

Long term results of the revisited Meary closing wedge tarsectomy for the treatment of the fixed cavo-varus foot in adolescent with Charcot-Marie-Tooth disease

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ARTICLE INFO

Article history:

Received 26 January 2018

Received in revised form 28 October 2018

Accepted 5 November 2018

Keywords:

Charcot-Marie-Tooth foot

Fixed cavovarus

Adolescent

Tarsectomy

Salvage procedure

ABSTRACT

Background: Various techniques have been proposed for the treatment of cavovarus feet (CVF). The aim of this study was to report outcomes of the revisited Meary's dorsal closing wedge tarsectomy for fixed CVF secondary to Charcot-Marie-Tooth (CMT) disease.

Methods: All CVF operated on between 1977 and 2011 were included. The tarsectomy design was modified from its original description and systemically combined with a plantar fascia release, a Dwyer osteotomy and a proximal extension osteotomy of the 1st metatarsal bone if required. Outcomes were assessed by 2 functional scores and radiographically.

Results: Among the 26 feet (20 patients), the Wicart and Seringe score was very good or good, fair and poor in respectively 58%, 23% and 19% of the feet. Hindfoot and midfoot AOFASs were of 95.5 and 75 respectively. All radiographic measures were significantly improved.

Conclusions: This complete revisited procedure is an efficient and safe surgical technique for the treatment of the CMT disease CVF.

Level of evidence: Level IV.

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1. Introduction

Neurological cavovarus foot (CVF) secondary to Charcot-Marie-Tooth (CMT) disease, is a complex three-dimensional deformity, that combines medial cavus to forefoot pronation and hind-foot varus [1,2]. Hindfoot varus, which is reducible at an early stage and that becomes more rigid at the end of growth, is induced by the irreducible forefoot pronation [3,4]. Indeed, as reported by Meary in the pes cavus symposium of 1967, it is a compensatory mechanism to restore the tripod foot support [5]. Underlying neurological degenerative disease and growth may induce worsening of the deformity during childhood and particularly during adolescence leading to rigid and severe deformities in young adults.

CVF surgery aims to obtain a painless and well-balanced plantigrade foot as flexible as possible. Therefore, triple arthrodesis, which is still commonly performed, should be avoided in this indication because of the long-term risk of ankle or tarso-metatarsal joints degenerative arthritis [6–9]. Various therapeutic options have been proposed, consisting on bone procedures more or less associated to soft tissue releases and tendon transfers [10–19]. A dorsal mid-foot tarsectomy combined with a plantar fascia release has been published in 1976: the Meary dorsal wedge tarsectomy [15]. This procedure consisted on a triangular tarsectomy with the largest wedge located dorsally and medially on the medial cuneiform bone. However, if the results showed an efficient correction of the cavus and the forefoot pronation deformity, forefoot adduction was increased and hindfoot varus was not properly corrected. Therefore, the original technique was revisited in order to correct all the components of the deformity [10,11].

The aim of this study was to report radiological and functional outcomes of the revisited Meary's dorsal closing wedge tarsectomy

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in neurological cavovarus foot (CVF). The hypothesis was that the revisited procedure improves clinical, functional and radiological parameters.

2. Materials and methods

2.1. Patients

All adolescents with a CMT disease and a fixed CVF operated on a revisited Meary's closing wedge dorsal tarsectomy combined with a Dwyer calcaneal osteotomy and a plantar fascia release between 1977 and 2011 were retrospectively reviewed in a single institution [4,11,15]. Twenty patients (10 females and 10 males) were included in the study. All patients were skeletally mature and with a fixed deformity of the foot. Patients with prior foot surgery were excluded. CMT disease was genetically confirmed in all patients. The exact neurological status was not reported but all the patients were addressed by a neurologist that diagnosed the CMT disease and all were already to an advanced stage of the foot deformity. Age at surgery averaged 15.8 years, range 15.3 to 20.4 years. The surgical procedure involved 7 right feet, 7 left and was bilateral 6 times.

