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PREVALENCE OF CLINICAL CHARACTERISTIC AND DIAGNOSIS OF CONGENITAL PSEUDOARTHROSIS OF THE TIBIA: ILIZAROV SURGERY

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ABSTRACT

Congenital pseudarthrosis of the tibia (CPT) which is characterized by persistent nonunion because of fractures of the tibia and the presence of fibrous tissue between parts of the fractured bone. The study aimed to Prevalence of clinical characteristic and diagnosis of congenital pseudoarthrosis of the tibia: Ilizarov surgery. This retrospective study included patients with congenital pseudarthrosis of the tibia (CPT) treated by segmental resection and the Ilizarov technique with a minimum follow-up period of 2 years. Surgeries were performed between 20013 and 2018 by a single orthopedic surgeon. In this study showed 17 patients: 12 (70.5%) males and five (29.4%) females. The mean age at the index surgery was 6.4 ± 3.8 (range, 3–10) years. The right leg was affected in ten (58.8%) patients, while the left was affected in seven (41.1%) patients. Ten patients had not been operated on for CPT before, while seven patients had one to five previous failed surgeries. Time from the first surgery to our interference was 3.8 ± 3.0 (range, 1–2.5) years. Seven patients had neurofibromatosis. Ilizarov surgery has a high association rate and can simultaneous synchronous powerful administration of issues with pseudarthrosis, including non-association, distortion, appendage shortening, and contiguous joint contracture and subluxation.

Key words: Congenital Pseudarthrosis, Limb Length Discrepancies, Intramedullary Rod.

INTRODUCTION

Congenital pseudarthrosis of the tibia (CPT) which is characterized by persistent nonunion because of fractures of the tibia and the presence of fibrous tissue between parts of the fractured bone. It is characterized as a "misleading joint" and is a break in the bone that neglects to mend all alone. The pseudarthrosis usually develops within the first two years of life; the diagnosis may be missed until pathologic fractures occur with weight bearing or on first attempts to walk in toddlers. Close to one-half of the breaks happen before the 2 years old. The tibia shows anterolateral bowing too as segmental dysplasia, which can bring about tibial nonunion and LIMB length shortening.

According to studies, ~ 50–90% of cases of CPT develop from neurofibromatosis type 1. Pseudoarthrosis results from abnormal tumor growth from neurofibromatosis and impaired bone healing due to dysplasia. This eventually leads to abnormal bone growth and bowing of the tibia 2.

The essential objectives of the different careful strategies right now accessible for CPT are to acquire the drawn out hard association of the tibia, forestall or diminish limb length discrepancies (LLD), forestall mechanical pivot deviation of the tibia and close-by joints, and prevent refracture of the bone.

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According to Children's Tumor Foundation, the ideal treatment for CPT associated with NF1 would encompass bone fixation, debridement, vascular bed implantation and promotion of osteogenic growth, control of bone resorption, prevention of pseudarthrosis tissue growth, and achievement of mature healthy bone³. The most common fixation treatments include using an intramedullary rod (IR)/screw fixation or Ilizarov/external fixation techniques for mechanical stability⁴. Both of these techniques utilize bone grafts for correcting LLD after excision of the pseudarthrosis site⁵.

The Ilizarov strategy was promoted as a suitable treatment choice as it can address pseudarthrosis, LLD, and the related complex staggered and multidirectional distortions. Furthermore, it can be used in cases where other methods have failed⁶. The generally announced association rate for the Ilizarov strategy goes from 60 to 100 percent⁷. The primary standards of the Ilizarov treatment incorporate careful resection of the pseudarthrosis, adjustment of precise distortions and joint direction, stable ossification for recuperating, and length gain by interruption osteogenesis. In the current study, we aimed to Prevalence of clinical characteristic and diagnosis of congenital pseudoarthrosis of the tibia: role of the Ilizarov treatment.

MATERIAL AND METHOD

This retrospective study included patients with congenital pseudarthrosis of the tibia (CPT) treated by segmental resection and the Ilizarov technique with a minimum follow-up period of 2 years. Surgeries were performed between 20013 and 2018 by a single orthopedic surgeon (A.I.Z.) who is highly experienced in pediatric deformity correction. The present study was conducted at Sree Balaji Medical college and Hospital, Chennai and Sri Lakshmi Narayana Institute of Medical Sciences, Pondicherry, India. Institutional review board (IRB) approval and consent from the parents of patients were obtained prior to conducting the study.

