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Original article

Relapse in surgically treated clubfoot: treatment approach and midterm results of revision surgery

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Abstract. *Background and aim:* The rate of recurrence of surgically early treated clubfoot is around 25% and the treatment of clubfoot recurrence remains debated. The aim of the study is to report a case series of 15 patients (16 feet) surgically treated for relapse of surgically treated clubfoot. *Methods:* A careful clinical and radiological evaluation of each deformity was made. The treatment algorithm was based on the pathological anatomy of the relapse, on the patient's age and on the use of a combination of surgical steps involving bones, soft tissue or both. *Results:* The average age of patients at the time of relapse treatment was 8 years and 6 months, with an average follow-up of 2 years. The average Avatar score was 77 (good result). The 16 feet submitted to evaluation obtained the following scores: 6 excellent, 4 good, 4 mediocre and 2 poor. *Conclusions*: The number of previous interventions does not seem to be related to the outcome. The clinical and radiological evaluation of the deformity is the most important step for the right application of the algorithm. The use of a treatment approach based on age and on the systematic treatment of bony and soft tissues leads to reproducible clinical results with functional improvement. (www.actabiomedica.it)

Key words: Relapse clubfoot, surgical treatment

Introduction

Congenital talipes equinovarus (i.e. congenital clubfoot) is a common congenital disorder occurring in approximately 1-2/1000 live births (1, 2). The typical tridimensional deformity is characterized by adductus forefoot, cavus midfoot, varus and equinus hindfoot, with skeletal and soft tissues abnormalities (3). Nowadays, serial casting associated or not with minor surgery (e.g. Achilles tendon tenotomy or lengthening, tibialis anterior tendon transfer) is widely accepted as first line treatment to restore shape and good function with a low rate of complications (4,5). In the past many surgical procedures and techniques for early extensive soft tissue release have been described, with the belief that the earlier the medial and posterior contractures were released, the better the deformity correction was (6, 7). Nevertheless long-term follow-up demonstrated unsatisfactory results of these procedures with high risk of post-operative (e.g. failed correction, wound breakdown, skin necrosis, and overcorrection) and long-term (e.g. scarring, growth disturbance, muscles weakness and stiffness) complications (8). Furthermore, some Authors, reported high recurrence rate after posteromedial release (9,10,11). It is important to differentiate relapse, which means recurrence of deformity in a previously well corrected foot, from residual clubfoot, which can be defined as previously undercorrected deformities (12). However, sometimes it is not possible to differentiate relapse from residual deformity and it was reported that recurrence is most often the consequence of insufficient primary surgery (13).

The most common forms of recurrence are forefoot and midfoot deformities (14). Tarraf and Caroll

reported that adduction and supination are present in 95% of clubfoot recurrences (15). The treatment of clubfoot recurrence remains an object of debate. In 1994 Lehman et al. (16) proposed an algorithm for clubfoot recurrence treatment and followed by a similar algorithm by Raab and Krauspe (17) in 1999.

The first aim of the study is to report the results of a retrospective evaluation of fifteen patients (sixteen feet) surgically treated for relapse of previously surgically treated clubfoot. The second aim is to show that the use of a treatment approach based on age and on the systematic treatment of bony and soft tissues could leads to reproducible clinical results with functional improvement.

Materials and methods

After approval of the study by the local ethics Committee, all patients surgically treated for relapse of previously surgically treated clubfoot presenting at our Department between January 2014 and February 2016 have been selected from the database of our Hospital. Indication for surgical treatment was a symptomatic non-plantigrade foot due to the relapse of previously surgically treated clubfoot. Before being enrolled in the study, all patients or their guardians signed written informed consent. Inclusion criteria for enrolment in the study were: age at surgery older than six years, previously surgically treated idiopathic clubfoot, rigid deformities not responsive to manipulation or recasting without evidence of arthritis signs at X-rays. Patients with secondary clubfoot and those who denied their informed consent were excluded.

Surgical planning

The goal of surgical treatment of residual deformities is to obtain an asymptomatic plantigrade foot suitable for wearing normal footwear (18). The decision-making process was made following the concept of "menù a la carte" treatment based on the type of the deformities, the age of the patient and the amount of stiffness. First of all is useful to decide whether to treat soft tissues, bones/joints or both. Bony procedures are usually performed before soft tissues procedures. The Authors started to address the hindfoot deformity, varus first and then equinus.