2.2. Surgical procedure

All surgeries were performed by one of the two senior foot surgeons of the department during a single stage procedure. The first step of the procedure was a selective plantar fascia release through a short medio plantar approach. The dorsal tarsectomy was performed with osteotomes through two longitudinal incisions (dorsolateral and dorsomedial) in the axis of the 1st and the 4th commissures directly to the bone with sub periosteal exposure in order to prevent skin healing problems. The posterior frontal plane of the osteotomy was delimited by two perpendicular and convergent k-wires (one vertical in the navicular bone and one horizontal in the cuboid bone)

introduced under fluoroscopic control. Posterior osteotomies of the navicular and the cuboid bones were performed through both incisions. Then, the anterior osteotomy was performed through the cuneiform bones and the cuboid. Consequently, the anterior facet joint of the navicular bone and the posterior facet joints of the 3 cuneiforms bones were removed. Instead of a triangular shape, the revisited osteotomy was curvilinear. The width was maximal in the intermediate cuneiform, which is the apex of the cavus and progressively decreased medially through the medial cuneiform avoiding undesired adduction. The width also decreased laterally through the cuboid and should remove the correct amount of bone to correct forefoot adduction (Fig. 1). A plantar hinge between the two osteotomies must be maintained to prevent foot shortening. The tarsectomy was closed and fixed by two staples. A Dwyer osteotomy completed the procedure for the correction of hindfoot varus [11]. The calcaneal osteotomy was not fixed since the Achilles tendon was not lengthened and the dorsal flexion in the postoperative cast allowed the closing of the osteotomy into valgus. A proximal 1st metatarsal bone extension osteotomy was also required in 16 cases (61.5%) for persisting plantar sticking [20]. Postoperatively, a posterior cast was applied during 7 days in order to control wounds healing, ankle in a slight dorsi-flexion for the closing of the calcaneal osteotomy. Thereafter, a short-leg cast was applied during 2 months including a full month of non-weight-bearing. After cast removal, physiotherapy consisted on gait training and restoration of foot and ankle range of motion.

2.3. Clinical assessment

Besides recurrent pain and ankle sprains reported by the patients, pre- and postoperative, physical examination also described the presence and location of plantar callosities and claw toes. Scaled plantar footprints pictures were recorded for 6 patients with unilateral surgical procedures in order to compare pre- and postoperative foot length (Fig. 2).