Inclusion criteria:

Patients with segmental tibial dysplasia with or without previous surgeries, aged 3 years or older, without ischemia or serious neurological deficits
With or without associated deformities.

Excluded Criteria:

Under 3 years of age, those with impaired foot circulation, and those with serious posterior tibial nerve damage.

Preoperative assessment

- The affected limbs were examined for deformities, skin condition, neurovascular status, the site of the lesion, ranges of motion of the hip, knee, ankle, and foot joints, and motion at the site of pseudarthrosis.

- All patients were examined for leg length discrepancy and stigmata of neurofibromatosis.
- The type of CPT was categorized according to the El-Rosasy–Paley classification for CPT⁸.
- Patients were categorized into primary cases presented to us without prior surgeries or revision cases who had previous surgeries elsewhere.

SURGICAL TECHNIQUES

Under general anesthesia and fluoroscopic guidance, surgeries were performed on a radiolucent table in a supine position. A tourniquet was used in all cases. The pseudarthrosis site was approached anteriorly by incising the skin, subcutaneous tissue, and deep fascia opposite the diseased part of the tibia. The pseudarthrosis was then excised using a saw till healthy bone edges were reached, and the medulla was gradually opened by applying drill bits of increasing size and reamers (4–11mm) that were suitable for the tibial size until a normal medullary canal was obtained. Great caution was used to not excise too much normal bone and to preserve the maximum available healthy bone in these patients with an already small-sized tibia. Removal of the entire surrounding periosteum was done. At this stage, the tourniquet was deflated and removed to accomplish hemostasis. A retrograde intramedullary rod was inserted in nine cases with large residual bone defects of more than 4 cm after intraoperative excision of the CPT and applying some shortening. The components of the applied Ilizarov frame varied according to the age of the patient and the length of the remaining available bone. In most cases, there were three levels of fixation, including the proximal tibia, the transported middle segment, and the distal tibial metaphysis. In some cases, an additional level of fixation was applied in the distal femur to correct or prevent joint contracture and subluxation, and another level was added below the ankle joint in cases with a very short remaining distal tibia. In only one patient, two tibial rings were applied proximal to the corticotomy site, one middle ring was applied in the transported segment, and two distal tibial rings were used.

Intraoperative acute leg shortening was done in some cases with smaller intraoperative bone defects to a safe distance of 3–4 cm, guided only by intraoperative monitoring of distal pulse and capillary circulation. This intentional shortening allowed primary bone grafting in two cases at our first surgical interference, while in 10 cases, a secondary bone graft was done at a later stage once the main tibial segments met at the docking site.

Primary iliac bone grafting at the docking site was done in two cases. Proximal tibial corticotomy was then done, with great care taken to resuture the periosteum after corticotomy to get the best chance of a good regenerate. Closure of the wound was then done in layers with or without a suction drain. In cases with retained hardware, it

was removed first, and then the completion of surgery was resumed as described.

In one patient with previous multiple surgeries and atrophic bone ends and non-union, there were genu recurvatum and posterior subluxation of the knee. Therefore, the knee subluxation was first corrected by anterior translation of the tibia using a translation mechanism within a few days following surgery, regardless of the initiation of distraction at the corticotomy site 1 week postoperatively. A trial of the gradual correction of the genu recurvatum through the site of the regenerate was executed in the distraction phase and before full consolidation of the regenerate.

In two patients with sclerotic-type CPT, the fibula was hypertrophied and deformed, so segmental excision was done to correct leg alignment and facilitate leg lengthening. In the single patient with a normal fibula, we kept it intact and only did tibial segment transport. In patients with an atrophic but intact fibula, segmental excision was done, while in patients with an atrophic fibula with pseudarthrosis, there was no need for any special maneuver. Great care to increase the diameter at the tibial docking site was taken, either by bone graft in our first surgery or weeks later, when the main tibial segments met.

Bone graft was indicated if the cross-sectional diameter at the docking site was narrow, and it was also used to fill small residual defects after the main tibial segments met at the docking site. In some patients who still had a small narrow tibia despite adequate debridement, multiple longitudinal osteotomies were done at the end of the proximal main tibia at the docking site, with the cross diameter of this tibial end enlarged by the gentle spreading of these longitudinal bone chips across the longitudinal osteotomies while they were still enclosed in their surrounding healthy periosteum

Postoperative care

On the first postoperative day, anteroposterior and lateral radiographs from the knee to the ankle joints were taken. Meticulous care was taken of the fixator pin sites. The latent period was between 5 and 7 days postoperatively. Then we always started with a 1 mm per day rate, with a rhythm of 0.25 mm every 6 h. This rate and rhythm continued for 3–4 weeks and was sometimes modified later to a lesser extent according to the shape and quality of the regenerate.

