

Original Article

Efficacy of triple surgery for cavovarus foot in Charcot-Marie-Tooth disease

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Abstract

Objective: to evaluate the efficacy of surgical treatment to correct the main deformities associated with flexible cavovarus foot due to Charcot-Marie-Tooth disease.

Methods: Fifteen patients (18 feet) with flexible cavovarus feet due to Charcot-Marie-Tooth disease were evaluated and underwent surgery between 2000 and 2015. We used a “triple surgery” protocol: a combination of plantar fasciotomy, valgus-inducing osteotomy of the calcaneus, and lengthening osteotomy of the first metatarsal. After a mean follow-up time of 105 (48 to 198) months, we developed a numerical scale to assess the main aspects of patient complaints: pain (3 points), function (3 points) and deformity (4 points). The scale considered the results of the visual analog pain scale, the American Orthopedic Foot and Ankle Society Scale, and the Coleman block test, as well as clinical and radiographic evaluation of gait and cavovarus deformities.

Results: According to the numerical scale, the results were considered satisfactory in 15 of the 18 feet (84%) and unsatisfactory in 3 (16%).

Conclusion: In the medium term, the “triple surgery” protocol proved efficient for correcting cavovarus deformities, providing functional improvement while preserving mobility without pain complaints. In the final analysis, through the “triple surgery” protocol, early indication for arthrodesis can be avoided, postponing sacrifice of the hindfoot joints.

Level of Evidence IV; Therapeutic Studies; Case Series.

Keywords: Charcot-Marie-Tooth disease/surgery; Foot deformities; Osteotomy/methods; Fasciotomy; Bone Lengthening/methods; Treatment outcome.

Introduction

Cavovarus foot is often caused by neurological disorders and is triggered by an imbalance between antagonistic muscle groups⁽¹⁾. Among the neurological diseases related to cavovarus foot deformities, Charcot-Marie-Tooth disease (CMT) stands out since it is associated with approximately 80% of the reported cases⁽²⁾. A common feature of CMT is progressive motor paralysis, which results in worsening deformities and joint stiffness in the foot and ankle⁽³⁾. Initially mild and flexible deformities in childhood tend to become accentuated and rigid in adulthood⁽⁴⁻⁶⁾.

In CMT, peripheral nerve involvement progresses from distal to proximal⁽⁷⁾. The intrinsic foot muscles are the first to suffer paralysis, followed by the tibialis anterior and fibularis brevis

muscles⁽⁷⁾. On the other hand, the strength of tibialis posterior and fibularis longus muscles is affected later, causing their actions to occur without due opposition, which triggers typical cavovarus foot deformities⁽⁸⁾.

The greatest treatment dilemma is choosing the most appropriate surgical intervention and the right time for it. Nevertheless, the surgery's corrective effect is not permanent due to the disease's progressive character, ie, as it evolves, other muscles may be affected⁽⁹⁾.

Since some degree of flexibility remains in the joints in the initial stage of the deformities, surgeries are aimed at both correcting the deformities and preserving joint mobility. The main procedures indicated at this stage are: releasing the plantar fascia (popularized by Steindler)⁽¹⁰⁾; hindfoot realignment by calcaneal osteotomy (described by Dwyer)⁽¹¹⁾;

Study performed at the Santa Casa de Misericórdia de São Paulo, São Paulo, SP, Brazil.

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and lowering the medial longitudinal arch with a dorsal closing wedge osteotomy at the first metatarsal head. When correcting deformities, it is also necessary to consider adding procedures that rebalance the deforming forces⁽¹²⁾. Given that no single surgery can correct all cavovarus foot deformities, an association of procedures is frequently needed⁽⁷⁾. Since the literature is not specific about the best combination of surgeries for correcting flexible cavovarus foot associated with CMT disease, we developed a specific treatment protocol that combines the following three procedures: 1) plantar fasciotomy; 2) valgus-inducing osteotomy of the calcaneus; and 3) lengthening osteotomy of the first metatarsal.

The aim of this study was to evaluate our protocol's effectiveness for treating flexible cavovarus feet associated with CMT disease.

Methods

This retrospective observational study was approved by the Institutional Review Board and registered on the Plataforma Brasil database under CAAE (Ethics Evaluation Submission Certificate) number: 03686718.1.0000.5479.

