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CONGENITAL PSEUDARTHROSIS OF THE TIBIA: ANALYSIS OF FACTORS INFLUENCING OUTCOMES

Orthopaedics

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ABSTRACT

Background: Congenital pseudarthrosis of the tibia (CPT) is a challenging pediatric condition characterized by difficulty in achieving union, high refracture rates, limb-length discrepancy, and progressive ankle deformity. The influence of various clinical and surgical factors on these outcomes remains controversial. **Objective:** To evaluate the variables influencing union, refracture, limb-length discrepancy, and ankle deformity in children with CPT treated with a uniform surgical pz1rotocol. **Methods:** A retrospective review was conducted of 35 children with CPT treated at a single center over a 20-year period using excision of pseudarthrosis, intramedullary rodding, and cortical bone grafting. The mean age at surgery was 2.0 ± 2.32 years, with a mean follow-up of 8.62 ± 5.45 years. Outcomes assessed included union (primary and secondary), refracture, limb-length discrepancy, and ankle valgus deformity. Radiographic union was defined by cortical continuity on anteroposterior and lateral views. Cox regression analysis was used to identify factors influencing outcomes. **Results:** Primary union was achieved in 65.7% of patients within a mean duration of 4.5 ± 2.5 months, while all but one patient achieved union at final follow-up (91.4% sound union). No studied variable, including age at surgery, neurofibromatosis, fibular status, or implant type, significantly influenced union or refracture rates. Refractures occurred in 25.7% of patients at a mean of 4 years postoperatively. Limb-length discrepancy averaged 4.67%, with significantly less shortening in children operated before 3 years of age (p=0.02), and increased shortening associated with multiple surgeries (p=0.01) and refracture (p=0.04). Ankle valgus deformity was observed in 40% of patients and was significantly associated with fibular pseudarthrosis (p=0.013) and refracture (p=0.005). **Conclusion:** Intramedullary rodding combined with cortical bone grafting provides high union rates in CPT. While no variables significantly influenced union or refracture, early surgical intervention, fewer subsequent procedures, and prevention of refracture were associated with improved limb-length outcomes. Fibular pseudarthrosis and refracture significantly contribute to ankle valgus deformity. Larger multicenter studies are required to further clarify prognostic factors in CPT.

KEYWORDS

Congenital Pseudarthrosis of Tibia, Intramedullary Rodding, Refracture, Limb-Length Discrepancy, Ankle Valgus, Pediatric Orthopaedics

INTRODUCTION

Congenital pseudarthrosis of the tibia (CPT) remains one of the most challenging conditions in pediatric orthopaedics, owing to the difficulty in achieving and maintaining union, the high incidence of refracture, limb-length discrepancy, and progressive deformities of the leg and ankle[1,4]. Despite numerous surgical techniques described in the literature, the influence of various clinical and surgical factors on these outcomes remains incompletely understood, with conflicting results reported across studies.

In particular, the role of variables such as age at surgery, presence of neurofibromatosis, fibular status, and surgical technique in determining union, refracture, limb shortening, and ankle deformity has not been clearly established.

The aim of the present study was to evaluate the effect of these variables on union, refracture, limb-length discrepancy, and ankle deformity in children with CPT treated with excision of pseudarthrosis, intramedullary rodding, and cortical bone grafting.

MATERIALS AND METHODS

The case records of 50 consecutive children with congenital pseudarthrosis of the tibia treated in the single center over the last 20 years were reviewed to identify cases that were treated by excision of pseudarthrosis, intramedullary rodding and cortical bone grafting. Thirty-seven children were treated by intramedullary rodding and cortical bone grafting. Two of these children lost follow-up. Remaining 35 children formed the study group which included 21 boys and 14 girls. Twenty-two children had features of neurofibromatosis. Associated fibular pseudarthrosis was present in 29 children whereas 9 children had intact fibula. The site of pseudarthrosis was lower third of tibia in 27 cases, middle third in 6 cases and middle third lower third junction of the tibia in 2 cases. Thirty-two patients were classified as Crawford type 4 whereas 3 patients were classified as Crawford type 3.

The mean age at presentation was 2.0+2.28 years, while the mean age at index surgery was 2.0+2.32 years. The mean follow up duration was 8.62+5.45 years.

All these children underwent excision of pseudarthrosis, intramedullary rodding of tibia and cortical bone grafting (Figure 1). No fixation of the fibular pseudarthrosis was attempted. Intramedullary rush rodding through calcaneum was performed in 31 children, rush rodding through knee in 2 and Sheffield telescopic rodding through ankle was done in 3 children. The intramedullary rod was retained till skeletal maturity to support the pseudarthrosis site. This entailed revision of the rod when the child outgrew the initial rod. Protective bracing was continued till skeletal maturity in the form of clamshell orthosis.