Postoperative lengthening was done in two cases in which we achieved complete closure of the docking site and did a primary bone graft. In the remaining 14 cases, middle segment transport was done until it reached the docking site, and secondary bone grafting was done in 10 cases. Then we continued the lengthening to the targeted length. X-rays were obtained every 2 weeks for 2 months,

and then every month till the removal of the fixator. Weight-bearing was allowed when tolerated, and the frame was removed after full solid union and the maturation of at least three cortices of the regenerate.

After removal, transcalcaneal wire was applied for an additional 1 or 2 months in four cases. In another five cases, a single intramedullary rod replaced the fixator to guard against refracture, and was exchanged periodically one to three times at intervals of 6–10 months in three cases according to tibial growth. The duration of use of the IM rod after frame removal in these cases ranged from 6 to 24 months. After frame removal, a below-knee cast was applied for 1–2 months, which was then replaced by a leg–ankle–foot orthosis till the end of follow-up. During the followup period, union, refracture, progression of deformities after full union, and our interference were all reported.

Methods of evaluation

The American Orthopaedic Foot and Ankle Society (AOFAS) scale^{9,10} was used for functional assessment. Radiographic and clinical results were classified into excellent, good, fair, and poor using the criteria previously reported in the study by Inan et al.¹¹; see Table 1.

The Paley classification¹² for pin-site problems was used to evaluate and guide the treatment of pin tract issues. Valgus deformity at the ankle was graded according to the Malhotra classification¹³. The residual pain was described as mild (pain after sports activity), moderate (pain during walking), or severe (pain at rest).

The satisfaction of patients and parents was evaluated using a questionnaire of five items measuring function, appearance, duration of treatment, how likely the patient would be to agree to undergo the procedure again, and overall satisfaction, with scores ranging from + 2 (the best) to – 2 (the worst)¹⁴.

Statistical analysis

Data were analyzed using the IBM SPSS software package, version 20.0 (IBM Corp., Armonk, NY). Categorical data were represented as numbers and percentages. The chi-square test was applied to investigate the association between the categorical variables. Quantitative data were expressed as a range (minimum and maximum), mean, standard deviation, and median. The Wilcoxon signed-rank test for abnormally distributed quantitative variables was used to compare two periods. The Mann–Whitney test was used to compare two groups based on non-normally-distributed quantitative variables. The significance of the obtained results was judged at the 5% level. A *P*-value of < 0.05 was considered significant.

Table 1 Classification of clinical and radiological results according to the study by Inan et al.

Excellent	Unrestricted activity Contact sports No LLD No pain No restriction of ankle and subtalar motion
Good	Noncontact sports LLD 2.5–4 cm Shoe modification compensates for limping Mild pain Slight restriction of ankle and subtalar motion
Fair	No sports LLD 4–5 cm Moderate pain Restriction of ankle and subtalar motion
Poor	Crutches for walking LLD > 5 cm Severe restriction or no ankle and subtalar motion

RADIOLOGICAL

Excellent	Radiographic union of pseudoarthrosis No axial malalignment in sagittal and coronal planes No ankle valgus No deformity in calcaneus > 30 degrees No LLD
Good	Healed pseudoarthrosis Axial malalignment 1–7 degrees in coronal or sagittal plane Valgus 1–5 degrees Calcaneal pitch (30–60 degrees) LLD 2.5–4 cm
Fair	Axial malalignment > 7 degrees in coronal or sagittal plane Ankle valgus > 8 degrees Calcaneal pitch > 60 degrees LLD 4–5 cm
Poor	Persistent pseudoarthrosis

Table 2 Demographics and baseline characteristics.

