Can Selective Soft Tissue Release and Cuboid Osteotomy Correct Neglected Clubfoot?

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Abstract

Background Neglected clubfoot in older children is characterized by a stiff, nonreducible deformity with relative elongation of the lateral column of the foot with respect to the medial column. Surgical correction often has involved a double osteotomy with elongation of the medial column and shortening of the lateral column, or the use of an external fixator to achieve more gradual correction. Both approaches have shortcomings.

Questions/purposes We therefore (1) used objective physical examination measurements and a functional assessment to evaluate the effectiveness of cuboid osteotomy combined with a selective soft tissue release to achieve correction of neglected clubfoot in older children, (2) determined the rate of complications, and (3)

ascertained whether the initial correction achieved was maintained.

Methods We reviewed 31 patients (56 feet) older than 5 years with severe, neglected nonreducible clubfoot deformity who underwent the index procedure. Minimum followup was 2 years (average, 6 years; range, 2–9 years). Postoperatively, the Laaveg and Ponseti classification and Dimeglio score were used to grade correction. Complication rates were tallied.

Results According to the Laaveg and Ponseti classification, 24 feet showed excellent correction, 20 good, nine fair, and three poor at 1-year followup. These results were maintained up to the latest followup. Patients showed significant improvement of Dimeglio score after surgery ($p < 0.0001$). Two patients had postoperative skin-related complications that healed without additional surgery.

Conclusions Cuboid subtraction osteotomy combined with posteromedial release is an effective approach to manage a stiff nonreducible neglected clubfoot deformity in older children.

Level of Evidence Level IV, therapeutic study. See Guidelines for Authors for a complete description of levels of evidence.

Introduction

Clubfoot is a congenital deformity estimated to occur in 150,000 children each year worldwide, with 80% of these

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Each author certifies that his or her institution approved or waived approval for the human protocol for this investigation and that all investigations were conducted in conformity with ethical principles of research.

This work was performed at The Rizzoli Institute (Bagheria, Italy) and University Campus Bio-Medico of Rome (Rome, Italy). Surgery was performed at the Mlali Children Hospital (Kongwa, Tanzania).

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in low- and middle-income countries [6, 9, 36, 37]. Non-operative treatments provide lasting correction in more than 80% of patients in which treatment is begun in the first 2 years of life [8]. In many countries, however, patients do not have access to medical treatment at an early age, and present only after walking age with what has been termed neglected clubfoot [13, 37]. Many children with clubfoot in low- and middle-income countries do not receive treatment and eventually have neglected clubfoot develop, which represents a cause of societal stigma, limiting their participation in activities ranging from school attendance to work and marriage [36]. If the foot is not corrected by walking age, the effect of weightbearing worsens the equinus and varus deformities, making the clubfoot non-reducible [37]. Eventually, the lateral column of the foot (calcaneus plus cuboid) elongates more than the medial one (talus, navicular, and cuneiform) and renders the deformity stiff [28, 38]. Severe neglected clubfoot is associated with pain and difficulty in locomotion; moreover, recurrent skin breakdown with infections is observed, which sometimes can lead to amputation, especially in patients who are unable to wear shoes [37].

Soft tissue procedures previously were developed with the aim of releasing tendons, capsule, and ligaments [34, 48]. In 1905, Codivilla [1] conceptualized his own surgical technique, whose modifications are known as extensive soft tissue release (STR); the Turco technique is one such approach [46]. STR can be used to correct some cases of neglected clubfoot [20, 46, 48], but in patients with more severe deformities as a result of the lateral column being longer than the medial one, STR allows for only partial correction of the deformity. In addition, extensive STRs are known to predispose the clubfoot to vascular complications related to its congenitally reduced vascular supply, leading to local tissue necrosis [24]. For this reason, some authors advocate a combined procedure of bony osteotomies with shortening of the lateral column and lengthening of the medial column as the most logical approach to address the forefoot deformities seen in more rigid neglected clubfeet [31]. However, this approach is associated with some drawbacks, including that it does not address the rigid hindfoot deformities present in most neglected clubfeet, and that it requires a mature ossific nucleus of the medial cuneiform, which usually is not present until the patient is 4 years of age [35]. Moreover,