Clinical and radiographical examination

At the time of the first evaluation the Authors recorded personal data and clinical history and then clinical and radiographical examination was performed.

With the patient in standing position on a podoscope it is possible to evaluate any cavus or metatarsus adductus deformities (e.g. "peek a boo" sign), feet plantar arch and heel alignment. In case of varus heel the Coleman (19) and the Andreasis test (20) are performed to differentiate a primitive hindfoot varus from a varus heel secondary to a forefoot cavus. Gait observation is useful to exclude any equinus (tiptoes) or equinovarous gait and dynamic supination deformity (21, 22). The evaluation of the sole of the foot is useful to identify skin thickening (callosity) or tenderness areas. Finally, we evaluate the tibial torsion (by thigh-foot angle) (23), the strength of the leg and foot muscles and the active and passive range of motion of the ankle, midfoot and forefoot. In case of limited dorsiflexion of the ankle (normal range: 3 to 15 degrees past perpendicular), it is important to differentiate an equinus deformity, caused by an osseous condition, from an equinus contracture (caused by only soft tissues) and from a pseudoequinus (plantarflexed forefoot without ankle equinus). The Silfverskiöld test is useful to differentiate gastrocnemius versus soleus contracture and gastrocnemius versus achilles tightness (24).

Anteroposterior (AP) and lateral weight-bearing radiographs of the feet were performed preoperatively and postoperatively (25). In AP radiogram we evaluated adduction deformity by the talo–first metatarsal angle, the calcaneo–fifth metatarsal angle and the second metatarsal–tarsal angle (25,26,27,28) and the the varus deformity of the hindfoot by AP talo-calcaneal angle (Kite I angle).

In lateral radiograph we evaluated the equinovarus deformity of the hindfoot by the lateral talocalcaneal angle (Kite II angle) and the cavus deformity by the talar–first metatarsal angle (Meary's angle) (26).

The radiographic measurements also included the talocalcaneal index (sum of the lateral and anteroposterior measurements of the talocalcaneal angle) and talus-first metatarsal angle (26).

Bony and joints deformity, as talar dome and talar head flattening and degenerative arthrosis were also evaluated (29). The tarsal dome shape and its deformities were classified as normal, mildly flattened, moderately flattened, severely flattened using the criteria published by Dunn (41) and Kolb (42).

At follow-up, the overall outcome was assessed using the rating system described by Atar et al. (11), specific for clubfoot treatment evaluation. This score contains subjective (functional limitation, pain and satisfaction) and objective (dorsiflexion of the ankle, mobility of the subtalar joint, position of hell in standing position, foot shape and gait) clinical parameters and specific radiographic angles.

The Atar's score ranges from a maximum of 100 points, which corresponds to a normal foot, to a minimum of 0. The range between 100 and 85 corresponds to an excellent result, 84-70 points correspond to a good outcome, 69-60 points fair and less than 60 is considered a poor result.

Surgical techniques

Soft tissue procedures

Tibialis Anterior Tendon Transfer (TATT): The main indication was the presence of dynamic supination, although tibialis anterior is also involved in cavus deformities, hindfoot varus and forefoot adduction [30]. Our surgical technique is similar to that described by Ippolito et al. [31]: the TAT was splitted and its transfer, underneath the extensor retinaculum, was fixed to the lateral cuneiform by a bone tunnel using a Mg screw (Magnezix, Syntellix AG, Hannover, Germany) that acts as a resorbable interference screw (fig. 1, 2).

Plantar fasciotomy: the main indication is forefoot equinus (cavus) (12). We performed an open complete plantar fascia release using a modified Steindler technique (32).

Lengthening of the Gastrocnemius-Soleus Complex (LGSC): the main indication was equinus deformity, defined as the impossibility to achieve the neutral ankle position due to a short and tight Achilles tendon, evaluated at the end of all bony procedures. The LGSC was performed according to Baker technique (33).

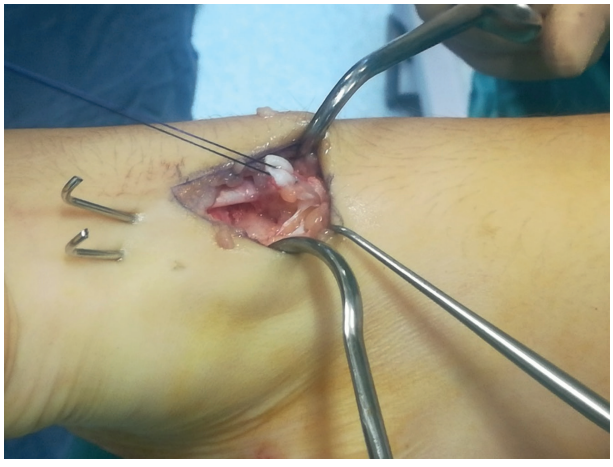

Fig. 1 Intraoperative image of the tibial anterior detachment from its distal insertion.

