

Naviculectomy for two ambulatory children with intractable congenital vertical talus: redefining the indications of an old technique

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Congenital vertical talus is a rare and complex foot anomaly. Serial casting with or without minimally invasive surgery is a universal management strategy especially for children in the first year of life. Nevertheless, extensive surgical treatment of late-presenting, neglected and multiple operated children with congenital vertical talus may be required with guarded results. The results of naviculectomy as a more conservative intervention and directed exclusively at ambulatory children with intractable congenital vertical talus have not been reported. We present the radioclinical outcomes of two ambulatory children with intractable congenital vertical talus treated by naviculectomy/midtarsal resection and limited soft tissue release. One child had an isolated congenital vertical talus whereas the other had a non-isolated etiology. Generally, naviculectomy/midtarsal resection revealed a positive benefit-risk profile in children with intractably severe congenital vertical talus on the

short-term. We reported favorable results in terms of foot appearance, function and radiology. We believe that a less invasive procedure like naviculectomy/midtarsal resection is an encouraging technique to investigate in children with intractable congenital vertical talus. *J Pediatr Orthop B* 29:387–391 Copyright © 2019 The Author(s). Published by Wolters Kluwer Health, Inc.

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Introduction

Congenital vertical talus (CVT) is a rare and complex foot anomaly. It can present as an isolated foot anomaly or as a nonisolated anomaly, namely, in association with neuromuscular disorders such as arthrogyposis and myelomeningocele, and chromosomal aberrations [1]. The clinical diagnosis is largely based upon the presence of a rigid hindfoot equinus and valgus, and forefoot dorsiflexion and abduction [1]. On lateral radiographs, the talocalcaneal angle, talar-axis-first metatarsal base angle (TAMBA) and tibio-calcaneal angle can verify the diagnosis and confirm the rigidity of all components of the deformity [1]. Studies have demonstrated that the manipulative technique with minimally invasive surgery is superior to extensive soft tissue release in terms of surgical trauma inflicted and subsequent ankle range of motion and foot pain on the long term [1–3]. Although some have advocated the use of the manipulative technique with minimally invasive surgery as a first-line treatment irrespective of patient's age and subcategory, there is a universal agreement in the literature that this technique is most suited to children younger than two years [4–7]. Nevertheless, some patient subsets with

CVT may not fall under the above-mentioned treatment recommendations and may be more amenable to salvage procedures such as naviculectomy. Historically, naviculectomy was practiced as a routine procedure for CVT children above the age of three months irrespective of ambulatory status and CVT subtype [8]. The intermediate term results were satisfactory [8]. In fact and particularly after the recognition of the successful results of manipulative casting techniques, the practice of naviculectomy seemed to fade in regard to ambulatory CVT children. Additionally, naviculectomy was practiced on four nonambulatory CVT children with irreducible talonavicular joint following a classic soft tissue release. Naviculectomy achieved favourable short-term results [9]. Likewise, naviculectomy has been employed in conjunction with lateral column shortening to treat children with severely rigid pes cavovarus caused by multiply operated isolated congenital club feet and arthrogyposis where the classic soft tissue releases and osteotomies were deemed unlikely to achieve a plantigrade foot [10]. Longstanding congenital dislocations are likely to undergo secondary adaptive bony changes, thus making anatomic reconstruction unachievable. Furthermore, the extensive surgical trauma required to achieve a plantigrade foot in postsurgical recurrence and certain neuromuscular subtypes of CVT and club feet alike is fraught with serious complications that usually compromise the overall functional outcome of surgery [1,3]. We postulate

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that naviclectomy as a salvage procedure may present a workable and surgically affordable option for a carefully selected subset of complex CVT, where the risks incurred through extensive surgical release are expected to overpower the potential benefits of surgery. Concomitantly, it may avert the need for revision surgeries. There are insufficient literature citations to properly inform decision taking in regard to the indications and outcomes of salvage procedures particularly in ambulatory children with CVT. This study includes two young ambulatory children (two feet) with CVT. The first patient had a neglected isolated CVT that was never subjected to manipulation and the other patient had arthrogyposis and bilateral CVT that was multiply and extensively operated together with unilateral avascular necrosis of the head talus. The objective of this study is to evaluate the short-term radioclinical outcomes of naviclectomy/midtarsal resection with limited tailored releases in patients with complex CVT together with a comprehensive literature update. Additionally, we will detail the particulars of the surgical techniques implemented.

