Zagazig University Medical Journal
www.zumj.journals.ekb.eg
https://doi.org/10.21608/zumj.2024.234154.2873 Volume 30, Issue 1.6, September 2024, Supplement Issue
Manuscript ID ZUMJ-2310-2949 (R1) DOI 10.21608/ZUMJ.2023.241857.2949

ORIGINAL ARTICLE

Surgical Treatment of Foot Deformity in Charcot Marie Tooth Syndrome

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Submit Date 19-01-2023
Revise Date 21-10-2023
Accept Date 25-10-2023

ABSTRACT

Background: Foot deformities were regularly seen in patients with Charcot Marie tooth disease (CMT), also, orthopedic surgery frequently needed. Right now, there was no proof based on guideline for surgery treatment and few researches studied the long-term effects of surgery intervention. This study aimed to assess the early functional and anatomical results of surgical treatment of foot deformities in Charcot Marie Tooth syndrome patients. **Methods:** A prospective study was carried at the Orthopedic Surgery Department, Faculty of Medicine, Zagazig University on 18 cases with Charcot Marie tooth disease (CMT) in the period from January 2022 to January 2023 to evaluate surgery treatment for Charcot-Marie-Tooth syndrome's foot deformity. **Results:** There is highly statistically significant decrease (improvement) in the Meary's angle, the AP Talocalcaneal angle (Kite's angle), the lateral talocalcaneal angle, the calcaneal pitch angle, the AP talo first metatarsal angle. 38.9% of patients had a good outcome, 22.2% had excellent outcome, 27.8% had a fair outcome while only 11.1% had a poor outcome. **Conclusion:** Surgical foot deformities management in a CMT patients has a significant reduction of pain, alignment of foot and patients satisfaction.

Keywords: Charcot Marie tooth disease; foot deformity, Osteotomy.

INTRODUCTION:

Charcot Marie tooth disease (CMT) is a clinical hereditarily heterogeneous problems with expected predominance of 1 in 2500 persons and is a well-known acquired neuromuscular disorder. CMT is length-subordinate neuropathy described by leisurely moderate shortcoming and tangible misfortune in upper and lower appendages. Foot distortions, for example, pes cavus and hammertoes, were successive entanglements in patients with CMT and were seen in 71% of patients with CMT. The principal reason for cavovarus foot deformity is the muscle imbalance between the more fragile tibialis foremost overwhelmed by the more grounded peroneus longus muscles and the shortcoming

of characteristic foot muscles. Orthopedic surgeries are frequently needed to treat severe foot deformities and in a in a new report, 30% of patients with CMT underwent correction surgery. The goal of correction surgery deformities correction of bones and imbalance of muscle producing a very much adjusted foots. Surgeries is mostly indicated in case of conservative treatment failure, for instance, physiotherapy and orthotics **[1]**.

Charcot-Marie-Tooth illness is the absolute normal finding related with a cavus foot. The imbalance including intrinsic & extrinsic muscles were proposed as the principal pathogenetic reason for cavus foots sickness. The objective of surgical management is correction deformity to get adjusted foots. When a flexible defect is present without degenerative arthritis, preserving the overall motion range of foots and ankle is indicated **[2].**

In the CMT diseases, front tibial muscle, peroneus brevis and intrinsic muscular weakening are the main causes of the deformity, with the peroneus longus overcompensating as a result, bringing about articulated pronation and hindfoot varus Delicate tissue decay and auxiliary contractures can additionally convolute the generally fixed deformation. So, the correction of soft-tissue is important to recreate physiologic balance of soft-tissue and correction of pronation and foot drop. The correction of soft-tissue is essential for restoring balance of soft-tissue including tenodesis or active tendon transfers, when the active muscle control is impossible. Despite the fact that a tenodesis prevents dynamic soft-tissue balance, the correction of soft-tissue can be obtained to prevent drop of foot. The posterior tibial tendon can be transferred to the foot dorsum as one of these tendon transfers to correct the anterior tibial tendon deficient, which permit active dorsiflexion **[3]**.