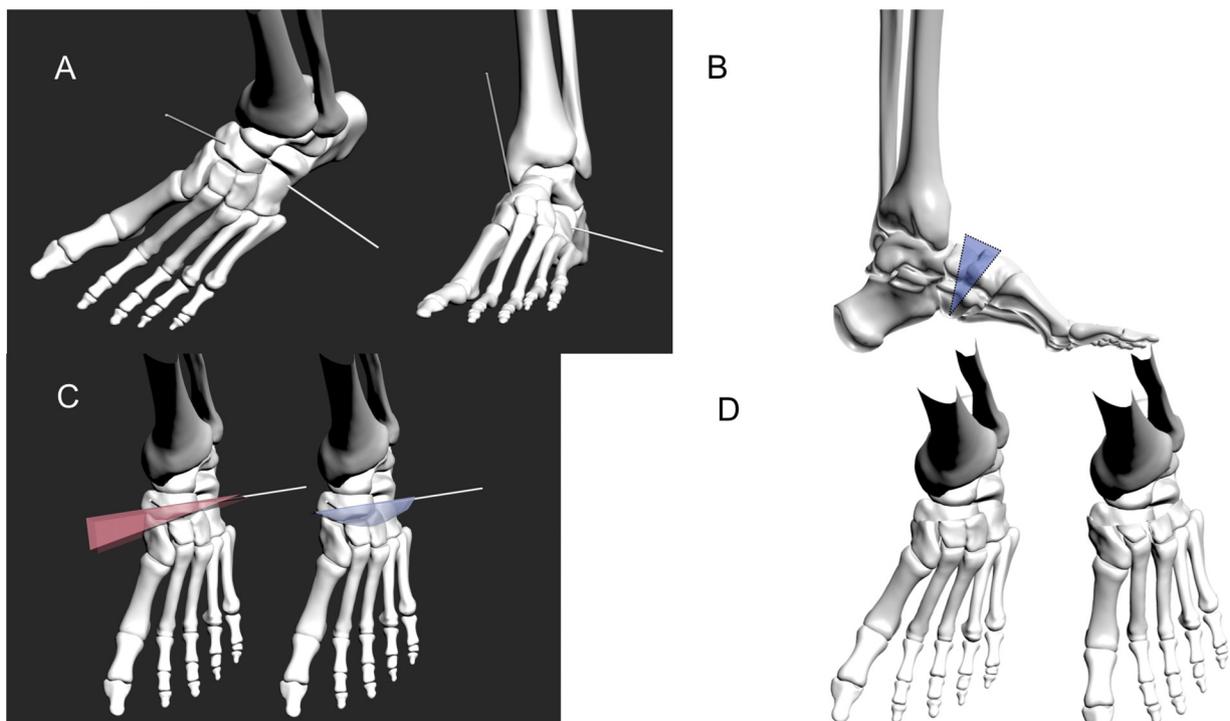


Fig. 1. Surgical procedure and comparison between the original and the revisited technique. A: Pin placement; B: lateral aspect of the osteotomy; C: design of the original and the revisited technique; D: comparison of the aspect after tarsectomy.

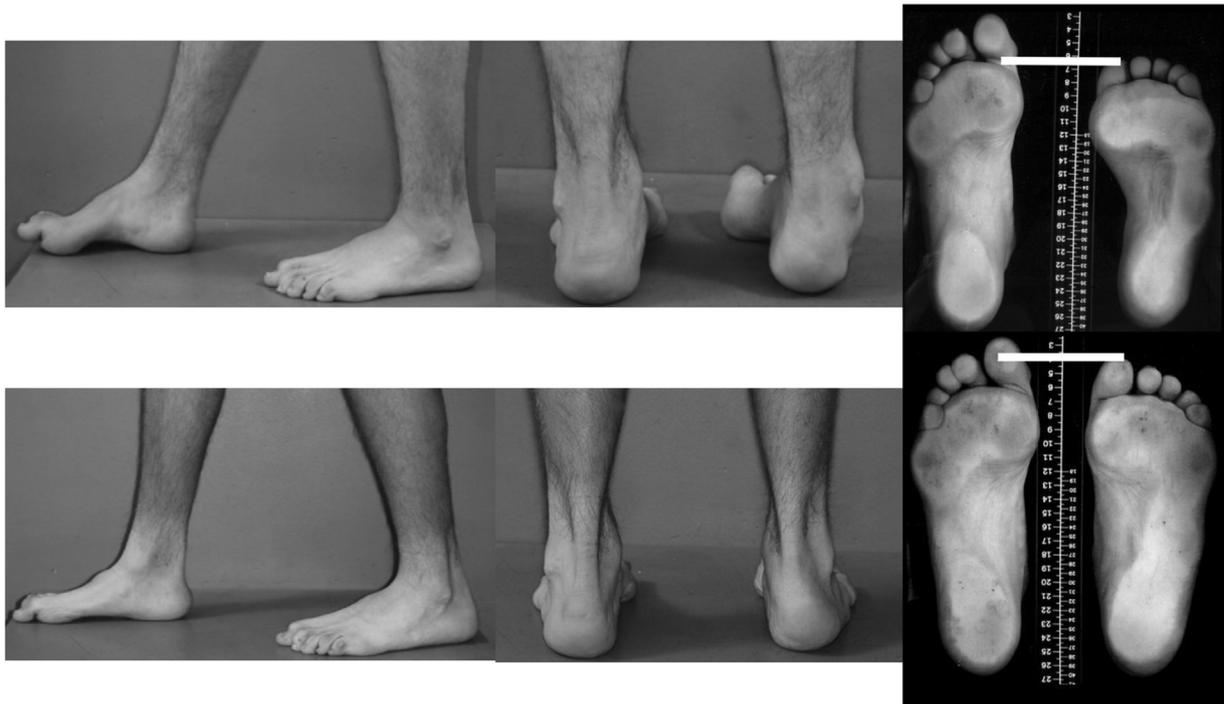


Fig. 2. Preoperative and 3-year postoperative clinical outcomes of a CMT patient operated unilaterally at 15.5 years old. A plantigrade foot with foot lengthening was obtained as well as the correction of the hindfoot varus.