Radiographs of the children taken at six-weekly intervals were reviewed to determine the time to union. If the pseudarthrosis united following the index surgery, it was termed as primary union whereas if any form of additional surgery was needed to obtain union it was termed as secondary union.

Figure 1: Treatment Technique Showing Excision of Pseudarthrosis, Cortical Grafting, and Transcalcaneal Rodding.

Two independent investigators concurred that cortical continuity was clearly evident on both the antero-posterior and lateral view radiographs in order to minimize inter-observer bias. Union was further quantified as per table 1.

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Table1. Union Quantification as Per Cortical Continuity Restored in Antero-posterior and Lateral View Radiographs.

Cortical continuity	Union
Four cortices	Sound Union
Three cortices	Satisfactory Union
Two or less cortices	Unsound Union

The frequency, timing and nature of additional surgical intervention during the period of follow-up and the frequency, timing and treatment of re-fractures were noted from the case records. At last follow up, clinical evaluation was done to not deformities of the tibia, deformities of the ankle and hind foot, range of motion of the ankle joint and the morbidity at the bone graft donor site. The function of the ankle was assessed by applying the American Orthopaedic Foot and Ankle Society (AOFAS) Ankle-Hind foot Score. Radiographic evaluation from AP & lateral films was done to note status of union of the tibia, status of the fibula, angular deformities of the tibia in both planes and tilt of the ankle mortise. Limb lengths were measured from grid films.

Statistical analysis

The data was analyzed using Cox regression analysis to identify various variables influencing the union rate, re-fracture rate, limb-length discrepancy and ankle deformity.

RESULTS

Variables Influencing Union of Tibial Pseudarthrosis

Out of 35 children, 23(65.7%) achieved primary union of pseudarthrosis following index surgery within mean (SD) duration of 4.5(2.5) months. Secondary union was achieved in remaining 12 children following subsequent surgeries. At last follow up 32 children had sound union (table 2) (Figure 2).

Table 2. Union of Tibial Pseudarthrosis at Final Follow up.

Final Union	Number (n=35)	Percentage
Sound union	32	91.4%
Satisfactory union	2	5.7%
Unsound union	1	2.9%

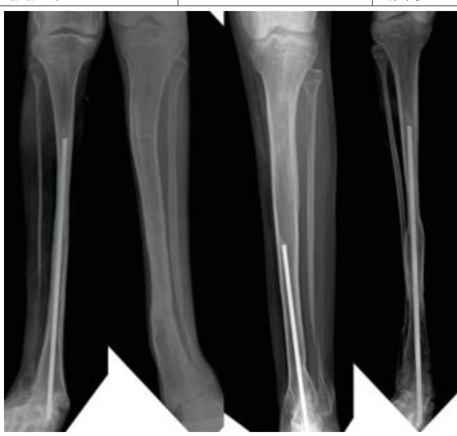

Figure 2: Examples of Sound Union of Tibial Pseudarthrosis

None of the variables including age at surgery (figure 3 and 4), presence of neurofibromatosis, associated fibular pseudarthrosis (figure 5 and 6), type of intramedullary rod or source of bone graft show any statistically significant influence on union of pseudarthrosis (table 3).

Table 3. Variables Influencing Union of Pseudarthrosis of the Tibia.

Variables	(n)	Union	Significance
Age at surgery	<3 years (23)	22	p=0.19
	>3 years (12)	12	(NS)
Neurofibromatosis	present (22)	21	p=0.89
	absent (13)	13	(NS)
Fibular pseudarthrosis	present (29)	28	P=0.76
	absent (6)	6	(NS)
Site of pseudarthrosis	middle third (6)	5	p=0.14
	lower third (27)	27	(NS)
	junction (2)	2	
Type of I.M. rod	rush rod (32)	31	p=0.94
	telescopic rod (3)	3	(NS)
Type of bone graft	autogenous (32)	31	p=0.13
	maternal (3)	3	(NS)

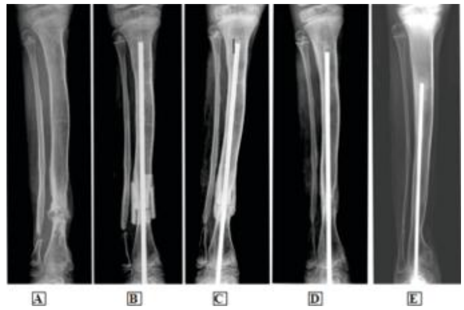

Figure 3: An example showing age at index surgery does not influence union of tibial pseudarthrosis

A. Seven years of age male child with CPT.
B. Three months following index surgery.
C. Six months following index surgery.
D. One year following index surgery.
E. Sound union at last follow up.