Surgery is indicated in case of failure of conservative measures, like physical treatment and orthoses use, which considered the first-line management of foot destoration in CMT. A few methods were mentioned such as soft tissue methods either alone or in combination for management of foots deformity, for example tendon transfers, tendon lengthening and plantar fascia release are procedures utilized for mild flexible deformities. Osteotomies correction, like calcaneal osteotomy and first metatarsal osteotomy, can used in case of great deformity correction or for a rigid deformity. Fusion methods, like triple arthrodesis, mostly reserved for severe degeneration of joints disorder & deformities. There is no one surgical procedure accomplishes this large number of objectives at the same time, and careful choices are typically individualized **[4]**.

We hypothesize that surgical treatment of foot deformities in Charcot Marie Tooth syndrome has a better functional radiological and correction outcome. Thus in the current study was aimed to assess the early functional and anatomical results of surgical treatment of foot deformities in Charcot Marie Tooth syndrome patients.

METHODS

This prospective study was carried in the Orthopedic Department of Surgery, Zagazig University's Faculty of Medicine on 18 Charcot Marie tooth disease (CMT) patients during the period from January 2022 to September 2023. After trying conservative procedures like physiotherapy and orthotics without success or to the patient's satisfaction, all patients were referred to the foot and ankle orthopaedic surgery section. Final assessment of any patient was done after 21 months of follow up.A written informed consent was taken from all participants and the study was approved by the Zagazig University Faculty of Medicine's Research Ethical Committee (ZU-IRB#9525-27-4-2022). The study was carried out according to the Ethical code of the World Medical Association (Declaration of Helsinki) for Studies including humans.

Inclusion criteria were**:** failure of conservative management, patients with foot symptoms especially deformity**.**

Exclusion criteria were: unfit for surgery, Charcot Marie Tooth syndrome without foot symptoms.

All participants were subjected to the following:

Comprehensive history taking including CMT, injuries and ankle instability, musculoskeletal pain & pain distribution, callosities distribution and skin condition. Full clinical and neurological examinations. Radiological evaluation including X-ray (anteroposterior, lateral and oblique views) on the infected side of ankle & foot X-ray and CT. Electromyography and nerve conduction studies. Operative data were recorded involving surgery type, operation time and complications.

Surgical technique (Mubarak and van Valine technique):

1) First ray osteotomies:

Opening wedge osteotomy of the medial cuneiform and closing-wedge osteotomy of the first metatarsal. The wedge from the metatarsal was inserted into the plantar area of the medial cuneiform later. These osteotomies were fixed with one to 2 K-wires.

2) Cuboid closing-wedge osteotomy:

A longitudinal lateral incision overlying the cuboid was performed. All patients underwent a closing wedge to correct the forefoot varus and to aid the dorsiflexion of the forefoot. The removed wedge can be used as supplemental

bone grafting for the cuneiform as necessary. This osteotomy was fixed with 1 K-wire.

3) Metatarsal osteotomies:

After the above osteotomies of all the patients if there is plantar prominence of the second and third metatarsal heads, dorsal closing-wedge metatarsal osteotomies was done in three patients. A single incision was made over the dorsum of the foot between the second and third metatarsals where closing abduction wedge osteotomies at the base of these metatarsals were performed. These were fixed individually also with intramedullary K-wires.

4) Calcaneal osteotomy:

The calcaneus was exposed subperiosteally through a lateral incision a wide wedge of done based laterally was resected and it was large enough to correct the heel varus without injury to the peroneal tendon The heel was placed into the corrected position and the incision was closed with interrupted sutures and was fixed the osteotomy with a kirschner wire.

5) K-wire fixation:

All of the osteotomy sites were then fixed with K-wires starting with the first metatarsal where pinning of the first ray and then the cuneiform is accomplished with the bone graft placed in the open wedge osteotomy of the cuneiform. The pins were inserted in position in the cuboid and metatarsals osteotomies for sequential pinning. Finally, the calcaneus was positioned and also fixed usually with 2 K-wires from a plantar-to-dorsal direction.