2.4. Functional assessment

Postoperative functional outcomes were evaluated by the global Wicart and Seringe (WS) score (Table 1) [21,22]. The score evaluated simultaneously function, clinical and radiological outcomes. In addition, the 100 points hindfoot, midfoot, hallux and lesser toes American Orthopaedic Foot and Ankle Society (AOFAS) scores were also available for 6 patients (10 feet) who accepted to fulfilled the questionnaire [23].

2.5. Radiological assessment

Pre- and postoperative dorsoplantar and lateral weight-bearing radiographs were performed. Radiographic measures in the sagittal plane included the Meary angle, calcaneal pitch and metatarsal stacking angle measurements (Fig. 3). The Meary angle was defined by the angle between the talus long axis and the first metatarsal axis. It is a measure of the global cavus deformity. The calcaneal pitch angle is formed by the horizontal and a line from the base of the heel and the inferior cortex of the calcaneus. It measures the amount of cavus deformity due to hindfoot. The

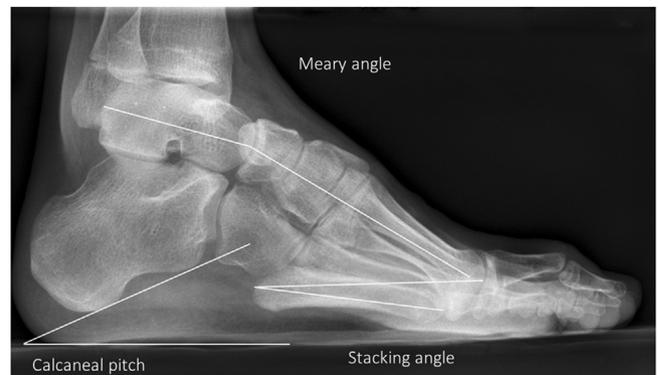


Fig. 3. Sagittal plane measurements appreciated by the Meary angle, calcaneal pitch and stacking angle.

metatarsal stacking angle was defined by the angle between the axis of the 5th metatarsal bone and the line drawn between the head of the great toe metatarsal and the base of the 5th metatarsal. It measures the amount of forefoot pronation [24]. Radiographic

Table 1
Wicart and Seringe score: an outcome grading classification.

Results	Symptoms	Meary angle (degrees) and clinical position of heel
Very good	None	0° ≤ Meary angle ≤ 15° and valgus Or 0° ≤ Meary angle ≤ 5° and neutral or varus
Good	None	15° < Meary angle ≤ 20° and valgus Or 5° < Meary angle ≤ 20° and neutral Or 5° < Meary angle ≤ 15° and varus Or -15° ≤ Meary angle < 0° (minor overcorrection)
Fair	None	Meary angle > 20° and valgus or neutral Or Meary angle > 15° and varus Or Meary angle < -15° (major overcorrection)
Poor	Recurrent pain and/or sprain	Recurrence of the deformity requiring triple arthrodesis

measures in the coronal plane included talocalcaneal angle (TCA), talo-first metatarsal angle (TM1) and calcaneo-fifth metatarsal angle (CM5) measurements (Fig. 4). The talocalcaneal angle measures hindfoot medial rotation. TM1 measures both medial rotation and forefoot adduction and CM5 measures forefoot adduction.

2.6. Statistical analysis

Statistical analyses were performed using SPSS statistics 23.0 (SPSS Inc., Armonk, NY, USA). A Shapiro–Wilk test was performed to assess data distribution. Continuous numeric data was expressed as means and ranges of minimal and maximal values. Pre- and postoperative results were compared with two-tailed



Fig. 4. Frontal plane measurements appreciated by the talocalcaneal angle (TCA), calcaneo-5th metatarsal angle (CM5) and talo-1st metatarsal angle (TM1).

Student t-tests for variables with normal distribution. Pre- and postoperative results were compared with 2-tailed Fisher's exact tests for nominal variables. A $p < 0.05$ was considered significant.

3. Results

3.1. Patients

A total of 26 feet (20 patients) were included with a mean follow-up of 6.2 years, range from 1.2 to 32.2 years. Age at follow-up averaged 22.1 years, range from 15.3 to 46.7 years. Four patients (6 feet) were followed more than 20 years after surgery (Fig. 5).

3.2. Clinical outcomes

At last follow-up, sprains and calluses were significantly reduced ($p < 0.05$) (Table 3). Among the 7 feet with persisting pain, 3 were connected with the development of claw toes deformities. However, claw toes were not significantly improved in the study.