the excised cuboid bone is sometimes soft or the wedge is too small to support the needed lengthening of the medial cuneiform. Finally, graft wedge extrusion after the medial cuneiform elongation osteotomy can complicate this approach and require revision surgery [24, 30]. To overcome such problems in children older than 5 years with neglected clubfoot, we describe an approach consisting of a single-step combination of a modified STR with the addition of a cuboid subtraction wedge osteotomy. This procedure has the theoretical advantage of providing good correction but with less bone work than traditional approaches to this problem, and without sacrifice of any joint at the medial column. Moreover, we believe this can be performed safely without the need for bone-grafting procedures, as the medial cuneiform is not addressed.

In this study, we therefore (1) used objective physical examination measurements and a functional assessment to evaluate the effectiveness of cuboid osteotomy combined with a selective STR to achieve correction of neglected clubfoot in older children, (2) determined the rate of complications, and (3) ascertained whether the initial correction achieved was maintained.

Patients and Methods

We conducted an observational study on a patient population of children affected with congenital clubfoot who presented to our institution. Between 2000 and 2009, we operated on 469 patients affected by congenital clubfoot. Sixty-seven of 469 patients had surgery for rigid neglected untreated clubfoot with an available followup of at least 2 years. Of these, we report on 31 of the 67 patients (56 feet) older than 5 years at the time of initial treatment (average age, 7 years; range, 6–9 years) who underwent STR plus cuboid osteotomy (Table 1). Of the remaining 36 patients, 17 of 36 were lost at followup, and 19 of the 36 underwent another kind of surgery because of different age range. Minimum followup was 2 years (average, 6 years; range, 2–9 years). The Mlali Children Hospital in Kongwa, Tanzania, was built with donations and private funding and has been active since 1999 in creating a structured environment to deliver orthopaedic surgery in the disadvantaged areas of Tanzania where poor economic resources preclude treatment of most of the resident rural

Table 1. Characteristics of the study population receiving posteromedial release plus cuboid osteotomy

Number of patients	Age at initial treatment (years)*	Involvement (number of feet)				Followup (years)*
		Total	Right	Left	Bilateral	
31	7 (6–9)	56	4	2	50	6 (2–9)

* Values are expressed as mean, with range in parentheses.

population. Since its opening, approximately 2000 children have been treated surgically there, and missions are performed for 2 consecutive weeks, six times a year. However, owing to the difficult context, not all patients are available for clinical followup in a regular fashion. Of the available 31 patients, six had unilateral and 25 had bilateral clubfoot involvement. No patients had casting or any corrective surgery before treatment at our institution.

All patients' feet were evaluated preoperatively in terms of skeletal maturity by preoperative radiographs and to assess the degree of rigidity using the classification systems of Dimeglio et al. [7]. The score by Dimeglio et al. [7] is based on the assessment of four parameters, which are categorized in terms of reducibility with gentle manipulation and measured with a goniometer: (1) equinus deviation in the sagittal plane, (2) varus deviation in the frontal plane, (3) derotation of the calcaneopedal block in the horizontal plane, and (4) adduction of the forefoot relative to the hindfoot in the horizontal plane. To each one of the four parameters, a score is assigned on a four-point scale (4 points = reducibility from 90° to 45°; 3 points = reducibility from 45° to 20°; 2 points = reducibility from 20° to 0°; 1 point = reducibility from 0° to -20°; and 0 points = reducibility of less than -20°). Single scores then are summed up to a maximum of 16 points. One point each is added for a posterior or medial crease, in case of evidence of plantar retraction or cavus deformity or in case of poor muscle condition. The feet then are classified into four categories with respect to the severity of the deformity: Grade I = greater than 90% reducible; score = 0 to 5 points; Grade II = moderate deformity, score = 5 to 10 points; Grade III = severe deformity, score = 10 to 15 points; and Grade IV = very severe deformity; score, 15 to 20 points; arthrogryptic appearance. In our cohort, all patients had severe or very severe deformity, with 12 feet presenting with Dimeglio Grade III deformity and 44 feet with Grade IV deformity (Table 2).