6) Plantar fasciotomies:

Plantar fasciotomies were performed through a small percutaneous incision if fascia was tight after the osteotomies and this was needed for all patients.

7) Peroneus longus-to-brevis transfer:

The peroneus longus tendon was released just under the cuboid and reattached to the peroneus brevis to decrease the plantar pull of the first metatarsal for correction of the cavus and to aid the abduction on the forefoot. This step was performed after the cuboid closing wedge osteotomy.

8) Clawing corrections

The Jones procedure was done for correction of the big toe deformity and extending the metatarsophalangeal joint. Clawing of the lesser toes was donethrough a dorsal incision of metatarsophalangeal joint and cutting extensor digitorum brevis tendon with deep and lateral to extensor digitorum longus tendon; the later was lengthed by Z plasty and capsule was released to correct the metatarsophalangeal hyperextension. Flexion of proximal IP joint was corrected through a dorsal cresent incision over proximal IP joint and the head of proximal phalanx was excited to correct the flexion deformity correction of clawing was needed in four feet.

Summary of all deformities and corrections

In all patients we used Surgical Mubarak and van Valine technique (medial cuneiform and closing-wedge osteotomy of the first metatarsal, Cuboid closing-wedge osteotomy and Peroneus longus-to-brevis transfer) to correct of cavus and to aid the dorsiflexion and abduction on the forefoot. Calcaneal osteotomy was used to correct the heel varus.

In four feet where we used the Jones procedure for clawing deformities correction.

Postoperative care:

After the bones were fixed and wounds closed, all the patients were placed in a below-knee cast, which was split. The casts and pins were removed under local anaesthesia at outpatient clinic about 4 weeks, and radiographs were obtained. A new below-knee walking cast is applied for a second 4 weeks, for a total of 8 weeks of immobilization.

Statistical Analysis: Information gathered since forever ago, fundamental clinical assessment, research facility examinations and result measures coded and entered. Information examination was performed utilizing the product SPSS (Measurable Bundle for the Sociologies) rendition 26. Shapiro-Wilk test is utilized to confirm suppositions for using in parametric tests. Quantitative factors were depicted utilizing their means and standard deviations or middle and reach as per sort of information. To look at quantitative information between two gatherings, to quantify changes in one variable between two time marks, Wilcoxon marked rank test was utilized. The level factual importance was considered at $P < 0.05$. Exceptionally tremendous distinction was available if $P \leq 0.001$.

RESULTS

Table (1) indicated that the study group's average age was (12.5±3.1) ranged between 9 to 19 years with the majority of them (83.3%) were less than or equal 15 years.

Table (2) indicated that more than half of them (55.6%) were males, and females (44.4%) were.

Table (3) showed a high statistically significant decrease (improvement) in the Meary's angle from (28.1±5.1) ranged between 15 to 35 to be (3.1±6.7) ranged between 0 to 22 (p-value<0.001) with a percent of improvement (89.9%) ranged between 42.9% to 100.0%.

Table (4) revealed that there is a highly statistically significant change (improvement) in the AP Talocalcaneal angle (Kite's angle) from (10.6±1.15) ranged between 8 to 12 to be (20.3±4.1) ranged between 9 to 26 (p-value<0.001) with a percent of improvement (100.25%) ranging from (18.18% to 177.8%). There is high statistically significant improvement in the lateral talocalcaneal angle from (16.7±3.5) ranging from 10 to 22 to become (29.8±4.3) ranged between 17 to 35 (p-value<0.001) with a percent of

improvement (84.7%) ranging from (36.4%-170.0%). There was a high statistically significant decrease (improvement) in the calcaneal pitch angle from (33.5±2.3) ranged between 25 to 36 to be (19.5±3.3) ranged between 17 to 32 (p-value<0.001) with a percent of improvement (41.6%) ranging from (11.1%-50.0%). There was highly statistically significant improvement in the AP talo first metatarsal angle from (15.2±1.9) ranging from 13 to 19 to become (6.8±2.3) ranged between 5 to 13 (p-value<0.001) with a percent of improvement (55.1%) ranging from (13.3%-68.8%).

