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Review: Congenital Pseudarthrosis of the Tibia

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ABSTRACT

Congenital pseudarthrosis of the tibia (CPT) is one of the most challenging conditions in paediatric orthopaedics . It often requires multiple surgical procedures to achieve bone union and therefore demands a carefully planned, individualised treatment strategy. The ultimate goal is to ensure that patients can carry out their daily activities without any difficulties. Achieving this outcome requires prolonged follow-up and continuous patient education to monitor recovery and prevent complications, including postoperative infection, progression of deformity, refracture, and subsequent foot deformities. The optimal treatment strategy remains unclear. Conventionally, the defect created after resection of the pseudarthrotic segment is filled with a free bone graft, followed by internal fixation. Another option is the use of a vascularised free fibular graft harvested from the contralateral limb. More recently, advances in external fixation systems, including the Ilizarov method and Taylor Spatial Frame, have enabled simultaneous correction of deformities and correction of leg length discrepancy (LLD). Intentional tibiofibular cross-union has also been described. Some reports have recommended periosteal graft and biological adjuncts to promote bone union. Refracture or failure to achieve union is widely recognised as a common aspect of this condition's natural course. Reported union rates without refractures vary widely, ranging from approximately 50%-100%. The secondary changes include foot deformity, LLD, knee malalignment and even hip dysplasia due to valgus deformity of the proximal femur. These findings highlight the importance of individualised treatment plans, including multiple surgeries and post-operative management to prevent complications. In addition, long-term follow-up is necessary.

Keywords: Congenital disease, Pathological fracture, Pseudarthrosis, Tibial deformities

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INTRODUCTION

Congenital pseudarthrosis of the tibia (CPT) is one of the challenging diseases in paediatric orthopaedics. It is a rare disorder, affecting 1 in 140,000 to 190,000 live births. The association between CPT and neurofibromatosis has been recognised since 1937, and 5.7% of patients with neurofibromatosis type 1 were found to have the deformity (Ducroquet & Cottard, 1937). Up to 55% of cases of anterolateral bowing and pseudarthrosis are associated with neurofibromatosis (Andersen, 1976; Strong & Wong-Chung, 1991; Whitehouse, 1966).

CPT often requires repeated surgeries to achieve bone union. Various treatment methods and their long-term outcomes have been reported, but the standard treatment guidelines for CPT have not been established. A characteristic feature of CPT is that, even after achieving bone union, patients remain at high risk of refracture and/or recurrence of the deformity.

CAUSES OF CPT

The exact aetiology of CPT remains unclear. Neurofibromatosis type 1 (NF1) is considered one of the major conditions associated with CPT, whereas fibrous dysplasia and osteofibrous dysplasia are associated with only a few cases. NF1 is an autosomal dominant disorder associated with various cutaneous manifestations and skeletal abnormalities. The NF1 gene on chromosome 17 typically produces the protein neurofibromin, which controls cell proliferation. The protein is expressed within different types of cells, including osteoblasts, osteoclasts, and fibroblasts. Its dysfunction results in insufficient bone anabolism and excessive catabolism, which ultimately leads to weakened bone and suboptimal bone repair. Bones from NF1 mouse models (Elefteriou et al., 2006; Rhodes et al., 2013) and bones from individuals with NF1-associated CPT (Seitz et al., 2010) are characterised by an unmineralized and fibrous bone

matrix, despite having normal serum phosphate and calcium concentrations.

CLINICAL AND RADIOLOGICAL CLASSIFICATIONS

Radiographs of CPT often reveal changes along the middle and distal thirds of the tibia, with frequent involvement of the fibula. The three primary problems are anterolateral bowing, pseudarthrosis after the fracture of the tibia, and proximal migration of the fibula. Based on these characteristics, several classifications have been developed using radiographic findings. These include Crawford's (1986), Andersen's (1973), Boyd's (1982), Choi's and Paley's (2019) classifications (Table 1). The first three classifications are relatively traditional and are based mainly on morphological findings (dysplastic, cystic and sclerotic) on the radiographs. In particular, the preservation of the medullary canal and cortical thickness is also taken into consideration. The latter two classifications also evaluated the morphologic status of both the distal and proximal bone ends of the tibia, and the fibula. They are widely used to guide treatment selection. Boyd's classification can also be used for clinical prediction of CPT (Table 2).