Characteristics	
SEX	
Male	12(70.5%)
Female	5(29.4%)
Age at index surgery (years) Mean ± SD	
SITE	
Right	10(58.8%)
Left	7(41.1%)
Previous operation	
No	10(58.8%)
Yes	7(41)
Time from 1st surgery (years) Mean ± SD	3.8 ± 3.0
NEUROFIBROMATOSIS	
No	13(76.4%)
Yes	4(23.5%)
El-Rosasy–Paley classification	
Type I	8(47%)
Type II	7(41%)

Type III	2(11.7%)
Fibula status	
Dysplastic	15(88.2%)
HypertrophiedNorma	1 (5.8%)
Normal	1(5.8%)
Preoperative AOFAS score Mean ± SD	49.5 ± 8.6

RESULTS

In present study showed 17 patients: 12 (70.5%) males and five (29.4%) females. The mean age at the index surgery was 6.4 ± 3.8 (range, 3–10) years. The right leg was affected in ten (58.8%) patients, while the left was affected in seven (41.1%) patients. Ten patients had not been operated on for CPT before, while seven patients had one to five previous failed surgeries. Time from the first surgery to our interference was 3.8 ± 3.0 (range, 1–7.5) years. Seven patients had neurofibromatosis.

All patients had an uprooted crack with pseudoarthrosis with the exception of one female patient, who had inherent segmental tibial dysplasia that didn't advance into pseudoarthrosis until she had careful treatment by segmental resection and portion transport by the Ilizarov strategy till association was accomplished. Afterward, repeat and refracture occurred and the patient was overseen effectively till a strong association was accomplished.

The typical shortening of the impacted appendage was 3.6 ± 1.9 (territory, 1-9) cm. As per the El-Rosasy-Paley order of CPT, eight (47.0 %) cases were type I, seven (41.1%) cases were type II, and three (17.6%) cases were type III. The fibula was dysplastic in 14 (82.3%) patients, hypertrophied in one (5.8%) patients, and normal in two (11.7%) patient. Table: 1

The average preoperative gap at the CPT site was 2.3 ± 1.0 (range, 0.5–4) cm. After complete excision of the pseudoarthrosis, the mean gap was 7.1 ± 1.0 (range, 4.5–8.0) cm. The mean preoperative AOFAS score was 49.5 ± 8.6 (range, 38–69). Table :2

Complications

At the hour of fixator expulsion, the arrangement of the tibia in all cases was physical or possibly inside the acknowledged reach, yet with expanded development of the as of late joined tibia many years, slow malalignment was noticed. Valgus distortion at the lower leg was seen in 11 cases, with a scope of 5-15 degrees. In all instances of valgus distortion at the lower leg, fibular pseudoarthrosis was a successive affiliation. As per the Malhotra grouping, five cases were grade 0, four cases were grade I, three cases were grade II, and four cases were grade II.

Treatment of valgus distortion at the lower leg was by transitory average distal tibial hemiepiphyodesis in three cases with a deformation of in excess of 15 degrees and by perception in different cases. No preliminary of average wedge osteotomy was finished, as the patients and their families rejected any move that

included rebreaking their legs after they had proactively recuperated from an extremely lengthy history of pseudoarthrosis and a past failure to walk. Procurvatum distortion of the tibia of somewhere in the range of 7 and 20 degrees happened in four patients, and recurvatum of 10 degrees happened in one patient. Joined deformations existed in eight (half) cases.

Valgus distortion at the knee existed in four (30%) cases, going from 10-20 degrees, and was seen at 1-3 years' development. In two of those cases, impermanent average proximal tibial hemiepiphyodesis was finished, and the staples were taken out later. Afterward, varus disfigurement at the knee with discouragement of the average knee joint space in the coronal plane and a transformed slant of the upper tibial articular surface in the sagittal plane was accounted for in one case at 15 years of follow-up.

Pin parcel contaminations of various grades happened in all patients and answered treatment with oral anti-microbials and rehashed twisted dressing, besides in four (23.5%) patients, who had grade III pin diseases in two pins. These pins were taken out, their plots were debrided, and new wires were reapplied at different locales.

Tenacious non-association happened in two patient, and all preliminaries to accomplish association fizzled. In those patients, bone unite and intramedullary titanium bars were applied two times, yet non-association endured. Disease and resorption of the bone join muddled the two tasks; the contamination settled toward the finish of treatment, however the break didn't join together. A protective orthosis was applied while sitting tight for normal development of the tibia to give sufficient length of a distal bone portion to serious areas of strength for oblige equipment in a later medical procedure.

Unconstrained abundance of the tibia in regards to the length was accounted for in three patients at 5 and 6 years of follow-up. In two of them, this excess was 4 cm at 12 years old, yet this abundance was turned around to 1.5 cm shortening at complete conclusion of the development plates. In the subsequent case, this abundance was 3 cm at the last development before the development plate conclusion and didn't determine. Refracture happened in four patients; in one of them, it happened at a pin site opening, while in the other case it was at the repetitive pseudoarthrosis site. In the two cases, amendment medical procedure prevailed with regards to accomplishing association till late in the subsequent period.