Table (5), showed that about one-third of the studied group (38.9%) had a good outcome, 4 cases (22.2%) had excellent outcome, 5 cases (27.8%) had a fair outcome while only two cases (11.1 %) had a poor outcome.

Table (6) shows that 66.7% of the study group had no complications, three cases (16.6%) had a superficial infection treated by parental antibiotics for 2 weeks, two cases (11.1%) had residual deformity, one case (5.5%) had a painful neroma and one case (5.5%) had suedex atrophy.

Table (1): Age distribution among the study group

Characteristics	***The studied group No=18 (%)***
Age Mean ± SD Median (Range)	 12.5±3.1 12 (9-19)
Age group ≤ 15 years >15 years	 15 (83.3%) 3 (16.7%)

Table (2) : Sex distribution of the studied group

Sex	**The studied group**	
	No= (18)	**%**
Male	10	55.6%
Female	8	44.4%

Table (3): Comparing pre- and post-operative (Lateral talar - 1st metatarsal angle) Meary's angle in the study group.

Meary's angle	Preoperative Mean ± SD Median (Range)	Postoperative Mean ± SD Median (Range)	Paired t-test	p-value
Meary's angle	28.1±5.1 29.5 (15-35)	3.1±6.7 87.5 (0-20)	21.4	0.001**
Percent of improvement	89.9% (42.9%-100.0%)			

**Statistically highly significantly different.

Table (4): Comparing pre- and post-operative AP Talocalcaneal angle (Kite's angle), Lateral Talocalcaneal angle, Calcaneal pitch angl, AP talo first metatarsal angle among study group.

	Preoperative Mean ± SD Median (Range)	Postoperative Mean ± SD Median (Range)	Paired t-test	p-value
AP Talocalcaneal angle (Kite's angle)	10.6±1.15 11 (8-12)	20.3±4.1 20.5 (9-26)	9.4	0.001**
Percent of improvement	100.2% (18.18%-177.8%)			
Lateral Talocalcaneal angle	16.7±3.5 17.5 (10-22)	29.8±4.3 30 (17-35)	13.3	0.001**
Percent of improvement	84.7% (36.4%-170.0%)			
Calcaneal pitch angle	33.5±2.3 34 (25-36)	19.5±3.3 19 (17-32)	16.3	0.001**
Percent of improvement	41.6% (11.1%-50.0%)			
AP talo first metatarsal angle	15.2±1.9 15 (13-19)	6.8±2.3 6 (5-13)	14.9	0.001**
Percent of improvement	55.1% (13.3%-68.8%)			

Table (5): Postoperative functional outcome among the studied group.

The outcome	The studied group	
	No= (18)	%
Poor	2	11.1%
Fair	5	27.8%
Good	7	38.9%
Excellent	4	22.2%

Table (6): Postoperative complications in the study group.

Complications	The studied group	
	No= (18)	%
No	12	66.7 %
Superficial infection	3	16.6 %
Painful neroma	1	5.5 %
Residual deformity	2	11.1 %
Suedex atrophy	1	5.5 %

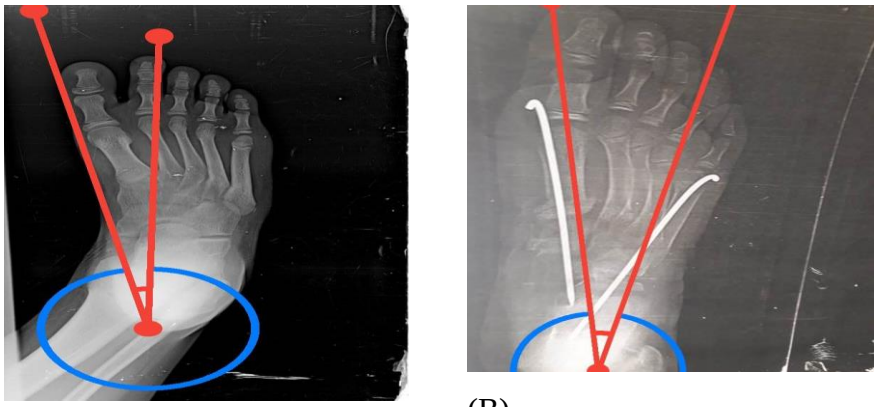
NB., One patient might have more than one complication.