In this series, the diagnosis of CMT disease was essentially clinical, based on a family history of progressive foot deformity, atrophy of the intrinsic musculature of the hands and feet, peripheral vibratory sense deficit, and decreased muscle strength of the tibialis anterior and fibularis brevis muscles. Foot flexibility was clinically determined by the Coleman block test⁽¹³⁾.

Between 2000 and 2015, 18 patients (21 feet) with flexible cavovarus foot and CMT disease underwent "triple surgery" (TS), a name our service coined for a combination of: 1) plantar fasciotomy; 2) valgus-inducing osteotomy of the calcaneus; and 3) lengthening osteotomy at the head of the first metatarsal. In 12 feet, additional soft-tissue procedures were associated with TS according to individual needs. For study inclusion, a minimum follow-up of 48 months was required.

The 15 included patients returned for clinical-functional and radiographic evaluation, three of whom underwent TS in both feet, totaling 18 feet. There were eight female and seven male patients, whose mean age at the time of surgery was 22 years (9 to 60 years). The mean postoperative follow-up time was 105 months (48 to 198 months).

We considered three criteria when assessing the TS results: pain, function and residual cavovarus deformity. To objectively compare the results, we developed a numerical scale with maximum score of 10 points. The pain and function criteria were assigned a maximum score of three points each and residual deformity was assigned a maximum score of four points. The scores were classified as follows: ≥ 8 points was considered good, 5-7 normal, and ≤ 4 poor. Good and normal results were considered satisfactory, while poor results were considered unsatisfactory.

We used two scales to assess pain: a visual-analog pain scale (range 0 to 10 points), and a subscale from the American Orthopedic Foot and Ankle Society (AOFAS) rating system⁽¹⁴⁾

(range 0 to 40). In our numerical scale, visual-analog pain scores from 0-2 were scored as 2 points, scores from 3-7 were scored as 1 point, and scores from 8-10 were scored as 0. For the AOFAS pain subscale, scores ≤ 20 and >20 were scored as 0 and 1, respectively.

We used three clinical parameters for functional assessment: 1) the AOFAS function subscale (range 0-50); 2) the presence or absence of claudication during gait; and 3) residual flexibility in the foot joints. In our numerical scale, AOFAS subscale scores ≥ 36 (equivalent to 70% functionality) were scored as 1 and lower scores as 0. Regarding gait, the absence and presence of claudication were scored as 1 and 0, respectively. Regarding foot flexibility, positive and negative Coleman test results were scored as 1 and 0, respectively.

We used two parameters to evaluate residual cavovarus deformities, one clinical and one radiographic. Of the 4 total points in this area, 2 points each could be scored for varus and cavus deformity correction.

Regarding residual varus, the clinical evaluation (1 point) was performed with the patient standing, and the angle between the axes of the heel and leg was measured. Radiographic evaluation (1 point) was performed by measuring the angle between the axes of the calcaneus and tibia, using radiographs according to Saltzman's method⁽¹⁵⁾. In both evaluations, angular values of up to 4 degrees of varus were considered satisfactory and were scored as 1, while values ≥ 5 degrees of varus were considered unsatisfactory and were scored as 0 (Figure 1).

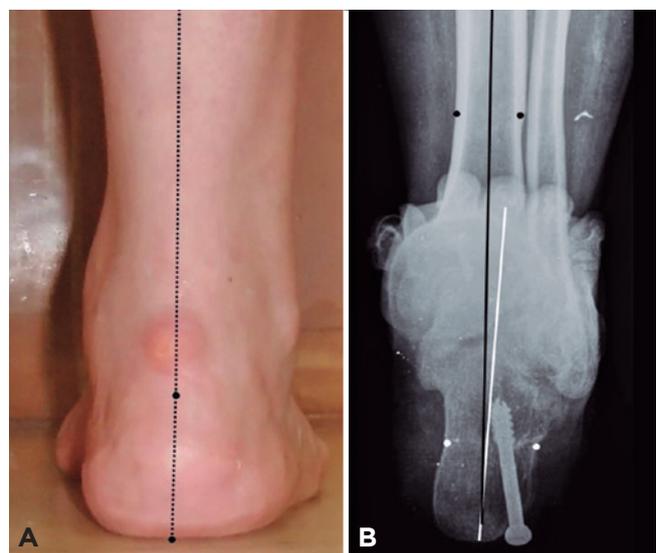


Figure 1. Posterior view of the right leg-foot (Saltzman) of patient 8, showing the clinical (1A) and radiographic (1B) methods for measuring the tibial and hindfoot axes, which are used to assess residual varus after surgical treatment.