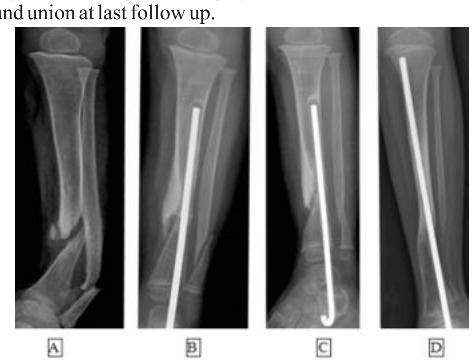

Figure 4: An example showing age at index surgery does not influence union of tibial pseudarthrosis.

A. Two years of age male child with CPT.
B. Six months following index surgery.
C. One year following index surgery.
D. Sound union at last follow up.

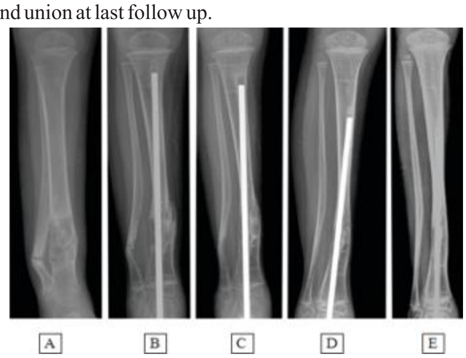

Figure 5: An example showing that associated fibular pseudarthrosis does not influence union of tibial pseudarthrosis.

A.Two years child with CPT & fibular pseudarthrosis
B. Unsound primary union 6 months - index surgery.
C. Six months following repeat bone grafting.
D. One year following repeat bone grafting.
E. Sound union at last follow up.

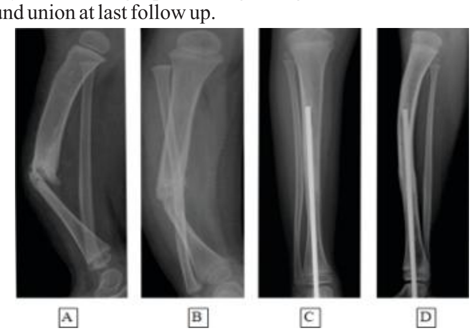

Figure 6: An example showing that associated intact fibula does not

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influence union of tibial pseudarthrosis.
A & B. Five years of age female child with CPT with intact fibula.
C&D. Sound union at last follow up.

Variables Influencing Refractures Following Union of Pseudarthrosis

Twelve (34.3%) re-fractures were encountered in 9 (25.7%) patients at a mean of 4 years after index surgery. Out of these 12 refractures, 9 had sound union, 2 had satisfactory union and one had unsound union at final follow up. None of the variables among age at index surgery, presence of neurofibromatosis, associated fibular pseudarthrosis or site of pseudarthrosis were found to significantly influence occurrence of refracture at pseudarthrosis site (table4).

Table 4. Variables Influencing Refracture at Pseudarthrosis of the Tibia.

Variables	(n)	Refracture	Significance
Age at surgery	<3 years (23)	5	p=0.84 (NS)
	>3 years (12)	4	
Neurofibromatosis	present (22)	5	p=0.79 (NS)
	absent (13)	4	
Fibular pseudarthrosis	present (29)	9	P=0.50 (NS)
	absent (6)	0	
Site of pseudarthrosis	middle third (6)	2	p=0.27 (NS)
	lower third (27)	6	
	junction (2)	1	

Variables Influencing Limb Length Discrepancy in Congenital Pseudarthrosis of the Tibia.

There was 11.47% mean tibial shortening, 1.22% mean femoral lengthening and 4.67% mean limb shortening. Those children who underwent index surgery prior to three years of age had statistically significant lesser limb shortening as compared to those who underwent index surgery after three years of age (figure 7 and 8). Children who required more than two surgeries to achieve sound union had significantly more limb shortening. Occurrence of refracture at pseudarthrosis site added to limb shortening (table5).

Table 5. Varibles Influencing Limb Shortening in Congenital Pseudarthrosis of the Tibia.