Surgery in all patients consisted of a modified STR as already described [1, 2], with the addition of a cuboid shortening wedge osteotomy. The surgery can be thought of as having three phases: (1) varus-adduction correction by the STR (Video 1; supplemental materials are available with the online version of CORR®); (2) shortening of the

lateral column by performing a superolateral wedge cuboid subtraction osteotomy (Video 2; supplemental materials are available with the online version of CORR®); and (3) correction of equinus and supination by elongation of the Achilles tendon and a posterior capsulotomy of the ankle and subtalar joint (Video 3; supplemental materials are available with the online version of CORR®).

In the first phase, a posteromedial incision is performed along the medial side of the Achilles tendon, curving below the medial malleolus to the base of the first metatarsal. Careful dissection under direct vision is performed to avoid traumatizing soft tissues and articular surfaces of the tarsal bones. The anterior tibial tendon sheath is gently sectioned, and the tendon is exposed on the medial plantar surface of first cuneiform and protected by a retractor. The posterior tibial sheath is gently excised; the section is performed around the medial malleolus as far as the navicular bone to expose the tendon. Without damaging the insertion on the navicular bone, the posterior tibial tendon is elongated by the z-lengthening technique. The flexor digitorum longus tendon is located just below the posterior tibial tendon. To better identify it, traction on the tendon is performed. The tendon is sectioned and elongated by a z-plasty. The tendon is sutured with the varus deformity of the foot reduced. On the posterior aspect of the feet, the neurovascular bundle is isolated. The flexor hallucis longus tendon is identified below the neurovascular bundle. Its traction produces flexion of the big toe. The tendon is lengthened using the z-technique and sutured with the foot in the reduced position, proximal to the knot of Henry. Attention is paid that the abductor hallucis tendon and muscle are not confused with the brevis hallucis flexor tendon and muscle; flexion of the big toe does not produce movement of the abductor hallucis tendon and muscle. The superficial and deep muscle belly are released and elongated using the z-lengthening technique. For better identification of the talonavicular joint, the distal fibers of the tibial posterior tendon are followed as far as the insertion on the navicular. The tibionavicular fibers of the superficial deltoid ligament are sectioned and the tibial anterior tendon retracted. The talonavicular ligaments are excised superficially. To complete the incision deeper, the blade is oriented in the direction of the talar head; an improper direction can damage the talus neck. The reduction of the navicular is completed by using a curved elevator. This release better corrects the varus deformity, although still being still incomplete (Fig. 1).

In the second phase of the surgery, a thin K-wire passing from medial to lateral is used to detect the talonavicular joint on the lateral aspect. The lateral approach to the varus deformity in the cuboid osteotomy is used to shorten the lateral column of the foot. A lateral incision is performed right by the cuboid bone. The dissection is performed superiorly to the brevis peroneal tendon and inferiorly to

Table 2. Preoperative and postoperative grading according to Dimeglio et al. [7]

Dimeglio grade	Preoperative	Postoperative at last followup
I (benign)	0	39
II (moderate)	0	17
III (severe)	12	0
IV (very severe)	44	0

Feet are evaluated separately (number of feet = 56).

Fig. 1A–F (A) The blade is orientated in the direction of the talar head during deep incision of the talonavicular ligaments. (B) Reduction of the navicular is performed by using a curved elevator that is inserted in the direction of the blade, and (C) then gentle reduction is performed. The drawings show the reciprocal positions of

the navicular and talus after reduction. (D) This drawing schematically shows the direction of the elevator while entering the talonavicular joint on the axial plane; (E) the elevator then is advanced and (F) rotated (red arrow) to perform the reduction.

the brevis extensor digitorum muscle. The brevis extensor digitorum muscle is retracted superiorly. Release of the bifurcate and interosseous ligament is performed using a curved elevator. The release of the talonavicular ligament is completed to perform the reduction of the talonavicular joint. The cuboid bone is isolated. The osteotomy is performed using an oscillating saw. A small wedge with the base on the superolateral aspect is removed. The wedge osteotomy of the cuboid bone achieved shortening of the lateral column; this process permits complete reduction of the varus and the adduction deformity of the foot.