Table 1: Radiological findings of congenital pseudarthrosis of the tibia based on different classifications

Classifications	Characteristics	Cortical density	Tibial morphology	Fibular morphology
Crawford (1986)				
Type 1	Preserved medullary canal	Normal	Anterior bowing	-
Type 2	Narrowed medullary canal	Sclerotic	Anterior bowing	-
Type 3	Recognisable medullary canal	Cystic	Anterior bowing	-
Type 4	Pseudarthrosis	Non-union	Anterior bowing	-
Andersen (1973)				
Dysplastic	Dysplastic hourglass	-	Anterior and lateral bowing	Lateral bowing
Club foot	Contralateral clubfoot	-	More anterior bowing	Anterior bowing
Sclerotic	Distal fibula and tibial fractures	-	Anterior and lateral bowing	Pseudarthrosis
Cystic	Cystic change in fibula & tibia	-	Little anterior bowing with fracture	Slight bowing
Boyd (1982)				
Type I	-	-	Anterior bowing	-
Type II	-	-	Anterior & hourglass	-
Type III	-	-	Pseudarthrosis	-
Type IV	-	-	Pseudarthrosis with fracture & non-union	-
Type V	-	-	-	Dysplastic fibula
Type VI	-	-	Intraosseous neurofibroma Schwannoma development	-
Paley (2019)				
Type 1	-	-	Absence of tibial fracture	Absence of fibular fracture
Type 2				
2A	-	-	Absence of tibial fracture	Presence of fibular fracture with no fibular migration

Classifications	Characteristics	Cortical density	Tibial morphology	Fibular morphology
2B	-	-		Presence of fibular fracture with proximal fibular migration
Type 3	-	-	Presence of tibial fracture	Absence of fibular fracture
Type 4				
4A	-	-		Presence of fibular fracture with no fibular migration
4B	-	-	Presence of tibial fracture	Presence of fibular fracture with proximal fibular migration
4C	-	-		Presence of fibular fracture with proximal fibular migration and tibial bone defect

Table 2: Clinical prediction of different types of Boyd's classification of Congenital Pseudarthrosis of the Tibia

Boyd's Classification (1982)	Clinical prediction
Type I	-
Type II	Recurrence of pseudarthrosis
Type III	Anterior bowing before or after fracture
Type IV	Stress fracture
Type V	Development of tibia or fibula pseudarthrosis
Type VI	Possible development of pseudarthrosis

TREATMENT STRATEGIES

Treatment often involves both surgical and non-surgical modalities. However, the optimal treatment method is still unclear. In most cases, surgical intervention is required. Nevertheless, the choice of treatment depends on several factors, including the type and location of the pseudarthrosis, age of the patient, and presence of associated conditions. Although used less frequently, magnetic resonance imaging (MRI) can also be a valuable imaging modality in the diagnosis of CPT (Teo et al., 2007). This can be especially useful in evaluating the soft tissues and helping to determine the amount of resection of the fibrous hamartoma.

Non-surgical treatment

Non-surgical management may be indicated primarily to prevent fracture and progression of deformity, particularly through braces before and after surgery. Non-operative treatment is unlikely to prevent the development of secondary changes such as atrophy, leg length discrepancy (LLD), fibular migration, ankle valgus and calcaneal deformity. Recently, studies investigating pharmacological treatment have primarily evaluated its use as an adjunct to surgical intervention, while its role as a standalone or standard treatment remains largely unexplored.

Pharmacological therapy

Pharmacotherapeutic adjunctive treatment has been employed as part of the treatment of CPT using bone morphogenetic protein (BMP)-2 or BMP-7, with or without bisphosphonate therapy (zoledronic acid, ZA) (Högler et al., 2004; Schindeler et al., 2008). Lee et al. (2006) reported on five CPTs treated with BMP-7 combined with allograft, intramedullary rodding and external fixation. The mean age was 6 years old, and the mean follow-up was 14 months. The use of recombinant human BMP-7 alone has been insufficient to overcome the poor clinical outcomes associated with CPT. Little et al. (2005) used bisphosphonate (ZA) for patients with CPT to control the activity of osteoclasts to promote a union. As bisphosphonate therapy was administered after bone graft harvesting, it might have provided adequate protection against resorption of the harvested bone graft.