Variables	(n)	shortening		Significance
		<4.6%	>4.6%	
Age at surgery	<3 years (23)	15	8	p=0.02 (S)
	>3 years (12)	6	6	
Subsequent Surgeries	<2 surgeries (29)	19	10	p=0.01 (S)
	>2surgeries (6)	2	4	
Refracture	present (9)	5	4	P=0.04 (S)
	absent (26)	16	10	
Fibular pseudarthrosis	present (29)	15	14	p=0.83 (NS)
	absent (6)	6	0	

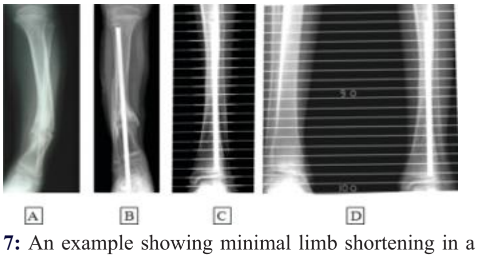

Figure 7: An example showing minimal limb shortening in a child who underwent index surgery before 3 years of age.
A. Three years of age female child with CPT.
B. Six months following index surgery.
C. Eight years following index surgery with sound primary union.
D. Minimal limb length inequality at last follow up.

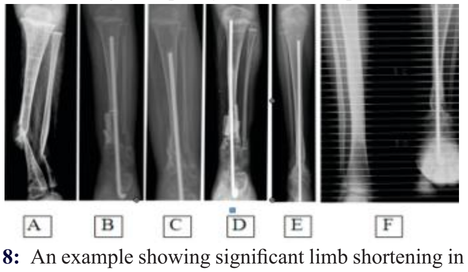

Figure 8: An example showing significant limb shortening in a child who underwent index surgery after 3 years of age and more than 2 surgeries to achieve union.
A. Five years of age male child with CPT.
B. Following index surgery.
C& D. Second and third bone grafting.
E. Sound union at last follow up.
F. Significant limb length inequality at last follow up.

Variables Influencing Ankle Deformity

Clinically, 14 (40.0%) children had ankle valgus at final follow up, while 21 (60%) had no appreciable ankle valgus. Mean (SD) ankle valgus deformity was 9.0 (9.2) degrees.Radiologically, frequency of ankle valgus was as shown in table 6.

Table 6. Ankle Valgus Deformity in Congenital Pseudarthrosis of The Tibia.

Ankle valgus (degrees)	Number (n=35)	Frequency
0 – 10	21	60.0%
11 – 20	12	34.3%
21 – 40	02	5.7%

Ankle valgus deformity was significantly more in children in whom there was associated fibular pseudarthrosis and in those who sustained refractures (table 7).

Table 7. Variables Influencing Ankle Valgus Deformity in Congenital Pseudarthrosis of the Tibia.

Variables	(n)	Valgus >10 deg	Significance
Age at surgery	<3 years (23)	7	p=0.93 (NS)
	>3 years (12)	7	
Refracture	present(9)	8	p=0.005 (S)
	absent (26)	6	
No. of subsequent Surgeries	<2 surgeries (29)	9	P=0.13 (NS)
	>2 surgeries (6)	5	
Type of I.M. rod	rush rod (32)	11	p=0.94 (NS)
	telescopic rod (3)	3	
Lower third location of CPT	yes (27)	11	p=0.09 (NS)
	no (8)	3	
Fibular pseudarthrosis	present (29)	13	p=0.013 (S)
	absent (6)	1	

DISCUSSION

Variables Influencing Union of Pseudarthrosis :

Age at Index Surgery:

According to Grill[7], Boero and Hardinge[8] the percentage of consolidation was better in patients who underwent surgery after 3 to 5 years of age. Grill[7] reported a healing rate of 64% in children under three and 92% in those between six and nine years old. Boero found the percentage of consolidation in those patients who were operated on after reaching the age of 5 years to be 87.5% in contrast to the rate of fusion of 14.2% in patients operated upon at ages younger than 5 years. On the contrary, Boyd and Sage recommended operation as soon as the procedure can be undertaken safely. While Gilbert[6], Fern[5] and Cho[3] did not find any difference in the rate of union in relation to age at surgery. In the present study it was found that union rate at final follow up was 86% in those who underwent surgery before three years of age as compared that of 92% who were treated after 3 years of age.

Nevertheless, when analyzed statistically using Cox regression analysis, age at index surgery was neither found to influence primary union nor final union at last follow up.

Site of Pseudarthrosis:

Kim[14] in his study stated that factor which is related to failure of union of pseudarthrosis was distal location of pseudarthrosis. He reported union rate of 58% in those with pseudarthrosis at distal third as compared to 100% union rate in those with pseudarthrosis at the junction of middle and proximal third. But site of pseudarthrosis did not influence the union significantly in our study group.