(A) (B)

Figure (1) preoperative (A) Shows an elevated medial longitudinal arch of the RT foot (B) Shows hindfoot deformity (hindfoot varus) of both feet.

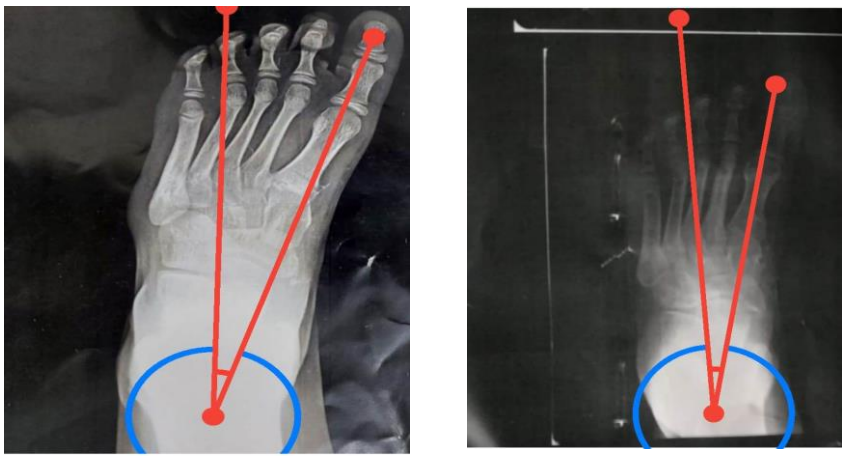
(A) **(B)**

Figure (2) postoperative follow-up A) shows a normal medial longitudinal arch of both feet after 6 months (B) Shows near the normal heel of both feet after 6 months.



(A) (B)

Figure (3) : A) pre-operative x-rays AP view of RT foot shows Talocalcaneal angle (Kite's angle) = 10° , B) post-operative x-rays AP view of RT foot shows Talocalcaneal angle (Kite's angle) = 22° after 1 month follow-up



(A) (B)

Figure (4) A) pre-operative x-rays AP view of Lt foot shows talo first metatarsal angle = 17°, B) Post-operative x-rays AP view of LT foot shows talo first metatarsal angle = 15°

DISCUSSION

The rate of peripheral neuropathies known as Charcot-Marie-Tooth disease (CMT), also known as genetic sensorimotor neuropathy, ranges from 15.2 to 40 per 100,000 people worldwide, with a prevalence of about 28.2 per 100,000 in Spain. Demyelination and axonal degeneration of the peripheral nerves were primary symptoms, and its clinical onset typically occurs in the first twenty years of life **[5]**.

Cavovarus foot disfigurement frequently results from strong unevenness. This lopsidedness could be of neuromuscular, horrible, innate or idiopathic beginning. Relative shortcoming of the peroneus brevis and tibialis foremost muscles major areas of strength for with back and peroneus longus muscles cause plantarflexion of the main metatarsal bone and varus of the hindfoot. Fringe neuropathy causes extra shortcoming of natural foot muscles bringing about a paw foot deformation. Clinical show that the indicative cavovarus foot deformation incorporates equinus, frequently brought about by flexion of the forefoot and not the lower leg joint, increased rearfoot varus and longitudinal curvature. Pain and callus formation were caused by the migration of the plantar fatpad at the metatarsophalangeal joints (MTP) and by stand impact as a result of deformities. **[6]**.