Operative treatment

Reinforcing and preventing fracture

In children younger than 3 years old, the deformity of the tibia may not be evident in radiographs yet. It is important to maintain proper mechanical alignment. A brace application will be helpful if the dysplastic and pathological changes are not remarkable. Trans-articular intramedullary rodding may be considered in patients with these pathological changes, even before 3 years of age (Joseph et al., 2003). Intramedullary rod with free bone grafts is necessary to maintain proper alignment if these changes are significant. This helps to prevent recurrence of deformity and reduce the incidence of refracture.

Correction of Deformity and the Maintenance to Achieve Bone Healing

Two recent retrospective studies on CPT supported the use of intramedullary fixation. Dobbs et al reported their long-term results in 21 consecutive patients using the Williams rod technique with free bone grafting, achieving initial consolidation in 18 (86%) patients (Dobbs et al., 2004). In the second study describing the use of intramedullary fixation in CPT, Johnston described important variations in the technique that might lead to more consistent union rates of the pseudarthrosis (Johnston, 2002). Concomitant fibular osteotomies, with or without fixation that were performed at the same time as the tibial pseudarthrosis repair, yielded superior clinical and radiographic results when compared to cases in which the fibulae were left intact. These two studies included patients whose treatment was augmented as much as possible with free bone grafts.

Paley recently recommended a more comprehensive surgical strategy: 1) pre-surgical infusion of ZA; 2) hamartoma resection around tibia and fibula with resection of the interosseous membrane; 3) insertion of a telescopic growing rod into the tibia; 4) insertion of wire rod into the fibula; 5) large cancellous bone graft harvest from the ilium; 6) concurrent periosteal graft harvest from the ilium, followed by 7) Ilizarov fixation or alternatively by a locking plate (Table 3) (Paley, 2012). A combination approach including intramedullary rod, Ilizarov apparatus, corticocancellous bone graft, and periosteal graft was reported for the severe type (Crawford type 4) (Zargarbashi et al., 2021).

Various external fixation systems, such as the Ilizarov frame and specialised frames, have been widely used for both correction and maintenance of deformity correction (Choi et al., 2011; El-Gammal et al., 2023). These techniques are also used for bone transport to fill the bone defect created after resection of the pathological bone tissue, and to preserve the original limb length in severe cases of CPT (Grill et al., 2000; Vanderstappen et al., 2015).

The cross-union procedure between the tibia and fibula has been reported as an advanced technique. This procedure has produced excellent outcomes, with multiple authors reporting a 100% success rate at follow-up (Rastogi & Agarwal, 2022; Vaidya et al., 2019). However, questions regarding the differential growth rates of proximal versus distal fibula and the potential ankle valgus created by subsequent growth remained unresolved.

Recently, guided growth has been employed to restore normal mechanical alignment, with the aim of preventing progression of anterolateral tibial bowing (Kennedy et al., 2017; Todderud et al., 2024).

Table 3: Surgical approaches based on Paley's classification of Congenital Pseudarthrosis of the Tibia

Paley's Classification (2019)	Surgical approach
Type 1	Do not release syndesmosis during hamartoma excision from fibula. Cut overlapping fibula
Type 2	
2A	Do not release syndesmosis during hamartoma excision from fibula. Cut overlapping fibula
2B	Release syndesmosis during hamartoma excision from fibula. Move distal fibula and cut ends of fibula
Type 3	Same as Type 1, resect sclerotic & atrophic bone tissue for bone contact
Type 4	
4A	Same as Type 1, separate ends of tibia & fibula, and cut overlapping fibula ends
4B	Same as Type 2B, separate ends of tibia & fibula: osteotomised fibula or tibia to correct the deformity, and resect sclerotic & atrophic bone tissue for bone contact
4C	Resect sclerotic & atrophic bone tissue for bone contact. If necessary, cut overlapping ends of fibula

Free vascularised fibula graft (FVFG)

The free vascularised fibular graft (FVFG) procedure is indicated for significant bone defects or shortening, or as a salvage procedure (El-Gammal et al., 2021; Simonis et al., 1991). The optimal timing for surgical intervention is at or before 3 years of age, with

intramedullary fixation reported to provide the most favourable outcomes, including prevention of refracture (El-Gammal et al., 2021). One of the disadvantages of FVFG is that it cannot correct LLD. A recent study examined the mid-term outcomes of a combined vascularised fibular graft with Ilizarov fixation, originally termed telescoping vascularised fibular graft. It showed favourable results through a combination of a vascularised fibular graft and large bone segment allograft, avoiding the higher reintervention rate and discomfort with ring frame bone transport, and the prolonged non-weight bearing with vascularised fibula transfer without reinforcement with a massive, large bone segment allograft (Van Den Heuvel et al., 2020).