On the six plantar footprints pictures, surgery lengthened the operated foot relatively to the opposite one in unilateral disorders. Preoperative measures found a median foot length of 22.2 cm, IQR [20.1; 22.9] compared to 23.0 cm, IQR [21.5; 24.5] postoperatively, without any significant difference ($p = 0.29$) (Fig. 2).

3.3. Functional outcomes

Global outcomes assessed by the WS score were very good or good, fair and poor in respectively 58%, 23% and 19% of the feet (Table 2). AOFAS score medians and IQR were 95.5, IQR [84.3; 97.8] for the hindfoot; 75, IQR [61.5; 80] for the midfoot; 100, IQR [95; 100] for the hallux and 92, IQR [91.5; 94] for the lesser toes.

3.4. Radiological outcomes

Radiological outcomes are summarized in Table 4. Significant improvements were found for the cavus deformity, the hindfoot varus deformity and the forefoot pronation deformity with a mean amount of correction of 92%, 58% and 73% respectively (Fig. 6).

3.5. Complications

Overcorrection with flatfoot (Meary angle $\leq 10^\circ$) was observed in 6 feet (4 patients, 23%) with Meary angle $\leq 15^\circ$ in 3 feet. Among these 4 patients, 2 were painful at last follow-up. After the exclusion of these feet, postoperative Meary angle improvement was still significant (mean angle 3.3° , range $[-14; 20]$, $p < 0.0001$).

Three feet (11.5%) required additional surgical procedures, including one patient with overcorrection, leaving a complication rate of 27% (5 patients). One patient had a skin necrosis which required early cast removal leading to CVF relapsed requiring a triple arthrodesis. At last follow-up, 21 years after initial surgery, this patient was the poorest result of the cohort. No other patient had wound healing issues, explained by the 2 incisions approach. A second patient required a complementary first metatarsal dorsal closing wedge osteotomy in one foot, which was not initially performed, for painful callosity under the first metatarsal head with plantar sticking. The third patient who required iterative surgery had hardware removal unilaterally for pain next to the staple. There was no non-union.

4. Discussion

This study is the first to describe the revisited Meary's procedure and its results, which was associated to the Dwyer

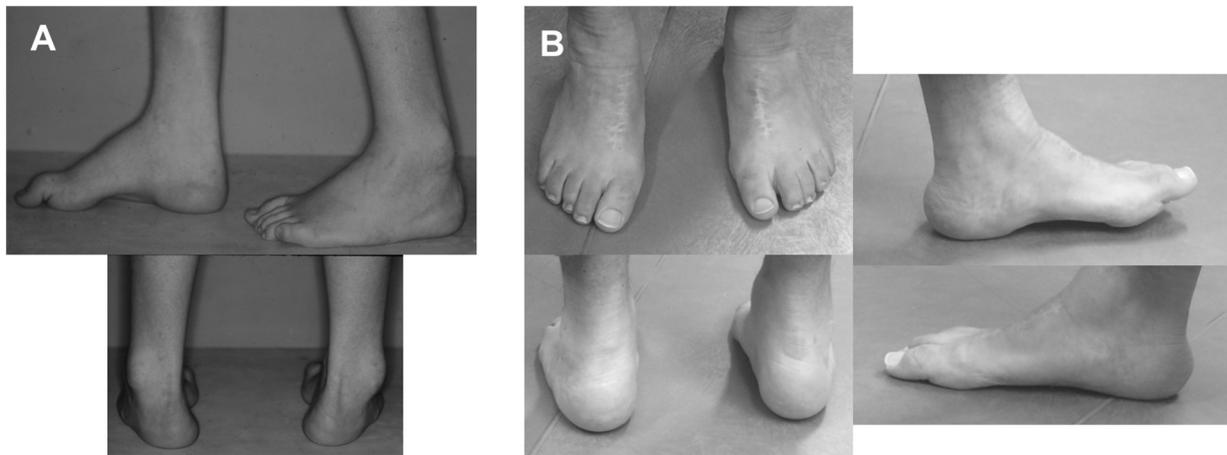


Fig. 5. Clinical outcomes after 32-years. Surgery was performed at the age of 14 years (A). At last follow-up, the patient was not limited in recreational activities (B). WS score was very good for both feet. All components of the AOFAS score were excellent: 97 and 100 for the left and the right hindfoot respectively, 100 for both midfoot and hallux and 92 for the lesser toes bilaterally.