DISCUSSION

Congenital pseudarthrosis of the tibia (CPT) is a rare condition in children and is known for non-union and refractures. Besides achieving union and correcting deformities and limb length discrepancies (LLD), the goal of treatment is also to ensure a functional limb with minimal interventions .

In present study, we treated 17 patients with CPT with pseudarthrosis excision by Ilizarov technique. Paley et al classification of CPT, eight patients had type I CPT, seven had type II, and three patients had type III. However, the case with segmental tibial dysplasia could not be classified before the fracture according to this classification as there was no atrophic or sclerotic bone end and no pseudarthrosis at this stage. After the initial treatment and refracture with pseudarthrosis established, it was classified as a type I CPT. We believe that there is no single comprehensive classification that can consider all the pathological, clinical, and prognostic aspects of CPT in all circumstances.

Agashe et al.¹⁵ treated 15 patients with CPT using a combination of the Ilizarov technique and intramedullary rodding, and the mean AOFAS score was 64 at a mean follow-up time of 4.5 years. which is correlated with the preoperative AOFAS score was low in most cases in our study, with impaired limb function and an inability to bear weight on these deformed limbs with pseudarthrosis, except in one patient who was able to walk on her leg before surgery, as she only had pain because of an early stress fracture in the diseased segment, which did not advance to pseudoarthrosis till our surgery. The mean AOFAS score improved significantly from 47.5 preoperatively to 78.9 at the latest follow-up in current study.

After the successful union of pseudarthrosis, residual long-term problems such as LLD, ankle valgus, diaphyseal malalignment, degenerative changes at the ankle joint, and calcaneus deformity were reported in patients with CPT similar studies in Seo SG, Lee DY et al proximal migration of the distal fibula accounts for progressive ankle valgus, whereas prolonged immobilization of the ankle during surgeries and later in orthosis leads to stiffness.

In current study, the clinical results were excellent in one patient, good in seven patients, fair in six patients, and poor in three patients, and the radiological results were excellent in one patient, good in eight patients, fair in seven patients, and poor in one out of the 17 treated

patients which is correlated with Inan et al.¹⁷ reported that out of 16 treated cases with CPT, the clinical results were good in 12 (75%) patients and fair in four (25%) patients, and the radiographic results were good in nine (56%) patients and fair in seven (44%) patients.

Nonetheless, we believe that this arrangement doesn't think about the extraordinary idea of the sickness of CPT and the incredible enduring of the patients and their families at the pre-association stage. In many occurrences, the patients were profoundly happy with simply accomplishing super durable association of the pseudarthrosis and recapturing the capacity to bear weight on these forbearing appendages, anything the leftover LLD and different distortions present (if any). Notwithstanding, in this reviewing framework, > 5 cm lingering shortening is viewed as a poor clinical outcome, equivalent to instances of tenacious non-association. We imagine that lingering shortening can be treated by appendage extending later while persevering CPT requests removal. In this way, we accept that simply accomplishing super durable association of the CPT ought to overhaul the outcome by one grade in any evaluation system, so that the results would be classified as fair at least.

CPT stays a genuine test, not just as far as accomplishing association and revising related disfigurements, yet additionally as far as expecting, foreseeing, and treating late-beginning postoperative lingering intricacies after the total association of CPT. To diminish non-association and refracture rates, we suggest keeping the cross sectional region at the docking site as wide as could really be expected and staying away from untimely expulsion of the fixator. To accomplish the best lower leg capability and reduction the frequency of firmness in the lower leg and rear foot, we prescribe transformation to intramedullary obsession after the total association of CPT to dispose of the orthosis and permit joint movement as soon as could be expected.

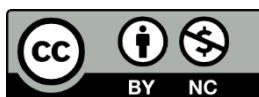
CONCLUSION

Inherent pseudarthrosis of the tibia can be enough overseen by the Ilizarov technique, which might be joined with an intramedullary pole and an essential or optional bone unite, with great long haul utilitarian and radiological results. This strategy has a high association rate and can furnish synchronous powerful administration of issues with pseudarthrosis, including non-association, distortion, appendage shortening, and contiguous joint contracture and subluxation.

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