CLINICAL AND RADIOLOGICAL OUTCOMES

Reported primary union rates range from 61% to 100%, while refracture occurs in 21% to 68% of cases (Paley, 2019). The average reported rate of union without refracture is approximately 50% (Kesireddy et al., 2018; Shah et al., 2018). The European Paediatric Orthopaedic Society (EPOS) multicentre study of 340 patients with CPT reported a 75% healing rate achieved with Ilizarov fixator and recommended the use of prophylactic intramedullary rodding to prevent refracture (Grill et al., 2000).

Union occurred in 86% of the patients over an extended period, averaging 16 months (Teo et al., 2007), whereas lack of union or union followed by refracture occurred in 50 - 68% of patients (Kesireddy et al., 2018; Paley, 2019). Unsound union at skeletal maturity was found to occur in 26 tibiae (24%) (Shah et al., 2018). The primary union rate was achieved in 17 of 21 (81%) patients. Refracture occurred in four of the 17 patients (19%), and the minimum follow-up duration was two years (Boero et al., 1997). Most recently, Laufer et al. (2020) reported on 26 patients treated for CPT between 1997 to 2019. Six were treated by resection of pseudarthrosis and bone transport with grafting of the docking site. It resulted in a 50% union rate with no refractures.

Amputation may be considered either as a primary treatment option in selected cases or as a last resort (Guille et al., 1998; McCarthy, 1982). It may be considered in the following conditions: foot conditions, number of previous operations, severity of LLD and the presence of severe infections.

The united bone is often of inferior biological and mechanical quality, so lifetime protection with intramedullary devices, braces or a combination of both is recommended (Grill et al., 2000).

FUTURE PERSPECTIVE

When NF1-deficient mice were treated with asfotase-α, they had increased tibial bone mineral density, increased trabecular bone volume, and increased bone strength (White et al., 2007). These findings suggest that CPT and other skeletal manifestations of NF1 may potentially respond to enzymatic therapy; however, this approach has not yet been translated into clinical treatment of CPT.

DISCUSSION

The management of CPT is complex, with treatment goals focused on correcting deformity, achieving bone union at the pseudarthrosis site, and preventing proximal migration of the fibula to avoid ankle valgus. Many long-term follow-up data have highlighted the persistent challenges in achieving stable, long-term union without complications (Joseph et al., 2003; Paley, 2019; Teo et al., 2007). The available evidence indicates that, although union can be achieved, the average number of surgical procedures remains substantial, particularly in more severe cases (Paley type III or higher) where intramedullary nailing appeared to be essential (Dobbs et al., 2004; Joseph et al., 2003; Teo et al., 2007). This aligns with current recommendations for CPT treatment, which emphasised the critical role of intramedullary nailing in conjunction with external fixation for deformity correction and maintenance.

The optimal age for surgical intervention remains a subject of debate. Some studies suggested early intervention (before 3 years of age), while others advocated for later surgery (El-Gammal et al., 2021; Joseph et al., 2003). The presence of prior inappropriate surgeries was identified as a factor affecting outcomes. This highlights the importance of timely and appropriate initial management. As demonstrated in this review, long-term follow-up is crucial in CPT, as the incidence of refracture tends to increase with longer observation periods. The observed complications, such as ankle valgus and LLD, emphasised the need for continued monitoring and potential further interventions throughout the patient's skeletal growth and into adulthood. Current recommended treatment strategies also include extensive resection of hamartomatous bone and soft tissue at the pseudarthrosis site, bone and periosteal grafting, tibiofibular cross-union (4-in-1 technique), and consideration of locking plates, vascularised fibular grafts, and the use of BMP-2 and bisphosphonates (ZA).

CONCLUSION

The aim of treatment for CPT is to achieve stable bone union and full weight-bearing ambulation, which is often difficult to attain. CPT requires multiple surgical interventions and careful long-term management due to the high risk of complications. Intramedullary nailing is a key component of treatment strategies with highly individualised selection of surgical

procedures,integrating the latest scientific evidence with the surgeon's clinical experience to optimise patient outcomes. Treatment should be individualised, and long-term follow-up is essential..

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