For clinical measurement of cavus deformity (1 point), we used photographs of a medial weight-bearing view of the foot (Figure 2). We proposed a ratio with the height of the medial longitudinal arch (measured from the ground to the highest point of the curve in the midfoot) as the numerator (X) and the length of the foot (measured from the head of the first metatarsal to the heel where they contact the ground) as the denominator (Y). When the ratio was $\geq 50\%$ lower in the postoperative image than the preoperative image, the result was considered satisfactory and 1 point was awarded. When the reduction was $< 50\%$, the result was considered unsatisfactory and 0 points were awarded.

For radiographic measurement of the cavus deformity, we used only postoperative lateral weight-bearing radiographs of the foot (Figure 3). Considering the distance from the lower edge of the navicular bone to the ground as the numerator (A) and the length of the longitudinal axis of the navicular bone as the denominator (N), we applied the ratio $A/2N$ as

an evaluation method. When the height of the navicular bone \leq twice its length, the result was considered satisfactory and scored as 1. When the value was $>$ twice its length, the result was considered unsatisfactory and scored as 0.

Results

Table 1 shows the demographic data and the clinical-functional and radiographic results of TS for cavovarus foot associated with CMT disease.

According to the numerical scale, after a mean follow-up of 105 months (48 to 198 months), we obtained good results in 10 feet, normal results in 5 feet, and poor results in 3 feet. Thus, the final results were considered satisfactory in 15 of 18 feet (84%) (Figures 4 and 5).

Regarding residual pain, the mean visual-analog pain score was 1 (0 to 7) point and the mean AOFAS pain score was 37 (20 to 40) points. Regarding functional analysis, the mean AOFAS subscale score was 41 (20 to 50) points. Claudication was identified in 4 feet, and satisfactory flexibility was maintained in 17 feet after surgery.

Regarding whether TS could correct hindfoot varus, means of 2 (14° to -12°) and 3 (9° to -8°) degrees were found in the clinical and radiographic measurements, respectively. Regarding cavus deformity, a reduction $\geq 50\%$ in the height of the medial longitudinal arch occurred in only 2 feet according to the clinical measurement, while in radiographic measurement, the height of the medial arch was two times higher the length of the navicular bone in only 1 foot.

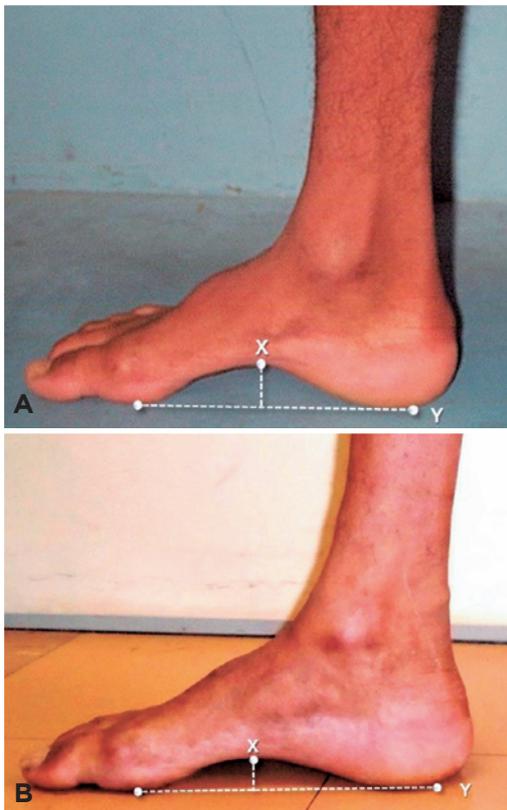


Figure 2. Pre- (2A) and postoperative (2B) medial view of the right foot of patient 5 in our series, showing the method used to assess the height of the medial longitudinal arch (X) in reference to the ground line (Y). Arch height variation was compared using a ratio of the lengths of X and Y.



Figure 3. Postoperative lateral view of the left foot (weight-bearing) of patient 10, showing the lines used in our radiographic method for evaluating the height of the medial arch. Line N represents the largest longitudinal axis of the navicular bone, while the line A measures the height of the navicular bone in relation to the ground. In this study, we used the ratio of N to A to calculate the magnitude of the residual cavus deformity after surgery.