Table 2
Global outcomes according to the rating scores.

Pt. No	Gender	Age at surgery	Wicart and Seringe score		AOFAS score			
			Results	Causes of poor results	Hindfoot	Midfoot	Hallux	Lesser toes
1	M	19	Good					
2	M	15	Fair					
3	F	18	Poor	Sprains, under correction 15°, no hindfoot deformity	55	83	100	100
4	M	15	Poor	Pain, under correction 20°, varus				
5	F	14	Good					
6	M	16	Very good		94	72	80	90
7	M	14	Very good					
8	F	20	Very good					
9	F	16	Good					
10 (Bilateral)	F	15	Fair					
			Poor	Pain, overcorrection 18°				
11 (Bilateral)	M	16	Good		82	85	50	68
			Good		82	75	75	68
12	F	16	Good					
13	M	15	Fair					
14 (Bilateral)	F	15	Fair					
			Fair					
15	M	17	Poor	Pain, sprains, overcorrection 14°, triple arthrodesis	54	48	31	18
16	M	14	Fair		87	95	90	90
17 (Bilateral)	F	14	Good		55	48	62	64
		15	Very good		55	48	57	64
18 (Bilateral)	F	14	Good					
		15	Poor	Sprains, overcorrection 20°				
19	F	14	Very good					
20 (Bilateral)	M	14	Very good		97	100	100	92
		15	Very good		100	100	100	92

calcaneal osteotomy and 1st metatarsal bone extension osteotomy for the treatment of the neurological CVF if necessary. All these procedures addressed to the specific components of the deformity. Results of the current study reported satisfactory functional and clinical outcomes, with a significant improvement of pain, callosities and sprains (Table 3). Furthermore, postoperative radiographs also reported significant improvements (Table 4).

Table 3
Clinical outcomes (number of feet).

	Preoperatively	Postoperatively	p-Value
Pain	14	7	0.08
Calluses	14	4	0.0006*
Sprains	16	4	<0.00001*
Claw toes	11	9	0.7

* Significant result $p < 0.05$.

This revisited procedure should therefore be considered as a good alternative to other CVF surgical procedures.

4.1. Surgical improvement: the revisited technique versus the original technique

As previously mentioned, the original technique did not meet all criteria to correct the 3D components of the deformity [4,15]. First, the triangular shape with dorsomedial base increased forefoot adduction. Second, hindfoot varus was not corrected, resulting in ankle instability found in 19% of the original cohort. This issue was resolved by the systematic Dwyer calcaneal osteotomy, which is also recommended by Japas [11,14]. Nonetheless, any other extra-articular calcaneal osteotomy such as a lateralizing osteotomy or a derotation osteotomy could have been used for the correction of the varus [25,26]. Third, remaining plantar sticking of the 1st metatarsal head was responsible of 30%

Table 4
Pre- and postoperative radiographic measurements (degrees).

Alignment measures		Preoperatively		Postoperatively		p-Value
		Mean	Range	Mean	Range	
Sagittal plane	Meary angle	16.7°	[4;28]	-1.4°	[-21;20]	<0.0001*
	Stacking angle	23.9°	[5;38]	14.9°	[2;24]	<0.0001*
	Calcaneal pitch	23.6°	[10;36]	12.5°	[0;34]	<0.0001*
Frontal plane	TCA	20.5°	[0;40]	26.8°	[14;42]	0.01*
	TM1	20.7°	[0;62]	8.1°	[0;28]	0.0007*
	CM5	14.9°	[0;28]	73.4°	[0;32]	0.0003*

TCA: talocalcaneal angle; TM1: talo-first metatarsal angle; CM5: calcaneo-fifth metatarsal angle.

* Significant result $p < 0.05$.