Fig. 2 Intraoperative image of the tibial anterior and fixation to the lateral cuneiform by a bone tunnel using a Mg screw (Magnezix, Syntellix AG, Hannover, Germany).

Bone procedures

Medial column lengthening with lateral column shortening: the main indication was the so called "bean-shaped foot" deformity (adduction and mid-foot supination) (28). We performed an opening wedge osteotomy of the medial cuneiform and a closing wedge osteotomy of the cuboid using the modified technique described by Pohl et al. (25). The opening wedge of the medial cuneiform was filled with the bone removed from the cuboid and with tibial autologous graft. The osteotomies were performed under X-ray control using an osteotome and fixed by one or two 1.6 mm K wires.

Calcaneal osteotomy: the main indication for this procedure was fixed varus heel. We used a closing wedge osteotomy as described by Dwyer (34) following the indications given by Lamm et al. (35). *(Case example:* Fig. 3a, 3b, 4a, 4b, 4c, 4d, 5a, 5b, 6a, 6b)

"Reverse Jones" procedure: it was performed in case of hallux flexus, a deformity consisting in a plantar flexion contracture of the metatarso-phalangeal joint with a dorsiflexion contracture of the tarso-1st metatarsal joint (36,37). We performed the modified technique described by Kuo (38): a plantar flexion osteotomy of the 1st metatarsal was performed and the flexor hallucis longus was trasferred to the head of the 1st metatarsal. (Fig. 7a, 7b, 7c)

Joint procedures:

Posterior release: it was performed in case of severe equinus deformity of the ankle which persists after all the planned bony procedures and LGSC were completed. This procedure was performed through a postero-lateral approach to the ankle and consisted in talo-crural capsulotomy and posterolateral knot release (including lower part of the ankle fascia, superior fibular retinaculum and calcaneo-fibular and posterior talo-fibular ligaments) (Fig. 8) (39,40).

Anesthesia and analgesic management

All surgical procedures were performed under spinal anesthesia. A spinal catheter was left in place for pain management for 48 hours after surgery in all patients treated by osteotomy.

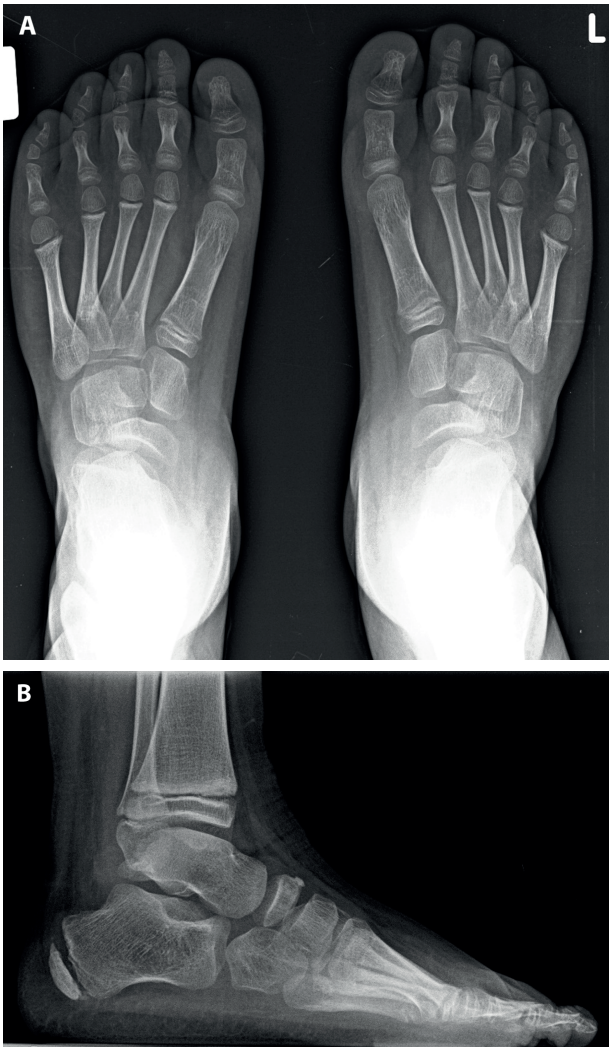

Figure 3 a) Pre-operative antero-posterior and lateral weight-bearing radiographs of the feet of a child with left relapsed clubfoot. b) Pre-operative antero-posterior and lateral weight-bearing radiographs of the feet of a child with left relapsed clubfoot.