In a separate analysis of the 3 (all unilateral) cases with poor results, the mean age was 42 (37 to 46) years at TS. The mean time between the initial and rescue surgery (triple arthrodesis) was 39 (16 to 74) months. Of the five patients (five feet) with normal results, the mean age at TS was 21 (9 to 60) years and the mean follow-up time was 122 (48 to 198) months. In the 7 patients (10 feet) with good results, the mean age at TS was 17 (10 to 28) years and the mean follow-up was 116 (48 to 192) months.

Discussion

CMT is a progressive neurological disease that causes muscle paralysis and affects the motor function of the foot^(16,17). Surgical interventions cannot permanently correct the associated deformities. Instead, soft tissue surgeries associated with osteotomies⁽¹⁷⁻²¹⁾ attempt to preserve joint mobility and postpone larger procedures, such as modeling arthrodesis⁽²²⁾.

The fundamental concepts of surgical treatment include correcting deformities, eliminating deforming forces, and

Table 1. Demographic data and clinical-functional and radiographic results for 15 patients (18 feet) who underwent triple-surgery treatment for cavovarus foot associated with Charcot-Marie-Tooth disease

Case	Sex, age, side	Additional surgeries/rescue surgeries (Y/N)	Follow-up (months)	Pain vaps (0-10) AOFAS (0-40)	Function AOFAS (0-50) coleman (Y/N) claudication (Y/N)	Varus deformity clinical/radiographic angle measurement	Cavus deformity clinical/radiographic qualitative measurements (satisfactory/unsatisfactory)	Decimal scale score (0-10) good/normal/poor
1	F, 58, L	Jones + FHLT/Y	27	7/20	20/N/Y	7°/4°	Unsatisfactory/satisfactory	3/poor
2	F, 33, R	Jones + TFL/N	155	3/30	40/Y/N	8°/3°	Unsatisfactory/satisfactory	7/normal
3	F, 26, L	LCT + TPTT + FHLT/N	198	0/40	27/Y/Y	14°/4°	Unsatisfactory/satisfactory	6/normal
4	F, 53, R	FHLT/Y	86	7/20	20/N/Y	14°/3°	Unsatisfactory/satisfactory	3/poor
5	M, 36, R	N/N	176	0/40	41/Y/N	-12°/3°	Unsatisfactory/satisfactory	9/good
	L	N/N	100	0/40	41/Y/N	-2°/2°	Unsatisfactory/satisfactory	9/good
6	M, 28, R	N/N	192	0/40	36/Y/N	5°/5°	Unsatisfactory/satisfactory	8/good
	L	N/N	175	0/40	36/Y/N	-9°/5°	Unsatisfactory/satisfactory	8/good
7	F, 23, L	Jones + FHLT/N	162	0/40	29/N/Y	-5°/8°	Unsatisfactory/satisfactory	6/normal
8	F, 30, R	Jones + TPTT/N	146	0/40	44/Y/Y	0°/4°	Unsatisfactory/satisfactory	8/good
9	F, 53, R	Jones + FHLT + LALR/Y	18	8/20	20/N/Y	6°/4°	Unsatisfactory/satisfactory	2/poor
10	M, 20, L	TFL/N	110	0/40	50/Y/N	-8°/-8°	Unsatisfactory/satisfactory	9/good
11	M, 26, R	TFL + LCT + LALR/N	90	0/40	50/Y/N	-8°/4°	Satisfactory/satisfactory	10/good
	L	TFL + LCT + LALR/N	76	0/40	50/Y/N	-5°/5°	Unsatisfactory/satisfactory	9/good
12	M, 14, L	TPTT/N	48	0/40	50/Y/N	-4°/1°	Satisfactory/satisfactory	9/good
13	M, 64, L	TFL + LALR/N	48	2/30	48/Y/N	4°/7°	Unsatisfactory/unsatisfactory	7/normal
14	F, 18, L	N/N	48	3/30	50/Y/N	2°/9°	Unsatisfactory/satisfactory	7/normal
15	M, 18, R	N/N	48	2/40	50/Y/N	3°/2°	Unsatisfactory/satisfactory	9/good

M: male; F: female; Y: yes; N: no; AOFAS: American Orthopedic Foot and Ankle Society Scale; COLEMAN: Coleman block test; FHLT: flexor hallucis longus tendon transfer to the fibularis brevis; Jones: extensor hallucis longus tendon transfer from the hallux to the first metatarsal bone; LALR: lateral ankle ligament reconstruction; LCT: lengthening of the calcaneal tendon; TFL: tenodesis of the fibularis longus to the fibularis brevis; TPTT: tibialis posterior tendon transfer to the dorsum of the foot; VAPS: visual analog pain scale.

attempting to restore the force of paralyzed muscles by performing muscle transfers. Occasionally, it is necessary to reconstruct incompetent lateral ligaments that are associated with muscle imbalance and gait instability⁽²³⁻²⁶⁾.