Several surgical methods were suggested either separately or in combination, to address foot abnormalities. which involve soft tissue methods, which manage the muscle imbalance and correction of osteotomies which treat deformities of bones. Careful choices were typically individualized as there was no proof put together rules with respect to the sort or timing of the medical procedure **[7]**.

The main cause of cavovarus foot deformity is muscular imbalance between the weaker tibialis anterior overpowered by the stronger peroneus longus muscles and weakness of intrinsic foot muscles. Currently, no proven medical treatment exists to reverse or slow the natural disease process for the underlying disorder. Orthopedic surgery is required to correct severe pes cavus deformities, scoliosis, and other joint deformities.**[4]**.

Foot deformities are frequently observed in patients with Charcot Marie tooth disease (CMT) and orthopedic surgery is often required. Currently there is no evidence-based guideline on surgical management and only a few studies which have evaluated long-term outcomes of surgical procedures. So this study aimed to assess the early functional and anatomical results of surgical treatment of foot deformities in Charcot Marie Tooth syndrome patients.

Up to knowledge, this is the first prospective study assessing the outcome and evaluation of surgical treatment of foot deformity in Charcot Marie Tooth syndrome. Direct comparison with previous studies is difficult since all the studies so far performed have been retrospective and only few studies have looked at the outcome of foot surgery in CMT.

This prospective study was carried out in the Orthopedic Surgery Department, Faculty of Medicine, Zagazig University on 18 patients with Charcot Marie tooth disease (CMT) during period from January 2022 to September 2023. Final assessment of any patient was done after 21 months of follow up.

The current study indicated that the study group's average age was (12.5±3.1) ranged between 9 to 19 years with the majority of them (83.3%) were less than or equal 15 years. more than half of them (55.6%) were males, and (44.4%) were females, which comparable to Zahid et al., [8] who found that in their study there were 21 males (70%) and 9 females (30%) with a mean age of (9.5 ± 3.44 years); ranged between 5 to15 years. Also, similar to our results Ramdharry et al. [1] who found that the mean age was 39.5 ±14.1, ranged from 19 to 79. Most patients (18/25, 72%) were males.

The current study revealed that there is high statistically significant decrease (improvement) in the Meary's angle from (28.1±5.1) ranged between 15 to 35 to be (3.1±6.7) ranged between 0 to 22 (p-value<0.001) with a percent of improvement (89.9%) ranging from (42.9%-100.0%), consistent with our results Shim et al., [9] found that the Meary angle was reduced from 10.7±5.5 degrees preoperative to 5.5±3.4 degrees postoperative with a significant difference (p=0.001). our results supported also by the study of Saragas & Ferrao [10], Pansini et al., [5] and Zhou et al., [11], where they concluded that there is a statistically significant difference a statistically significant difference in Meary's angle pre-operative and postoperatively (p <0.001).

The current study showed that there is highly statistical significant improvement in the AP talocalcaneal angle (Kite's angle) from (10.6±1.15) ranged from 8 to 12 to (20.3±4.1) ranged from 9 to 26 (p-value<0.001) with a percent of improvement (100.25%) ranging from (18.18% to 177.8%). Which comparable to Ozan et al., [12] and Joo et al., [13] whom concluded that there was highly statistically significant improvement in the AP talocalcaneal angle (Kite's angle) preoperative and postoperative respectively (P=0.0001).

The current study concluded that, there is a high statistical significant increase (improvement) in the lateral talocalcaneal angle from (16.7±3.5) ranged between 0 to 22 to be (29.8±4.3) ranged between 17 to 35 (p-value<0.001) with a percent of improvement (84.7%) ranging from (36.4%-170.0%). Which s comparable to Zahid, et al., [8] who reported that the mean Talo-calcaneal angle improved postoperative from 35.5 ± 7.22 to 26.3 ± 5.55 preoperative with high statistically significant (p=0.001).