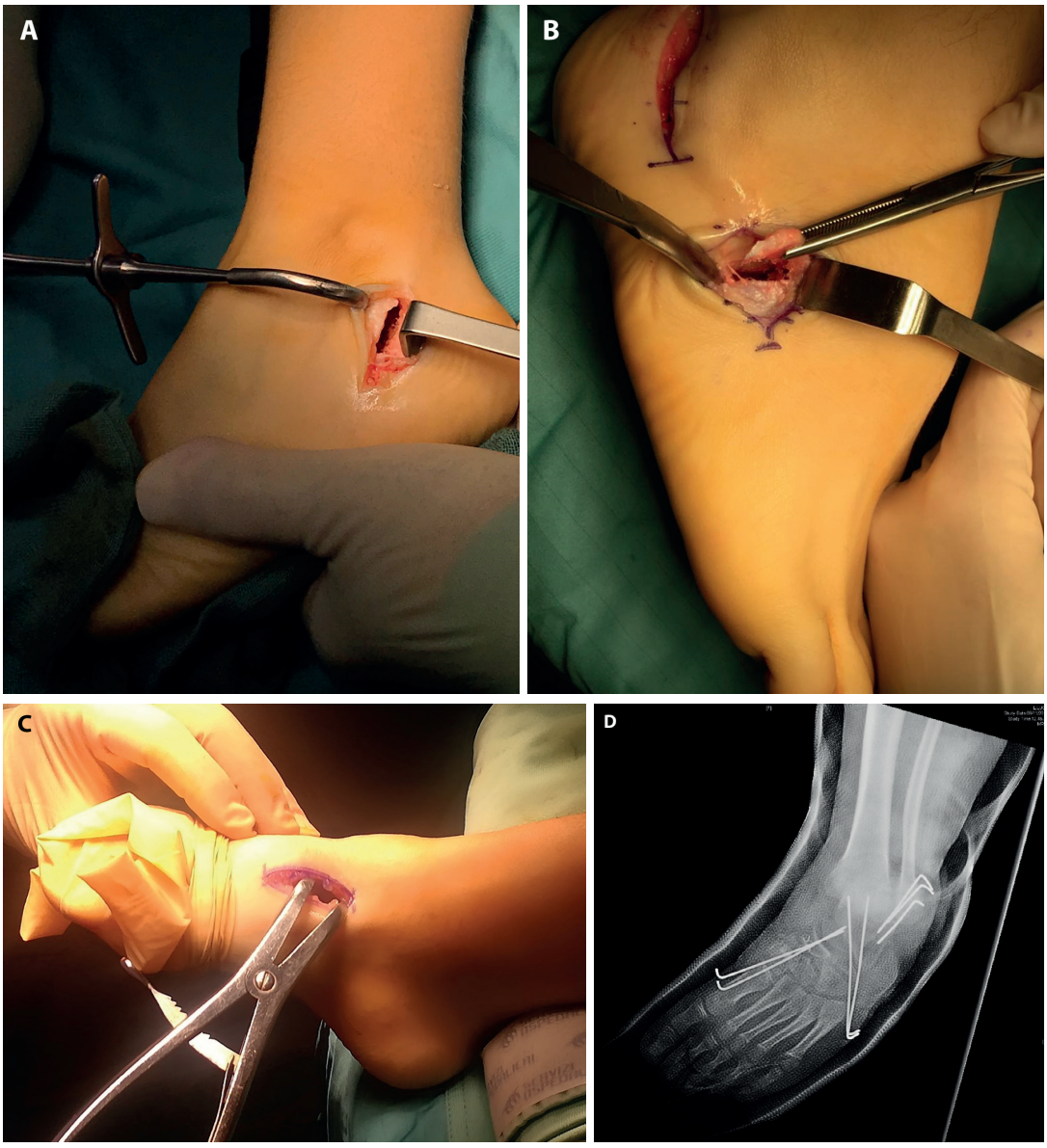

Figure 4. a) Intraoperative image of calcaneal osteotomy (6a), closing wedge osteotomy of the cuboid (6b) an opening wedge osteotomy of the medial cuneiform with autologous graft (6c) and and postoperative X-ray of calcaneal osteotomy, closing wedge osteotomy of the cuboid and opening wedge osteotomy of the medial cuneiform with autologous graft fixed by K-wires (6d) b) Intraoperative image of calcaneal osteotomy (6a), closing wedge osteotomy of the cuboid (6b) an opening wedge osteotomy of the medial cuneiform with autologous graft (6c) and and postoperative X-ray of calcaneal osteotomy, closing wedge osteotomy of the cuboid and opening wedge osteotomy of the medial cuneiform with autologous graft fixed by K-wires (6d). c) Intraoperative image of calcaneal osteotomy (6a), closing wedge osteotomy of the cuboid (6b) an opening wedge osteotomy of the medial cuneiform with autologous graft (6c) and and postoperative X-ray of calcaneal osteotomy, closing wedge osteotomy of the cuboid and opening wedge osteotomy of the medial cuneiform with autologous graft fixed by K-wires (6d). d) Intraoperative image of calcaneal osteotomy (6a), closing wedge osteotomy of the cuboid (6b) an opening wedge osteotomy of the medial cuneiform with autologous graft (6c) and and postoperative X-ray of calcaneal osteotomy, closing wedge osteotomy of the cuboid and opening wedge osteotomy of the medial cuneiform with autologous graft fixed by K-wires (6d)

