and fibula were shortened using a bone cutter, ensuring they were resected at a level with healthy bleeding tissue and minimal bone prominence. Bone ends were then smoothed with a rasp to facilitate closure.

A primary flap closure was achieved over the bone ends with low tension. The wound was irrigated and loosely approximated with chromic catgut sutures to allow for post-operative drainage to minimize the infection risk of a likely contaminated status of bone before surgery. Sterile dressings with petroleum gauze and stretch conforming gauze were applied to both amputation sites. The wound stump was mildly compressed with silk tape to minimize post-operative edema.

Ponseti casting and prosthesis

At the 3-month post-operative visit, the amputation site was healing well, with no evidence of overgrowth. The left foot continued to demonstrate a clubfoot deformity, to which we proceeded with the Ponseti method of casting correction [3]. The initial casting aimed to correct the cavus deformity by aligning the first metatarsal. In subsequent visits casting correction continued by maintaining the cavus correction and simultaneously correcting the adductus and varus deformities [3]. Starting on the 2nd week of casting, the patient had 0° of abduction which improved to 25° in the 3rd week. Finally, the neonate was able to passively abduct to 70° by the 4th week (Fig. 2a-d).

Achilles tenotomy

After 4 weeks of casting, the patient returned for an Achilles tenotomy to aid the correction of the equinus deformity. The tenotomy was performed under general anesthesia. A Beaver blade (BVI, Waltham, MA, USA) was introduced from the medial aspect of the Achilles tendon above the calcaneal apophysis. It was gently turned to the anterior aspect of the Achilles tendon. The ankle was held in maximum dorsiflexion while the beaver blade was passed through the tendon. Immediate improvement of dorsiflexion to 15–20° past neutral was seen. The wound was irrigated and closed with a wound closure strip. A dressing of petroleum gauze and gauze was applied. A well-padded and molded long leg Ponseti cast was applied. The orthotist measured and molded a fitting for the right residual limb to incorporate into the planned foot abduction brace.

Custom foot abduction brace

Three weeks following his heel cord tenotomy surgery (6 months old), the neonate was fitted for a custom foot abduction brace. Parents were instructed to have the patient wear the brace for 23 h a day until he begins to stand. The patient had no pain reported in his lower extremities at follow-up. The parents did however loosen the straps of the Mitchell sandal because it seemed to restrict circulation in his left foot, but the sandal itself remained secure. The parents expressed concerns about the fit of the foot abduction brace with the prosthesis, although there were no signs of brace related abrasions on the lower extremities. The 6-week follow-up visit showed an improvement of the left ankle dorsiflexion improved to 15° past neutral (Fig. 3a and b).

Discussion

Ponseti casting effectiveness

In our case, we successfully treated a case of clubfoot complicated by an amputation secondary to CABS. Bilateral clubfeet has been treated successfully by Ponseti casting which is today the standard of care. CABS-associated clubfeet have been proven difficult to manage in the past and had been seen as treatment resistant because of the propensity for multiple relapses [1]. Until recently, patients with CABS-associated clubfeet had largely been managed by extensive soft-tissue release [1]. The clubfeet associated with the constriction bands was challenging because they were reported to be more rigid than idiopathic clubfeet and requiring repeated surgical corrections and higher complication rates [2]. More recently, many have found success in treating CABS-associated clubfoot with Ponseti casting [1, 3, 5, 7].