Although several surgical techniques have been described for cavovarus foot due to CMT disease, it is necessary to combine procedures to address multiple deformities⁽¹⁶⁻²³⁾. The literature does not clearly define how to select the best combination of procedures⁽²⁸⁻³⁰⁾. In this study, we attempted to develop a systematized surgical protocol for correcting the main flexible deformities of the cavovarus foot.

In our series of 15 patients (18 feet), the 3 feet with poor results were operated on later in the course of the disease (mean age at intervention 42 years vs. 22 years overall). It would seem that, due to the progressive nature of the disease,

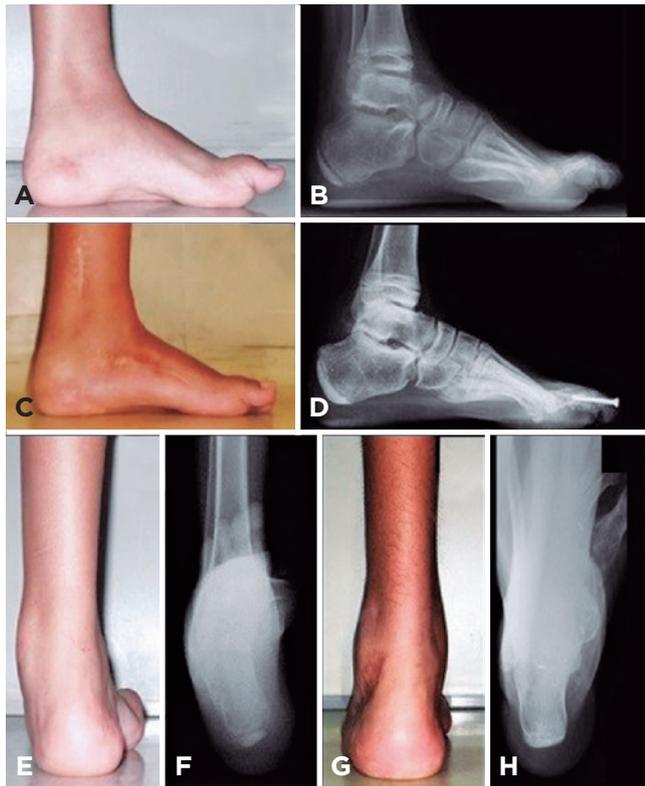


Figure 4. According to these clinical and radiographic images of the left foot and ankle of patient 12, the main deformities in the preoperative period were significantly corrected after surgery. Note the decrease in medial arch height before (4A) and 48 months after surgery (4C), which can also be seen in pre- (4B) and postoperative (4D) lateral weight-bearing radiographic images. Also note the correction of varus deformity of the hindfoot by comparing the pre- (4E) and postoperative (4G) clinical images. The correction can be better appreciated by comparing pre- (4F) and postoperative (4G) radiographic images in the axial leg-foot view. (Source: SAME Santa Casa de São Paulo).

there has been a neurological deterioration over the years, leading to greater muscle imbalance and more pronounced deformities. Thus, the point at which surgery is performed would directly influence the outcome. Better clinical-functional results would be expected of younger patients, whose deformities are less severe and rigid and whose degree of muscle impairment is lower.