Figure 5. a) Clinical image in front and lateral view in standing position of a child three years after surgical treatment for relapsed left clubfoot. b) Clinical image in front and lateral view in standing position of a child three years after surgical treatment for relapsed left clubfoot.

Aftercare

All patients were immobilized in a post-operative no weight-bearing short-leg cast for 4 weeks. After cast removal the patients were allowed to start to walk with crutches with partial weight-bearing (25% of body weight increasing every week till full body weight bearing recovery after one month).

Statistical method

Categorical variables are described by absolute and relative frequencies, while continuous variables are expressed by means, standard deviation (SD), medians and range. Parametric (paired t-test) or non parametric analysis (Wilcoxon test) was used for continuous variables. A p-value less than 0.05 was considered statistically significant; all p-values were based on two-tailed tests.

Statistical analysis was performed using SPSS for macOS (SPSS Inc., Chicago, Illinois, USA).

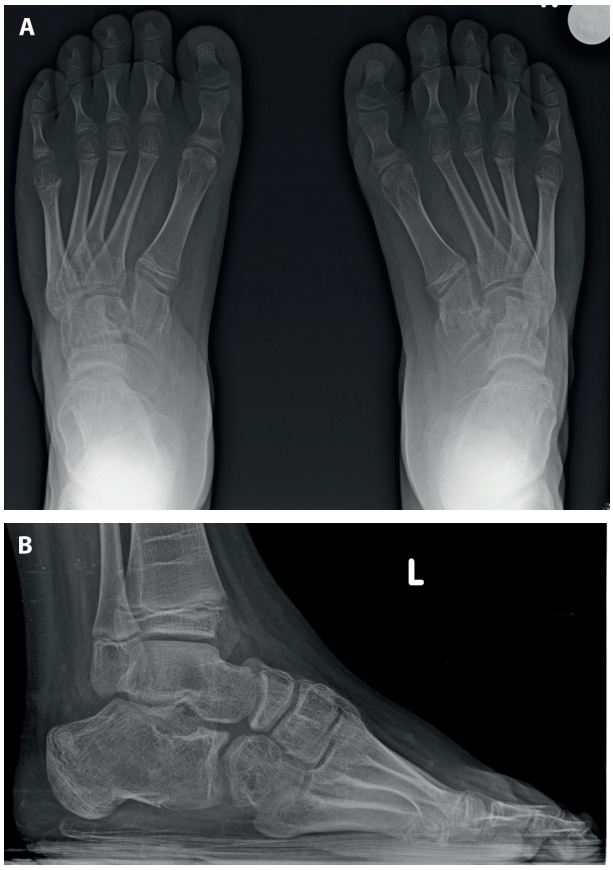

Figure 6. a) Anteroposterior and lateral weight-bearing radiographs of the feet three years after surgical treatment for relapsed left clubfoot. b) Anteroposterior and lateral weight-bearing radiographs of the feet three years after surgical treatment for relapsed left clubfoot.