Associated Fibular Pseudarthrosis:

As per Tudisco[15], Dobbs[8] and Kim[14], associated fibular pseudarthrosis was responsible for most of failure of union. Keret[13] in his EPOS multicentre study found that fusion rate of congenital pseudarthrosis of the tibia was higher when there was a pseudarthrosis of fibula (67%) than when there was a normal fibula (59%). However, in this study presence of fibular pseudarthrosis was not found to influence the union of pseudarthrosis.

Variables Influencing Refracture:

Cho[3] in his study of refracture after Ilizarov osteosynthesis stated that risk of refracture was higher when osteosynthesis was performed below age of 4 years, when tibial cross- section area was narrow and when associated with persistent fibular pseudarthrosis. Likewise, Dobbs[8] concluded that prevalence of refracture was higher (50%) in patients in whom fibular pseudarthrosis was persistent as compared to those who had a resection with or without internal fixation of fibular pseudarthrosis (33%). In the present study the rate of refracture was 21.7% if the index surgery was performed before 3 years of age whereas it was 33.3% if the surgery was done after 3 years of age. Fibular pseudarthrosis was present in all the patients who sustained refracture whereas none of the patients with normal fibula encountered refracture. However, when analyzed statistically, neither age at the index surgery or associated fibular pseudarthrosis show any significant influence on occurrence of refracture.

Variables Influencing Limb-length Inequality:

Dobbs[4] suggested that limb length discrepancy was primarily the result of chronic resorption of bone at the pseudarthrosis site prior to surgery and secondarily as a result of operative resection of pseudarthrosis.. In this study the same view was reinforced that patients who underwent index surgery prior to 3 years of age had relatively less limb-length inequality (4.5% mean final shortening) as compared to those who underwent surgery after 3 years of age (5.4% mean final shortening).

The other factors which were found to be significantly influencing the limb shortening were more number of subsequent surgeries following index procedure and occurrence of refracture. Those patients with refracture and those with more than two subsequent surgeries following index surgery had comparatively more limb length discrepancy.

Variables Influencing Ankle Valgus:

Anderson[1] and Dobbs[4] concluded that valgus deformity of ankle was due to altered growth of distal tibial physis and associated fibular pseudarthrosis. In our study, the variables which were found to be significantly influencing ankle deformity were associated fibular pseudarthrosis and occurrence of refracture. Those patients with associated fibular pseudarthrosis had more chances of ankle valgus deformity than those with intact fibula. Similarly, those patients who sustained refracture had more chances of ankle valgus deformity than those who did not sustain refracture. None of the other variables including site of pseudarthrosis, age at index surgery, frequency of subsequent surgeries, type of intramedullary rod (rush rod/ telescopic rod) and were found to influence ankle deformity.

CONCLUSION

Excision of pseudarthrosis combined with intramedullary rodding and cortical bone grafting provides a reliable method for achieving union in congenital pseudarthrosis of the tibia, with a high rate of sound union at long-term follow-up. However, complications such as refracture, limb-length discrepancy, and ankle valgus deformity remain significant challenges.

The present study demonstrates that none of the evaluated variables significantly influenced union or refracture rates, suggesting that achieving adequate mechanical stability and biological augmentation may be more critical than individual patient or disease-related factors. In contrast, limb-length discrepancy was significantly influenced by age at surgery, number of subsequent procedures, and occurrence of refracture, while ankle valgus deformity was strongly associated with fibular pseudarthrosis and refracture.

These findings highlight the importance of early and sustained union, as well as long-term mechanical support, in optimizing outcomes in CPT. Further large-scale, multicenter prospective studies are required to better define prognostic factors and guide treatment strategies.

Clinical Relevance

- Intramedullary rodding with cortical bone grafting is an effective strategy for achieving high union rates in CPT.
- Age at surgery does not significantly influence union; however, early intervention may help reduce limb-length discrepancy.
- Prevention of refracture is critical, as it contributes significantly to both limb shortening and ankle deformity.
- Fibular pseudarthrosis plays a key role in the development of ankle valgus and should be carefully monitored during follow-up.
- Long-term intramedullary support and protective bracing until skeletal maturity are essential to maintain union and prevent complications.

Limitation of the Study

All patients must be followed up to skeletal maturity and with uniform follow up, all variables should be assessed with logistic regression analysis. As the follow up period in this study was not uniform, so we applied Cox regression analysis.

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