The current study found that there is highly statistically significant decrease in the calcaneal pitch from (33.5±2.3) ranged between 25 to 36 to be (19.5±3.3) ranged between 17 to 32 (p-value<0.001) with a percent of improvement (41.6%) ranging from (11.1%-50.0%). Which comparable to Chen et al., [14] who concluded that there is significant decrease in the calcaneal pitch pre and postoperative (30.93° postoperative to 27.43° preoperative; P = .005) and study of Shim et al., [9] who found that the calcaneal pitch angle improved from 26.4±4.3° preoperative to be a 25.2± 4.1° postoperative; with no statistical significance which may be contributed to the size sample of patients involved in their study (p=0.066).

Our study showed that there is highly statistically significant improvement in AP talo first metatarsal angle from (15.2±1.9) ranging from 13 to 19 to become (6.8±2.3) ranging from 5 to 13 (p-value<0.001) with a percent of improvement (55.1%) ranging from (13.3%-68.8%) which coincide with Wen et al. [15] whom concluded that there is highly statistically significant improvement in AP talo first metatarsal angle from 17.2±4.7 preoperative to 6.8±2.4 postoperative (P <0.001). Also, Sanpera et al., [16] who found a high significant decrease in AP talo first metatarsal angle postoperatively (P <0.001).

The current study reported that one-third of the studied patients (38.9%) had a good outcome, 4 cases (22.2%) had excellent outcome, 5 cases (27.8%) had a fair outcome while only two cases (11.1 %) had a poor outcome which comparable with the study of Pansini et al., [5] who found that the postoperative AOFAS score in 13 patients (61.9%) was excellent, in five patients (23.8%) was good, in two patients (9.6%) was fair and one patient (4.7%) was poor. Also, our results supported by Zhou et al., [11] who concluded that 11 cases (64.7%) were very good, 4 cases (23.5%) were good and 2 cases (11.8) were fair.

The current study concluded that 66.7% of the study patients had no complications, three cases (16.6%) had a superficial infection treated by parental antibiotics for 2 weeks, two cases (11.1%) had residual deformity, one case (5.5%) had a painful neroma and one case (5.5%) had suedex atrophy, which comparable with the study of Carantini et al., [17] who reported that 5 patients (8%) suffered from postoperative complications. Two patients of them (aged 45 and 51) had surgical wounds; two patients (ages 55 and 65 years) had detachment of a transposed tendon, and painful retraction of the plantar fascia was recorded in one patient (aged 41).

Limitations: The study has some limitations. We were able to assess a large percentage of patients up to two years after surgery but the number of patients assessed after three to four years reduced due to loss to follow-up and follow-up sessions falling outside of the data collection window. Therefore, only conclusions for short-term postsurgical outcomes can be drawn. Second the cohort of patients studied was small, reflecting the rarity of the disease and was not homogenous in terms of genotype with various of forms of CMT in the cohort. As expected, the majority of patients evaluated had CMT1A, as this is the most common form of CMT, and most of the conclusions are applicable to CMT1A but are limited for the other rarer forms of CMT

CONCLUSIONS

Surgical foot deformities management in a CMT patients has a significant reduction of pain, alignment of foot and patients' satisfaction.

Future prospective and Recommendations: Surgery resulted in significant improvement of pain, foot alignment, callosities and quality of life. Although evidence-based guidelines on surgical management in CMT are not available, this study suggests that surgery for foot deformity in adults with CMT in a specialized foot and ankle unit is beneficial. Further studies looking at longer follow-ups and larger number of patients in different surgical settings will be required to provide further guidance on surgical management and to explore this aspect as CMT is a slowly progressive disease and the extent of recurrence of deformities after surgery and natural history of cavovarus foot deformity in CMT has not been well defined.

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Citation:

Alzarga, A., Abdo Mar’éi, M., Ahmed, A., Mostafa Elaidy, S. Surgical Treatment of Foot Deformity in Charcot Marie Tooth Syndrome. ***Zagazig University Medical Journal*****, 2024; (2941-2954): -. doi: 10.21608/zumj.2023.241857.2949**