Results

Fourteen patients (15 feet) treated by the senior surgeon in a single Operating Unit in 2017 were subjected to case review. The sample was composed by 3 females and 11 males. One patient had bilateral clubfoot.

The median age at revision surgery was 10 years (range 6 – 21). The average follow-up was 2 years (range 1 – 4 years).

The number of previous surgical procedures ranged from 1 to 4 interventions. In 8 (50%) cases they underwent a single intervention, 2 (12.5%) cases had two, 3 (18.6%) cases had three and 2 (12.5%) cases had four previous surgical interventions. Nine (56.2%)

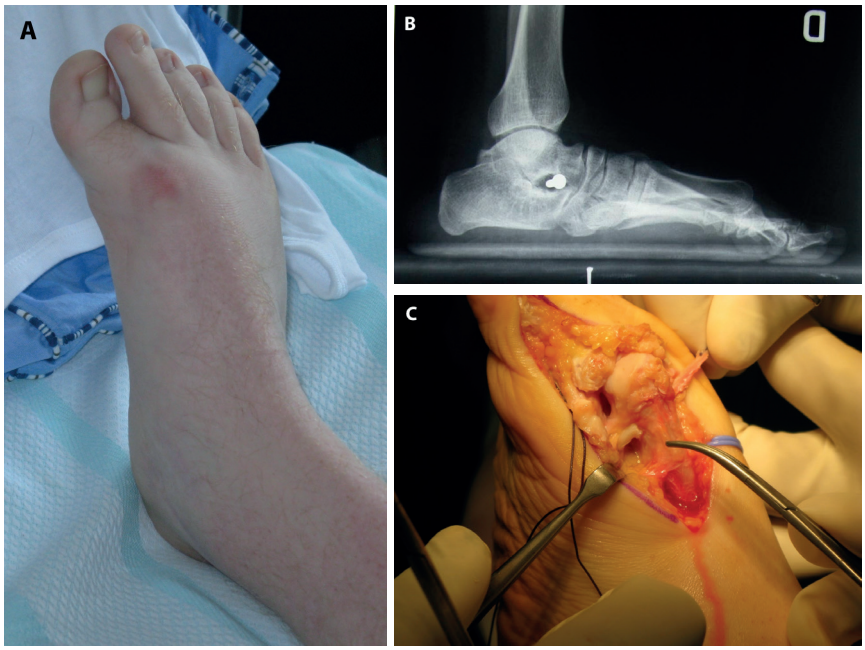

Figure 7. a) Clinical image of left foot of a child with hallux flexus (a), lateral weight bearing X-ray of left foot with hallux flexus (b). Intraoperative image of "Reverse Jones" procedure modified technique: plantar flexion osteotomy of the 1st metatarsal and flexor hallucis longus trasfer to the head of the 1st metatarsal (c). b) Clinical image of left foot of a child with hallux flexus (a), lateral weight bearing X-ray of left foot with hallux flexus (b). Intraoperative image of "Reverse Jones" procedure modified technique: plantar flexion osteotomy of the 1st metatarsal and flexor hallucis longus trasfer to the head of the 1st metatarsal (c). c) Clinical image of left foot of a child with hallux flexus (a), lateral weight bearing X-ray of left foot with hallux flexus (b). Intraoperative image of "Reverse Jones" procedure modified technique: plantar flexion osteotomy of the 1st metatarsal and flexor hallucis longus trasfer to the head of the 1st metatarsal (c).

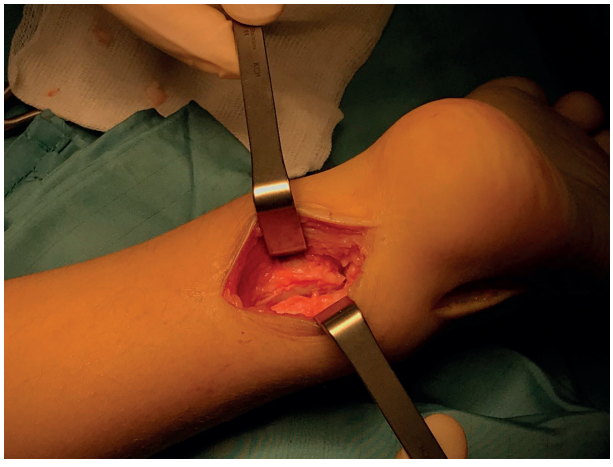

Fig 8. Intraoperative image of posterior tibio-tarsal capsule and sydemosis release.