After complete correction of the varus deformity, the third phase of the surgery, the posteromedial approach to correct the equinus and supination, begins. The Achilles tendon is excised and elongated using the z-technique. The foot is gently forced in talus and, if excessive resistance is seen, by protecting the neurovascular bundle with a retractor, it is possible to identify the posterior ankle capsule that is released with a blade. If residual supination deformity is observed after correction of equinus, a gentle opening of the subtalar joint is performed. As the foot achieves the neutral position, the elongated Achilles tendon is sutured.

After surgery, patients in this series have been immobilized in an above-knee cast in hypercorrection (talus-valgus-pronation) and left nonweightbearing for 6 weeks. After that period, a below-knee cast is placed to allow the patient to ambulate with crutches for the following 6 weeks. At 3 months, the cast is removed and the child is allowed unprotected ambulation during the day and use of a night ankle-foot splint in corrected position up to the end of growth.

Patients were reassessed at discharge at approximately 3 months after surgery, and then at variable followups depending on the availability of the parents and patients. At

final available followup, the patients' age averaged 14 years (range, 13–15 years). Reported clinical evaluation represents patients' assessments at last followup. The clinical outcome was measured qualitatively by visual assessments and by comparing preoperative and postoperative adduction, supination, and hindfoot varus through calculation of the Dimeglio score at latest followup.

Surgical results were evaluated in terms of recurrence of the deformity and by the postoperative Dimeglio score [7]. Relapse of the deformity, defined as the child walking on the side of the foot, or when shoe fitting is a problem [11], was noted. Functional results were evaluated according to the Laaveg and Ponseti classification [26]. This system quantifies the functional results of treatment of clubfoot. In this system, 100 points indicate a normal foot. Final points are the sum of the following: a maximum score of 30 points for amount of pain, 20 points each for level of activity and patient satisfaction, and 10 points each for motion of the ankle and foot, position of the heel during stance, and gait. As in the original article by Laaveg and Ponseti [26], we rated a score of 90 to 100 points as excellent, 80 to 89 points as good, 70 to 79 points as fair, and less than 70 points as poor.

Descriptive statistics were calculated. The chi-square test was performed to compare preoperative and postoperative-reported data in relation to Dimeglio score. Statistical analysis was performed with PASW[®] software package (SPSS, Chicago, IL, USA).

Results

All patients had improvement of forefoot adduction and midfoot supination deformities (Fig. 2). In the 49 feet

Fig. 2A–G Preoperative standing (A) posterior, (B) inferior, and (C) superior views show a 6-year-old patient with bilateral neglected clubfeet. Postoperative standing (D) anterior and (E) posterior views and (F) inferior and (G) superior views of the same patient show good correction with plantigrade feet.

having lateral calluses preoperatively, these calluses disappeared postoperatively. Qualitatively, the gait improved owing to plantigrade placement of the foot, with no relapse. Grading of the feet at final followup according to Dimeglio et al. [7] showed significant improvement (Pearson chi-square = 112; $p < 0.001$), with 39 feet graded as Grade I and 17 graded as Grade II (Table 2). According to the Laaveg and Ponseti classification (Table 3), at 1-year followup, the mean rating of function for all 56 clubfeet was 84.2 points. According to the scores, 24 feet showed excellent correction, 20 good, nine fair, and three poor. At last followup, the mean rating of function for all 56 clubfeet was 82.7 points; an excellent result was observed in 23 feet, while 18 of 20 kept a good result. Fair and poor results were observed in 12 and three patients, respectively.