Case 1

A four-year-old boy presented to our paediatric orthopaedic outpatient clinic with a neglected CVT of the right foot. The child's mother provided neither history of previous serial casting nor surgery. The child was neurologically and systemically intact. On examination, he exhibited a rigid hindfoot equinus and valgus, and forefoot dorsiflexion and abduction with extremely convex sole. The foot radiographs revealed marked abnormalities of the talocalcaneal angle, TAMBA and tibiocalcaneal angle on the lateral view and of the talocalcaneal angle and the talar axis-first metatarsal angle on the anteriorposterior view (Fig. 1a–d).

Surgical technique

We decided to subject the patient to a conservative soft tissue release with navicular excision. After tourniquet application in the supine position surgery was performed in the following sequence. A direct medial incision over the talar head extended slightly distal. Through it we

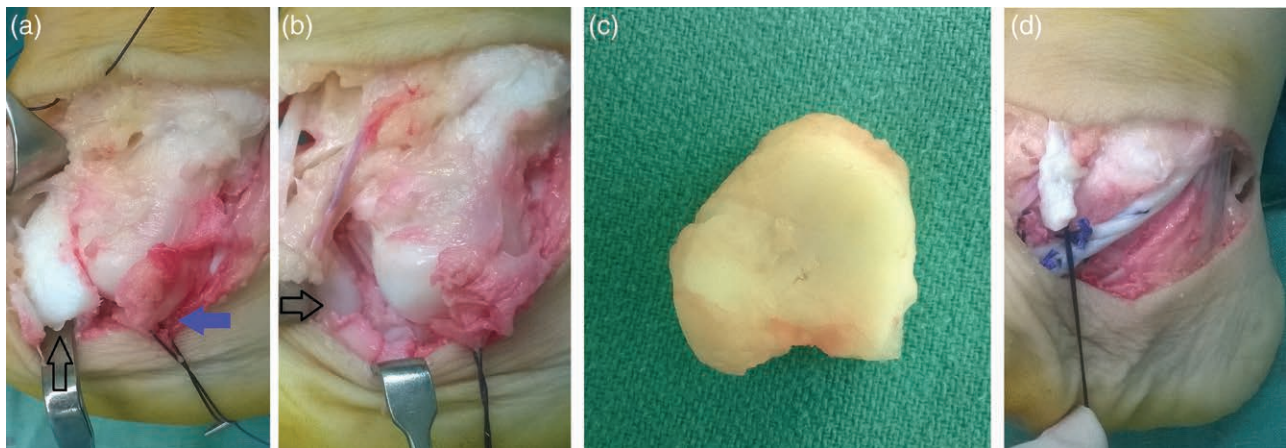
circumferentially released the talonavicular joint, the plantar calcaneonavicular ligament, and we disinserted the bony attachments of the tibialis anterior and tibialis posterior tendons in preparation for later rerouting after optimal foot correction. Then, all the bowstringing digital extensors including the peroneus tertius were released through mobilizing a small transverse incision over the anterior ankle. Then, we performed a curved incision over the cuboid through which the extensor digitorum brevis origin, the peroneus brevis tendon and capsule of the calcaneocuboid were released. Then, we tenotomized the tendon Achilles and released the posterior capsule of the ankle through a small direct transverse incision above its insertion. An attempted reduction of the talonavicular joint was unsuccessful and tensioned the neurovascular bundle. Therefore, we performed a subperiosteal naviclectomy. We took a retrograde K-wire through the cuneiform and first ray and foot alignment was restored by maximum forefoot adduction and flexion through the talo-cuneiform pseudoarthrosis, and concomitantly upward pressurizing the talar head. The correction was maintained by advancement of the K-wire proximally. The tibialis anterior tendon was reattached as medial and plantar as possible to the plicated remnants of the talonavicular capsule. Likewise, the tendon of the tibialis anterior was advanced and secured further medially (Fig. 2a–d). We checked foot alignment by image intensifier in orthogonal planes. A posterior slab was applied for 4 weeks after which a long leg cast was used for another 8 weeks. At the final follow-up, the patient was pain free. Hindfoot valgus and forefoot abduction were fully restored and the sole convexity disappeared. A residual flexible flattening of medial arch was noted which was compatible with normal pes planus frequently seen in that age group. Over the course of follow-up, the patient developed progressive tightness of the digital foot extensors with gradual limitation of ankle plantar flexion and excessive forward bending of the tibia which caused gait difficulties especially on running. On stance, an extension deformity of the second and third digits was noticed. The postoperative radioclinical outcome at two years follow-up are shown (Fig. 3a–d).