Fig. 6. Preoperative and postoperative radiographic outcomes 30 years after surgery. The patient did not complain about any symptoms and had an unlimited walking perimeter despite the staples broken spike.

of painful plantar callosities in the original cohort and 60% in the study by Naudi et al. [15,27]. Therefore, a 1st metatarsal bone extension osteotomy is recommended and should be based on perioperative findings [10,14]. This procedure was presently performed in 61.5% of the feet. Finally, the preservation of a plantar hinge combined with a plantar fascia release were also mandatory to avoid foot shortening [10,20]. This issue was reported by Wülker and Hirschler without providing a particular reason and plantar fascia release was not performed in their study [28].

4.2. Functional objectives of CVF bone surgeries: strengths and weaknesses

Sammarco and Taylor pointed out that the treatment of CVF must focus on pain-relief, correction of the deformity and function improvement [18]. Nevertheless, since CMT disease is slowly degenerative, other deformities might develop over time (claw toes) due to the progressive muscular impairment and the foot will be painful again [29]. Pain also depends on both the disease advanced stage and the severity of the deformity. The persistence of claw toes, which was not significantly corrected in the present study, was involved in painful conflicts with footwear and therefore pain disappeared in only 50% of the patients by the correction of the deformity.

Good foot function can only be maintained in the long term if joint motion is preserved by the surgery [29,30]. The approach inevitably sacrificed the cuneiform-navicular joints. As demonstrated by Lundgren et al., this joint has a significant range of motion (11.5°, 10.4° and 6.2° respectively in the sagittal, frontal and

transverse planes) [31]. This is no doubt why higher rates of very good results were not identified. However, the revisited procedure is a salvage procedure for an already fixed deformity, with a lack of joint motion. Furthermore, the concept of “joint-sparing” procedure has been questioned by Zide and Myerson [29,32]. These authors demonstrated that results of an arthrodesis that creates a plantigrade foot are superior to those of a theoretical full joint-sparing surgery that preserves a limited amount of motion and fails to gain complete correction.

Compared to other surgical procedures, the revisited procedure had similar functional outcomes (Table 5) [13,14,16,19,27]. The two approaches described by Jahss and Wülker required the same amount of resection as the revisited procedure [13,28]. Nonetheless, Wülker and Hirschler did not stipulate the exact location of the deformity [28]. The tarsometatarsal dorsal wedge osteotomy as described by Jahss should not be recommended because it is too distal of the apex of the deformity and all metatarsal joints are also sacrificed [13]. Similarly, the Akron osteotomy or any osteotomy located more distally (metatarsal osteotomies) should also be avoided for the same reasons except for the 1st metatarsal extension osteotomy that corrects plantar sticking [19]. Finally, the triple arthrodesis must be proscribed as first option in patients of that age because of the risk of degenerative osteoarthritis in the adjacent joints [6–9]. The literature had demonstrated poor functional outcomes in the long term and is certainly related to the association of the arthrodesis with the sensory loss induced by the disease. Triple arthrodesis should remain the last therapeutic option after failure of other bone procedures, as reported for one patient in the present study.

Table 5
Midtarsal osteotomies: comparison to the literature.

Studies	Osteotomy	Deformity	No. of feet	Follow-up (Years)	Etiologies	Results	Complications
Current study, 2017	Modified Meary osteotomy	Rigid pes cavovarus	31	5.7	Neurological	Good and very good 61%	1 Wound necrosis 3 Overcorrection >15° 1st Metatarsal osteotomy
Japas [14]	Tarsal V- osteotomy	Pes cavus	18	6	12 Neurological 2 Unknowns	Good 35% Very good 65%	2 Overcorrection 2 Undercorrection 1 Osteoarthritis 1st Metatarsal osteotomy
Jahss [13]	Tarsometatarsal truncated wedge arthrodesis	Pes cavus and forefoot equinovarus	34	6.3	6 Neurological 5 Residual clubfoot 7 Idiopathic 7 Equinus with forefoot adduction	Completely satisfactory	1st Metatarsal osteotomy
Weiner [19]	Akron dome midfoot osteotomy	Rigid pes cavus	139	7.6	86 ICF 19 Neurological 5 Arthrogyroses 7 Congenital metatarsus adductovarus 22 Others	76% Of satisfactory (pain free, foot alignment)	1 Skin slough
Naudi et al. [27]	Initial Meary osteotomy	Rigid pes cavovarus	39	9.8	20 Neurological 14 Idiopathic 2 Residual clubfoot	AOFAS 69.2/100 Metatarsalgia 60%	74.2% Osteoarthritis
Mubarak and Van Valin [16]	Combined bones procedures (1st ray, cuboid, metatarsal, calcaneus, cuneiform) + plantar fasciotomy + tendon transfer	Rigid pes cavovarus	13	4.1	8 Neurological 3 Idiopathic 2 Clubfeet	35% Very good 60% Good 5% Poor	2 Wounds dehiscence 1 Revision