Given that all patients with flexible deformities underwent the three combined procedures in our service, we cannot separately assess the corrections achieved by each procedure. Some authors consider calcaneus osteotomy unnecessary to correct deformities of the flexible cavovarus foot. They perform only lengthening osteotomy of the first metatarsal associated with plantar fasciotomy, reporting that they can correct the foot and prevent arthrodesis in most cases^(5,6,29,30). We believe that due to the varus component in the deformity, calcaneus osteotomy is important for complete correction in the hindfoot. It could be that feet which respond well to lengthening osteotomy of the first metatarsal associated

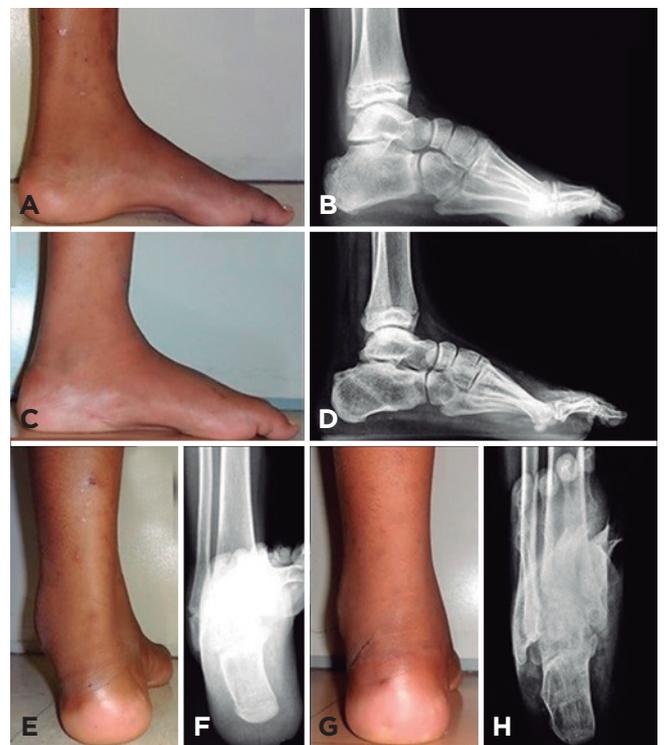


Figure 5. Clinical and radiographic images of the left foot of patient 14 pre- and 48 months postoperatively. The decrease in medial arch height can be seen by comparing the pre- (5A) and postoperative (5C) images. In the weight-bearing lateral view, the reduction in medial arch height between the pre- (5B) and postoperative (5D) periods is evident. Significant correction of the hindfoot varus deformity can be seen by comparing the preoperative clinical (5E) and radiographic (5F) images with those of postoperative period (5G and 5H). (Source: SAME Santa Casa de São Paulo).

with plantar fasciotomy are still in a very early stage deformity and have no major changes in the hindfoot, unlike some of the feet in our series. In addition, our clinical evaluation showed that there was no significant correction of the cavus deformity despite the lengthening osteotomy of the first metatarsal.

Due to the lack of a classification system to quantify deformities in the cavovarus foot, there are currently no formal indications regarding the ideal procedure for each type of deformity. There is not even a standardized system for evaluating the results of surgical treatment. Thus, we attempted to formulate a method for evaluating the main points we seek to improve with the surgical intervention. We developed a scale to more accurately evaluate the results and created objective parameters that allow a comparison of the initial and the post-surgery states for the main cavovarus foot deformities. This scale could also be used to assess different treatment modalities, allowing a comparison of the results of different techniques for correcting cavovarus foot.

Regarding study limitations, we point out that our preoperative documentation did not allow a complete comparison

between pre- and postoperative deformities in some cases. Thus, we could not accurately determine whether the cases that obtained satisfactory results were initially less severe than those that did not. Another important point is that the evaluation did not include soft tissue procedures performed in conjunction with TS.

Although we obtained satisfactory results with TS, it should be pointed out that these patients have a progressive neurological disease that can result in significant disability in foot function. The purpose of this protocol is not definitive treatment but rather to improve the symptoms and quality of life of these patients, reestablishing better foot support and possibly improved walking⁽³⁾.

Conclusion

As a proposal for cavovarus foot treatment in patients with CMT disease, our TS protocol efficiently corrected the hindfoot varus deformity in the medium term. The method can provide functional improvement while preserving mobility without pain complaints.

Authors' contributions: Each author contributed individually and significantly to the development of this article: LM (<https://orcid.org/0000-0001-5967-0541>)* data collection, clinical examination, wrote the article, interpreted the results of the study, participated in the review process; NMN (<https://orcid.org/0000-0001-7696-2220>)* wrote the article and interpreted the results of the study; RCF (<https://orcid.org/0000-0002-9886-5082>)* conceived and planned the activities that led to the study, wrote the article, participated in the review process. All authors read and approved the final manuscript. *ORCID (Open Researcher and Contributor ID) 

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