Table 1. Proportions of profiles of deformities in the relapse group. Values are number of feet

Profile	N° of relapse
Single deformity	**6**
equinus/decreased dorsifl exion (EqDD)	**3**
active supination	**2**
adduction	**1**
cavus	-
EqDD involved	**10**
EqDD + active supination	**2**
EqDD + adduction	**3**
EqDD + varus	**1**
EqDD + active supination + varus	**1**
EqDD + active supination + adduction	**1**
EqDD + adduction + cavus	
EqDD + varus + adduction	
EqDD + active supination + varus + adduction	**2**

patients had previous surgical interventions performed at other institutions.

The revision surgery was performed after a median of 8 years (range 3 – 16 years) from the last operation. The most common residual deformity was forefoot adduction and supination in 13 feet (81.3%). The pattern of the deformities is reported in Table 1.

The mean Atar score was 28.4 ± 8 at pre-operative evaluation and 74 ± 13.7 at follow up with a statistical significance ($p < .001$). Out of 15 feet included, 6 (37.5%) obtained an excellent score, 4 (25%) a good score, 3 (18.6%) a fair score and 2 (12.5%) poor. Nine patients were asymptomatic during daily activities, five patients reported partial pain after demanding activities. In one case the pain was reported as frequent. Five patients were able to use commercial footwear. Nine patients had to use orthotic insoles and one orthopedic shoes. Patients reported high and partial satisfaction.

As for the radiographic parameters the mean talus-first metatarsal angle was 21.6 ± 2.2 degrees preoperatively and 20.7 ± 2.1 at follow-up with a statistical significance ($p = 0.03$). The mean talo-calcaneal index passed from 48.2 ± 9.2 degrees preoperatively to 51.9 ± 5.8 at follow-up with a statistical significance ($p = 0.04$).

Osteotomies healed between 8 and 10 weeks.

The talar dome was deformed in 11 patients, slightly flattened in 5 patients and greatly altered or flat in 6 patients. Talar dome shape was normal in 4 patients. The analysis of the talar head sphericity at talo-navicular joint reveals a convex talar head in 6 cases, plana in 7 cases and concave in 2. In one case there was dorsal subluxation of the triangular-shaped navicular.

Complications

We recorded 2 cases of superficial infections that healed with dedicated dressings. No other complications have been reported.

Discussion

In this retrospective study, we found that systematic approach to relapsed clubfoot deformity following a decision-making approach is helpful to understand the deformities and their treatment.

Because of the efficacy of the Ponseti method, extensive surgery has become obsolete both for first line treatment and recurrences. However, in case of severe relapse, previous extensive surgery or syndromic cases, the Ponseti method may not be completely effective. Several Authors reported their treatment algorithm based on the age of the patient, the type of deformity and its severity and stiffness (11, 16, 17, 43).

In patients younger than 6 years the relapse is usually treated with soft tissue or "joint sparring" skeletal procedures while in older patients or in case of fixed deformities more extensive treatment might be required.

However, in case of recurrence of the deformity after corrective surgery, the approach is not completely codified and in the literature there are only few studies reporting the results of these procedures (12,17).

Atar and colleagues (11) in 1992 reported the results of the treatments performed on 29 feet with replased clubfoot. They used the age as the most important criterion for treatment planning. The Authors reported 8 excellent, 11 good, 8 mediocre and 2 poor results at 30 months. The number of excellent and good results is comparable to the results reported in our study.

Lehman (44) in 1999 reported the results of the treatments performed on 27 feet. The age of the patients was in the range from 4 to 8 years. In this case, a complete release associated with a calcaneo-cuboid arthrodesis was carried out. The Authors reported 8 cases with excellent results, 14 good, 4 mediocre and 1 poor at a follow-up of 5-14 years. Even in this case, they reported positive results (flexible and painless plantigrade feet) in 78% of the patients.

In our case series all the patients were older than 6 years without severe arthritic aspect at the X-rays. In case of multiplanar foot deformities the correction of the hindfoot was performed first and it becomes the keystone for forefoot correction (45). Furthermore, since it is well known that bony procedure could modify soft tissues condition (46, 47), in case of double or multiple deformities (e.g. hindfoot varus and equinus) we start from bony procedures and then we treat soft tissue if still necessary (e.g. Dwyer osteotomy before a possible LGSC). Following the concept of "menu a la carte" approach, the decision making was made

both in preoperative evaluation and during surgery. The deformity evaluation is mainly based on clinical examination while X-ray is used to confirm the clinical findings and to find and quantify the amount of bone and joint involvement (48).