There were two complications in this group (two of 56 feet; 3.6%). Both were skin-related. Delayed wound healing was observed in the two patients and involved the medial surgical approach; wounds healed spontaneously in 6 weeks after cast removal. No reoperations were performed in this series.

Discussion

Despite widespread worldwide use of the Ponseti method through targeted national programs [28, 36, 38, 40, 42], patients with neglected clubfeet still present for evaluation because of limited access to treatment facilities and lack of

awareness that treatment options are available [19, 28, 37]. In many low-income countries, patients with congenital clubfeet present to the physician late, when parents become concerned about their children starting to walk on the deformed feet. At that time, the foot is likely to have become rigid, and typically the child walks with weight-bearing on the dorsolateral aspect of the affected foot [5, 16]. Although much of the current literature reports the clinical results of treatment for congenital clubfoot in the early years, few reports discuss the results of surgical techniques required to treat the more rigid deformities found in older children [12, 24, 25]. We perform shortening of the lateral column consisting of a single cuboid osteotomy associated with a selected soft tissue release to correct the residual deformities. In the current study, we evaluated (1) the results of this approach for neglected clubfoot in older children through objective physical examination measurements and functional assessments of the patients postoperatively, (2) determined the frequency of surgical complications, and (3) ascertained whether the initial correction achieved was maintained without additional surgery.

This study has some limitations. First, and most important, a relatively high proportion of the patients we operated on were not available to us for followup. Seventeen patients were not available at followup, and even though it is unlikely that these patients underwent surgery, some could have had recurrence or a late undesired complication, or might have required additional surgery.

Table 3. Results according to Laaveg and Ponseti score [26] at 1 year and last followup

Score category	1-year followup (number of feet)	Mean Laaveg and Ponseti score (points)	Last followup (2–9 years) (number of feet)	Mean Laaveg and Ponseti score (points)
Excellent (90–100 points)	24	96.3	23	93.4
Good (80–89 points)	20	86.6	18	84.7
Fair (70–79 points)	9	77.7	12	72.5
Poor (< 70 points)	3	63.5	3	58.8
Total	56	84.2	56	82.7

Second, the study is a retrospective review of a single-center experience in the absence of a control group, so the findings may not be generalizable. The lack of radiographic followup data is also an obvious limitation. However, there has been no consensus concerning the use of radiography during followup of clubfoot treatment [3, 41, 47]. Although some authors found correlations between clinical outcome and radiographic parameters [26, 44], others did not see any relationship [4, 17]. Another limitation could be the followup time which averaged 6 years and ranged from 2 to 9 years. A previous study on patients who had surgery for clubfoot showed that most relapses occur at an average age of 3 years [43]. In this context, it is unlikely that further relapses will occur 3 years after surgery, but the six patients in our series with 2- to 3-year followup could still require additional surgery in the future. However, our results in a population with an average 6-year followup showed sound results and applied to a patient population with severe deformity. Finally, we used categorical variables in the Laaveg and Ponseti scoring system, as in the original article, but such ratings are prone to interobserver variability and may not match patient-generated assessments [45]. Despite the widespread use of the Laaveg and Ponseti scoring in the original form, to partially overcome this potential limitation and better reflect results inside categorical ratings, we also provided average scores in each categorical rank.

We consider STR to be the mainstay of surgery in children with stiff nonreducible equinovarus deformity of the foot. Progressive release of the soft tissue on the shortened aspect of the foot allows for elongation of the medial column without the need for a cuneiform opening wedge osteotomy. Using the combination of STR plus cuboid subtraction osteotomy, the surgeon can correct the varus of the hindfoot by elongation of the medial flexors and by direct reduction of the navicular bone over the head of the talus, and can shorten the lateral column via a cuboid osteotomy that allows for correction of the neglected