Shabani was able to treat a 5-day old with bilateral clubfoot and CABS fixed with Ponseti casting followed by heel cord tenotomy. In their case, they reported using the Ponseti method with a serial casting correction of the clubfoot deformities, followed by the foot abduction brace to maintain the correction. This included 23 h a day external rotation for the 1st 3 months tapered off to 12 h a day until the patient could walk when they were switched to nightly. The correction was maintained with no relapse and the patient was released at 2 years old [1]. In a 12-year retrospective study by Carpiaux et al., they observed the effectiveness of the Ponseti method correction in twelve patients with CABS associated clubfeet. They examined twelve patients with a total of 21 clubfeet, nine bilateral and three unilateral, for an average of 3.9 years. Out of the 12 patients maintained their corrections [3]. For this case series, their team was able to correct the clubfoot deformities through serial casting and bracing alone without the need for surgery, including in patients with neurological deficit [3]. In another case series, three patients were managed by Agarwal et al. primarily with the Ponseti method, but patients in this study did require surgery to fix the equinus deformity. They noted that a unique challenge posed in serial casting correction of patients with constriction band syndrome is the variability in toe anatomy and vascularity. Differences in the presence or absence of toes place additional challenges in manipulating the foot for casting. Changes from typical vascularity make it additionally challenging for parents to monitor for adequate foot perfusion. Additional education for parents to remove the cast when they have any doubt of decreased vascularity was beneficial [2]. Finally, they noted that to prevent cast slippage, more knee flexion and a slightly tighter fitting cast was applied.

Zionts and Habell reported a case series of five patients with one patient similar to our case. One of the patients was born with a contralateral amputation of the limb, requiring the use of a unilateral foot abduction orthosis rather than a conventional foot abduction orthosis [7]. This patient was managed in a custom molded knee ankle-foot orthosis due to a congenital amputation of the contralateral lower extremity [7]. In their study, Zionts and Habell followed their patients for a mean of 32.6 months. They found that CABS associated deformities were more difficult to treat than idiopathic deformities, and a relapse occurred in 4–5 patients attributed to nonadherence [7]. They also noted that with immediate serial cast application and correction, response to treatment was similar to that reported in idiopathic clubfoot.

Prosthetic management with ankle foot orthotics (AFO)

A key step of the Ponseti correction is the maintenance of correction through an abduction foot orthosis. In our patient, this was particularly challenging given the amputation of the right foot at birth. It was observed that bilateral abduction braces have had more success in maintaining corrections [8]. Alternatives for cases where a bilateral AFO cannot be used have been explored in the literature. One challenge for bilateral AFO is one of adherence to the bracing protocol is difficult and patients can resist their use [9]. In a 2011 study of 27 kids with 35 idiopathic club feet, George et al. offered a unilateral above the knee orthosis over the traditional bilateral AFO. Their team noted that the unilateral orthosis was preferred over the bilateral brace but did incur a higher recurrence rate [9]. Twenty-three of the total 27 parents reported that unilateral orthosis was easier to use and the children were less bothered by the above the knee orthosis versus the traditional bilateral AFO [9]. Another study hypothesized an increased patient adherence to bracing times

and reduced deformity relapse when using a unilateral AFO was conducted by Sætersdal et al. In this study, outcomes in 94 children (133 feet) between unilateral versus bilateral bracing outcomes were compared. The findings also supported a better perceived adherence to the bracing protocol, as the parents and patients were more receptive to it versus a bilateral brace; however, bilateral bracing had superior parent reported outcomes in addition to more flexible feet on examination than the unilateral bracing group [8].

Another attempt at a unilateral AFO was reported by Solanki et al., who studied the effectiveness of modified ankle foot orthosis fabricated from low-temperature thermoplastics [10]. They followed a group of 28 infants with idiopathic clubfeet for 6 months who were using a unilateral AFO which were custom molded [10]. In this study, there was no significant difference in the Pirani and modified Dimeglio scores in the posttreatment results in cases with and without Achilles tenotomy [10]. They pointed to this as a viable alternative in cases of both unilateral and bilateral idiopathic clubfeet.

Conclusion

In our case, we successfully treated a case of clubfoot complicated by a contralateral leg amputation secondary to CABS. Bilateral AFO's have superior outcomes over unilateral AFO's at preventing relapse of the correction. A custom fitted prosthetic allowed the application of a bilateral AFO for the maintenance of a Ponseti method correction of the clubfoot.

Clinical Message

CABS-associated clubfoot historically required extensive soft-tissue release due to its propensity for relapse. This case study supports recent literature that CABS associated clubfoot can be corrected with the Ponseti method. While a contralateral amputation can prevent the use of a traditional bilateral abduction foot orthosis, a custom fitted prosthesis can allow for its use and prevention of a relapse of deformity.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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