Fig. 1



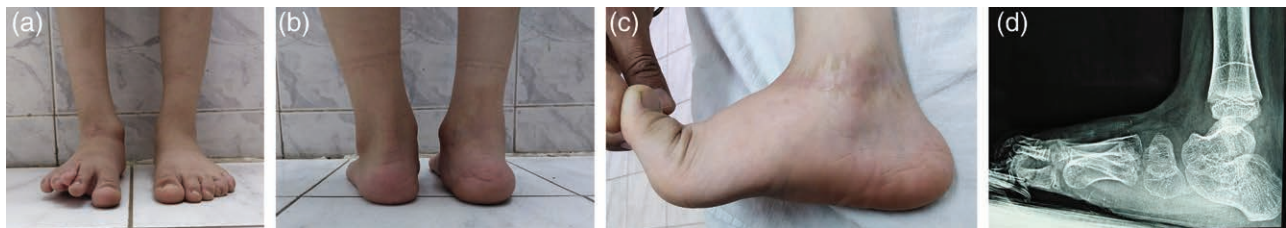
Radioclinical outcome of a four-year-old boy with neglected isolated congenital vertical talus. (a)–(c) Note the right foot exhibits forefoot dorsiflexion and abduction and an extremely convex sole with heel valgus. (d) Lateral foot radiographs show a severely abnormal talar-axis-first metatarsal base angle and tibiocalcaneal angle.

Fig. 2



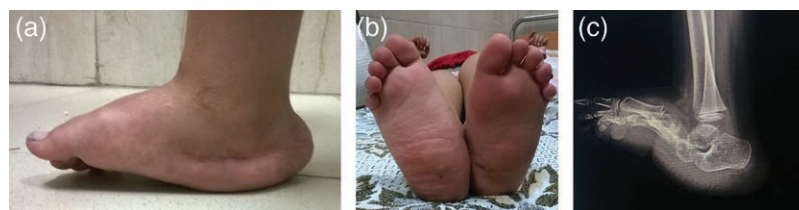
Intraoperative images of direct medial approach over the talonavicular joint. (a) The irreducible navicular (black arrow) after disinsertion of tibialis posterior (not shown) is seen articulating with the dorsum of the talar head (blue arrow). (b) After resection of the navicular. Note the articular cartilage of medial cuneiform. (c) The navicular bone. (d) Advancement of the tibialis posterior tendon and rerouting of the tibialis anterior tendon (black sutures).

Fig. 3



The radioclinical results at two-year follow-up. (a) and (b) Resolution of forefoot abduction and heel valgus. (c) Note flexibility of medial arch. (d) Restoration of talar axis first metatarsal base on standing lateral radiographs.

Fig. 4



A five-year-old girl with nonisolated congenital vertical talus. (a)–(c) Radioclinical relapse (incomplete correction) three years after first revision open reduction surgery on right foot.

Case 2

A five-year-old girl presented with a neglected bilateral CVT secondary to arthrogyripos multiplex congenita. At six months of age, she received bilateral simultaneous open soft tissue release through multiple incisions at another hospital. Early relapse of both feet deformity occurred together with avascular necrosis of the left head talus. She was reoperated at 9 months and one year of age on the left

and right foot, respectively. At five years of age – time of presentation to us – the right foot had developed a second recurrence and subsequent painful callosities corresponding to the plantar location of the talar head. The left foot was fairly corrected and asymptomatic in regard to radioclinical parameters except for slight residual sole convexity and heel valgus. The child had mild flexion deformity of her knees, but overall she had no functional limitations.

Fig. 5



(a)–(c) Radioclinical improvement one year following our index surgery on the right foot.

Surgical technique

On the right foot, we performed a naviculocuneiform resection through a direct medial incision over the mid-tarsal region, and limited releases of the tendon Achilles, and posterior ankle capsule through a direct incision. We released the extensor digitorum brevis and the calcaneocuboid capsule through a lateral incision. We restored the foot arch and maintained it with a retrograde K-wire. We tightened the capsuloligamentous remnants on the medial ‘talo–first metatarsal’ pseudoarthrosis. We subjected the left foot to tendon Achilles tenotomy and posterior ankle capsulotomy. At the final follow-up, the patient was pain free. Hindfoot valgus and forefoot abduction were fully restored and the sole convexity disappeared. A residual semirigid flattening of medial arch was noted. We noted residual subluxation of calcaneocuboid joint on plane radiographs. The postoperative radioclinical outcome at two years follow-up is shown (Figs. 4a–c and 5a–c).