The varus deformity of the hindfoot may be corrected with different type of calcaneus osteotomy (49). We performed the closing wedge osteotomy (Dwyer technique) because this approach, compared to open wedge osteotomy, is associated with less wound-healing problems and may reduce the tension on the Achille's tendon complex (50). To address equinus deformity it is mandatory to differentiate a bone equinus deformity from an equinus contractures. Indeed, the correction of equinus deformity need bony or joints procedures (e.g. supramalleolar wedge osteotomy (51)) while equinus contracture may be addressed by soft tissue procedures. Silfverskiöld test is important to differentiate an isolated gastrocnemius contracture from a gastrocnemius-soleus complex contracture (52). The first condition can be addressed by the so called gastrocnemius recession (53) while the second condition need a lengthening of the gastrocnemius-soleus complex (33). In our case series all the patients had an equinus due to gastrocnemius-soleus complex contracture with negative Silfverskiöld test and they were treated by a LGSC using the Baker technique [33]. The Authors prefer to avoid a direct Achilles lengthening due to its potential complications, as tendinosis, rupture, weakness and prolonged recovery (53). In case of incomplete equinus correction after LGSC during intra-operative evaluation, a posterior release of the ankle was performed (54).

As reported by many Authors (15) the deformities that most often recur in clubfoot is the adduction and supination of the forefoot. The chain of events that leads to the imbalance between lateral and the medial column leads to the so called "bean-shaped foot" (55). Walking is difficult, the adoption of commercial shoes is not comfortable and sport activities are sometimes abandoned (25). In such cases, the combination of an opening wedge osteotomy of the medial cuneiform and a closing wedge osteotomy of the cuboid is proposed [(28). Gordon and colleagues (56) suggest that this double osteotomy should be reserved for patients over 5 years of age because of for the partial ossification of the cuneiform which makes bone grafting difficult when filling the osteotomy in plus. Lourenco et al. [(55) reported the results of double osteotomy technique for the treatment of 39 feet. The 4.8-years follow up shows a significant improvement in both radiographic and clinical parameters.

Ettl and colleagues (17) investigated the effect of the peritalar release on clinical outcome, but did not report data to support the systematic use of this procedure in revision surgery. In our study extensive soft tissue release was avoided for the potential risk of wound healing problems.

In our opinion the strength of our study is that the group of patients enrolled was homogenous: all the patients affected by relapse of previously surgical treated clubfoot were in the same range of age and had no signs of severe arthrosis on the X-rays.

The present study has several limits which can be summarized in: a) we always reported "relapse" in the text but the differentiation between relapse and residual deformity may not be always clear. However, this problem is well known and reported in the literature and the importance for the decision of additional surgical interventions is minimal (8,13); b) a longer follow-up could help to better understand the natural history of surgical treated clubfoot; c) small number of cases reported, but as the Ponseti method is now extensively used, surgically treated clubfoot and its relative relapse are rare and our case series numerosity is in line with others reported in the literature. We reported a joint sparring approach which is possible only in the absence of severe degenerative changes. Furthermore, in older patients with severe rigid deformities the osteotomies and gradual correction using distraction osteogenesis could be useful (12, 57, 58).

In conclusion, relapse in previously surgically treated clubfoot is now a rare condition but still possible. It is usually associated with poor clinical findings and patients complains so it requires treatment. In all cases the goal of the treatment is to achieve a plantigrade painless foot. In case of relapsed clubfoot following Ponseti treatment, recasting can be effective sometimes, but in case of severe deformities and stiffness surgical approach may be required. Clinical and radiological evaluation of the deformity is the

most important step for the right application of the algorithm for treatment. At follow up examination, we found ankle dorsiflexion and subtalar range of motion increase and an improvement of the appereance of the hindfoot and the forefoot in standing position. The quantitative analysis of the measurements shows an improvement of the parameters considered, but it is not possible to demonstrate a statistical significance.

The approach adopted is useful to evaluate and understand the deformities and it is essential to guide a systematic approach for surgical treatment planning and timing.

The quantitative analysis of the measurements shows an improvement of the parameters considered but it is not possible to demonstrate a statistical significance. For example, the ankle dorsiflexion and the subtalar range of motion increased and the appereance of the hindfoot and the forefoot in standing position improved.

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Conflict of Interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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