deformity. The goal of treating a stiff nonreducible neglected clubfoot is to provide a corrected foot that is as plantigrade as possible, but with preserved growth ability [9, 10]. Numerous types and combinations of osteotomies have been described to achieve correction of complex deformities in the neglected clubfoot. At first, surgery was aimed at shortening of the lateral column [21] by wedge resection and enucleation of the cuboid, or conversely, at elongating the short medial column by medial wedge cuneiform osteotomy [18]. Later, the combination of medial and lateral osteotomies was described with the aim of addressing the disease at the midfoot and the forefoot [32], together with more complex osteotomies to ease correction of the rotatory component of the deformity [23, 39]; all of these were reserved for children older than 4 years where the medial cuneiform ossific nucleus is well developed [14, 29]. The most widespread combination of medial and lateral osteotomy is cuboid shortening and cuneiform opening wedge osteotomy [25]. The procedure consists of removal of a wedge from the shortened cuboid bone, which subsequently is inserted into a distracted osteotomy gap of the medial cuneiform bone. Sometimes the cuboid bone is too soft or too small for the length of the medial column to be corrected. Moreover, as in other lengthening procedures, mobilization of the bone wedge has been reported and requires revision surgery [24, 30, 46]. External fixation has been proposed to address stiff neglected clubfoot by progressive deformity correction during growth of the child [13, 27]; however, dedicated expertise, costs, and difficult care of the patient with an external fixator make this approach unsuitable in a rural context.

Extensive STRs may have short-term complications, and as much as 47% of patients may undergo additional surgery [10]. Moreover, repeated, soft tissue procedures may result in extensive postoperative scarring, with a high rate of complications such as skin necrosis, infection, or neurovascular damage [22]. Others have proposed a circumferential (Cincinnati) approach for these patients, used to address an extensive release at the posterolateral corner behind the lateral ankle or at the plantar fascia, but with the drawback of potential soft tissue loss that requires fasciocutaneous flaps [16]. We decided to perform two longitudinal incisions, on the posteromedial and lateral side of the foot, and a less extensive STR because of the contribution of the cuboid osteotomy, thus avoiding any additional plastic surgery which was not easy to perform in our clinical setting. In a study by Merrill et al. [33], four of 10 patients with isolated unilateral clubfoot had arterial anomalies in the clubfoot limb. Two of these patients had absent and/or hypoplastic anterior tibial arteries in combination with absent dorsalis pedis arteries; one had an absent posterior tibial artery and one had an isolated absent

dorsalis pedis artery. Merrill et al. concluded arterial deficiency is a marker of abnormal early limb development that correlates with other developmental abnormalities of the skeleton or muscle, which together cause clubfoot. Results from the arteriography study by Greider et al. [15] in children with clubfoot showed there is only one major vessel supplying the foot: the posterior tibial artery. They suggested any operation on the medial side of the foot therefore should include identification and adequate protection of this structure. Moreover, care must be taken when operating on the lateral side of a clubfoot to avoid injury to the lateral plantar artery during the approach for the cuboid. Finally, when correcting the position of a foot with a severe deformity, the surgeon must avoid stretching the only large artery that is present, or the operation may result in segmental thrombosis. Although numerous complications also might occur with the approach we describe [24], in this series the complications were uncommon and related mainly to wound healing. No patients in this series underwent accessory or subsequent surgeries in the study population; however, owing to the rural setting of this research, many were unavailable for followup. Even so, since this approach does not sacrifice any joints of the foot, these patients could undergo arthrodesis, talectomy, or osteotomy if late symptoms or recurrent deformities should arise. Triple arthrodesis or talectomy should be considered as salvage procedures because of the drawbacks associated to their use, such as a compromise in the longitudinal growth in arthrodesis, or incongruous joint and a less corrected foot for talectomy [37].

We found that combined STR plus cuboid osteotomy had a low rate of complications and led to a reasonably corrected foot in children older than 5 years with severe stiff nonreducible clubfoot. Performing a superolateral cuboid wedge subtraction osteotomy allowed for varus reduction and kept the foot in equinus. When combined with a progressive STR, stiff nonreducible neglected clubfoot deformity was managed safely and without the need to sacrifice a midfoot joint and with a low rate of complications.

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