4.3. The management of muscular imbalance

Some authors had more recently recommend to combine tendon transfer to bone procedures (Peroneus Longus to Brevis tenodesis/transfer most frequently to restore an eversion effect) according to the muscular imbalance found in neurological etiologies [1,16,30]. Mubarak and Van Valin recommended a Peroneus Longus to Brevis tendon transfer but their study included a large variety of non-progressive diseases and heterogeneous feet deformities [16]. However, the distal compartment is the first to be affected in progressive diseases, followed later by the proximal compartment with uncertainty regarding the progression speed. Therefore, the efficiency of the tendon transfer over time cannot be estimated, which is the reason why we did not associate these procedures. The split tibialis posterior transfer is another procedure considered in the literature for the correction of the hindfoot varus [33]. Nonetheless, all the patients considered in the present study had a fixed deformity, that cannot be correct by a soft-tissue procedure and the varus was adaptive of the forefoot and midfoot deformity. The role of tendon transfer (even combined with a triple arthrodesis) to reduce osteoarthritis is not yet establish and need further investigation in the CMT patients that become less ambulant over time.

4.4. Radiographic outcomes in CVF bone surgeries

Even though it has been reported in the literature that there is a lack of correlation between functional and radiological outcomes, a significant correction of all radiographic measures was found in the present study (Table 4) [30]. Despite the significant correction of the Meary's angle, 23% of the feet had an overcorrection into a

flatfoot, induced by an excessive dorsal bone resection. This complication has also been reported by Japas [14]. Therefore, we recommended to gradually remove bone in order to apply the correct amount of correction. Forefoot adduction (CM5 and TM1 angles) was also efficiently corrected. Nonetheless, the amount of forefoot adduction was mostly not reported in the literature, though it is essential for footwear [10,14,28]. Significant improvement of the stacking angle, expressing the amount of forefoot pronation correction, was also reported and correlate with 1st metatarsal osteotomies performed in 61.5% of the series. Finally, dorsoplantar TCA was surprisingly improved. Indeed, since the procedure was extra-articular regarding the subtalar joint, correction of forefoot pronation allowed the improvement of the talo-calcaneal divergence, within the limit of remaining motion.

4.5. Limitations

The main limitations of this study are its retrospective nature and the lack of clinical and functional data, that limited the study to four parameters. Therefore, the frequency of other symptoms could not be systematically found in medical records. Second, the postoperative WS score was performed for the functional assessment and the AOFAS score was only available for 10 feet. Functional assessment by the WS score was address by the lack of pain. This is certainly less precise than the functional assessment offered by the AOFAS rating system, which is currently the validated score predominantly used in the literature [23]. However, due to the progressive nature of the disease for most patients, functional outcomes cannot be exclusively attributed to surgery. Last limitation is the lack of control group and the

comparison to a validated clinical foot assessment index such as the Foot Posture Index [34]. Therefore, our results cannot conclude to the superiority of the procedure over another. Nonetheless, regarding the Foot Posture Index, its reliability in CVF has not been validated yet and its relevance in pathological cases has been questioned more recently [35]. Furthermore, regarding literature on CVF deformities, incomplete clinical and radiological data were also provided and most of the series were heterogeneous unlike the current series (Table 5) [13,14,16,19,27].

5. Conclusions

The revisited Meary's closing wedge dorsal tarsectomy combined with the Dwyer calcaneal osteotomy and plantar fascia release, completed with a first metatarsal closing wedge osteotomy if required, is an efficient salvage procedure for the correction of fixed adolescent neurological CVF. The combination of these different techniques led to significant clinical improvements and radiological correction of the deformity, while avoiding a triple arthrodesis. Functional outcomes were more limited but similar to previous studies. Recent literature suggests to combine tendon transfers to bone procedures. This was not performed in the study despite muscular imbalance, but required further investigations.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

The authors declare to have no conflict of interest.

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