Discussion

The surgical treatment of isolated and nonisolated CVT under the age of two years often follows a predictable and favourable course [1,2,4]. The current literature does not allow for strong treatment recommendations on how to manage nonisolated, recurrent and/or late-presenting ambulatory CVT children above the age of two years. It is believed that the use of serial casting with or without minimal soft tissue release in such cases will only make the likelihood of recurrence or undercorrection greater [1–3,11]. Likewise, the use of extensive soft tissue release in such cases has a tremendous potential in developing perioperative and delayed complications [1–3,9,11,12]. We believe that the scale of these complications may be underreported in the orthopaedic literature and it is likely attributed to publication bias. This is because there is a general untoward tendency to publish studies with significant results and leave studies with insignificant or negative results [13]. There are striking similarities between congenital talipes equinovarus and CVT in

regard to the complication potential following extensive soft tissue release for the nonisolated, recurrent, multiply operated and/or simply late-presenting categories above the age of two years [12,14,15]. In light of the growing consensus that the current treatment recommendations for such CVT subtypes are vague, we proposed our new strategy, namely, naviculectomy/mid-tarsal resection with limited soft tissue release. This treatment strategy entails a tradeoff between anatomic foot reconstruction with a high complication potential on one hand and simply restoration of foot function with a relatively low complication potential procedure on the other hand.

Naviculectomy versus talectomy

Talectomy has been used to address severe and rigid cases of congenital talipes equinovarus, mainly of the nonisolated types [16]. Resection arthroplasty has been advocated for nonambulatory children with CVT in the form of naviculectomy or talectomy [8,9,17]. And both types of resection arthroplasty have achieved satisfactory results for these nonambulatory children who were mainly nonisolated CVT [9,18]. However, we disagree about the use of talectomy for ambulatory counterparts of children with CVT. The talar bone is fundamental to optimal dynamic stability, kinetics and kinematics of the ankle joint and foot. Experimental studies have indicated that around 83% of forces in the ankle are transmitted through the tibiotalar joint, with the remaining 17% are transmitted through the fibula [19]. Talectomy is likely to significantly compromise the above gait parameters, especially in children already burdened by neuromuscular disorders. Although these potentially untoward effects may not be well pronounced in children with low ambulatory potential, it is certainly a point of central importance to children with high ambulatory capabilities and greater functional demands irrespective of the CVT subtype. Both of our patients were independent community ambulators. Besides, the functional and objective clinical outcomes of naviculectomy/mid-tarsal resection in our series were satisfactory. Because of the above reasons

we see no grounds for recommending talectomy to fully ambulatory patients with CVT like our two children. Thus, we believe that naviculectomy/midtarsal resection represents a functionally more conservative alternative to talectomy. It is noteworthy that we successfully implemented naviculectomy as both a primary procedure and a salvage procedure. This widens its indications and emphasizes its importance.

The radiologic parameters showed restoration of the TAMBA for our two children. A residual calcaneocuboid subluxation was demonstrated in case 2. It did not seem to have a functional impact. We attribute the progressive loss of plantar flexion and subsequent increase in the ankle dorsiflexion noticed in case 1 to an over lengthened Achilles tendon. We scheduled that patient for percutaneous release of the digital extensors with simultaneous shortening of the Achilles tendon.

Conclusion

Because we targeted patients of a certain subcategory of CVT, there is a plausible explanation why is our sample size is small. Yet, this limits the generalizability of our conclusions. The findings of this study illustrate that naviculectomy/midtarsal resection with limited soft tissue release can yield satisfactory short-term radioclinical outcomes in ambulatory children with multiply operated and/or neglected CVT. These findings have suggested a future avenue of a less invasive surgical management strategy for ambulatory children with resistant CVT. These subcategories of CVT children are otherwise left with treatment options that have a high complication potential or are functionally more depreciating. Long-term follow-up studies are more suited to settle the dispute as to what is the best evidence for the various surgical practices for children with intractable CVT.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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