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Association between preoperative concurrent fibular pseudarthrosis and risk of postoperative ankle valgus in patients with congenital pseudarthrosis of the tibia

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ABSTRACT

Background This study evaluated the correlation of preoperative concurrent fibular pseudarthrosis with the risk of ankle valgus deformity in patients with congenital pseudarthrosis of the tibia (CPT) who underwent successful surgical treatment.

Method The children with CPT who were treated at our institution between 1 January 2013 and 31 December 2020 were retrospectively reviewed. The independent variable was preoperative concurrent fibular pseudarthrosis, and the dependent variable was postoperative ankle valgus. Multivariable logistic regression analysis was performed after adjusting for variables that might affect the risk of ankle valgus. Subgroup analyses with stratified multivariable logistic regression models were used to assess this association.

Results Of the 319 children who underwent successful surgical treatment, 140 (43.89%) developed ankle valgus deformity. Moreover, 104 (50.24%) of 207 patients with preoperative concurrent fibular pseudarthrosis developed an ankle valgus deformity compared with 36 (32.14%) of 112 patients without preoperative concurrent fibular pseudarthrosis (p=0.002). After adjusting for sex, body mass index, fracture age, age of patient undergoing surgery, surgery method, type 1 neurofibromatosis (NF-1), limb-length discrepancy (LLD), CPT location and fibular cystic change, patients with concurrent fibular pseudarthrosis presented a higher risk of ankle valgus than those without concurrent fibular pseudarthrosis (OR 2.326, 95% CI 1.345 to 4.022). This risk further increased with CPT location at the distal one-third of the tibia (OR 2.195, 95% CI 1.154 to 4.175), age <3 years of patient undergoing surgery (OR 2.485, 95% CI 1.188 to 5.200), LLD <2 cm (OR 2.478, 95% CI 1.225 to 5.015) and occurrence of NF-1 disorder (OR 2.836, 95% CI 1.517 to 5.303).

Conclusion Our results indicate that patients with CPT and preoperative concurrent fibular pseudarthrosis have a significantly increased risk of ankle valgus compared with those without preoperative concurrent fibular pseudarthrosis, particularly in those with CPT location at the distal third, age <3 years at surgery, LLD <2 cm and NF-1 disorder.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Fibrotic lesions of fibular are thought to be a common pathophysiology that causes closely related ankle valgus in children with congenital pseudarthrosis of the tibia (CPT). Earlier research has demonstrated that shortening or pseudarthrosis of the fibula was associated with ankle valgus and proximal tibial valgus deformities. However, their conclusions need to be further confirmed through a study with larger sample size.

WHAT THIS STUDY ADDS

⇒ Patients with CPT having preoperative concurrent fibular pseudarthrosis had a significantly higher risk of postoperative ankle valgus than those without concurrent fibular pseudarthrosis. This risk further increased with CPT location at the distal one-third of the tibia, age <3 years of patients undergoing surgery, LLD <2 cm and occurrence of type 1 neurofibromatosis (NF-1).

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ Our study alerted paediatricians to the impact of fibular lesions on ankle valgus after CPT surgery. And further provides some clinical advice as follows: age at the index surgery for patients with concurrent fibular pseudarthrosis should be deferred to 3–10 years; postoperative ankle protection in these patients with CPT location at the distal one-third of the tibia, age <3 years of patients undergoing surgery, LLD <2 cm and occurrence of NF-1 need to be performed more carefully to avoid the risk of ankle valgus deformity.

INTRODUCTION

Congenital pseudarthrosis of the tibia (CPT) is one of the most challenging conditions in paediatric orthopaedics. Its incidence varies between 1 in 140 000 and 1 in 250 000.[1] Our latest study revealed that 68.9% of these cases were associated with type 1 neurofibromatosis

(NF-1) and 53.7% with fibrous dysplasia in China.[2 3] Almost half of the fractures occur before the age of 2 years, with most patients presenting unilateral signs, particularly occurring between the middle and distal one-third of the tibia. Notably, more than half of the cases simultaneously involve fibula.[4]

Various prognostic factors are considered to be associated with this disease.[5 6] Neurofibromatosis has an uncertain impact on the course of the disease.[7 8] In addition, the earlier a fracture occurs, the worse adverse consequences are expected.[6 9] Moreover, the more distal but closer to the ankle, the more complicated to fix the distal end of the fractured bone.[6] Severe atrophy and extensive sclerosis of bone fragments in pseudarthrosis, shortened tibia and the coexistence of fibular pseudarthrosis are also considered to be poor prognostic factors affecting CPT treatment.[6] The main goal of the current surgical treatment of CPT is to obtain long-term bone union, prevent limb-length discrepancy (LLD) and avoid mechanical axis deviation, soft-tissue lesions, nearby joint stiffness and pathological fracture.[10] Although the current surgical treatment has been partially successful, some patients showing good results initially have experienced serious functional complications such as ankle valgus, leading to deterioration of ankle function along with the extension of follow-up time.[6 11]

Although the aetiology of ankle valgus in children with CPT is poorly understood,[12] fibular lesions are considered as a closely related common pathophysiology.[13] Some authors believe that the presence or absence of fibular pseudarthrosis does not cause refracture of the tibia, whereas the concurrent fibular pseudarthrosis involvement is likely to cause ankle valgus deformity.[14–16] Deng *et al* demonstrated that shortening or pseudarthrosis of the fibula was associated with ankle valgus and proximal tibial valgus deformities.[17] However, their conclusions need to be further confirmed through a study with larger sample size. Accordingly, this study was aimed to evaluate the study findings of the correlation between the presence or absence of fibular pseudarthrosis and the risk of ankle valgus in patients with CPT showing successful healing; this study has a large sample size to identify a distinct population more likely to have postoperative ankle valgus with fibular pseudarthrosis.

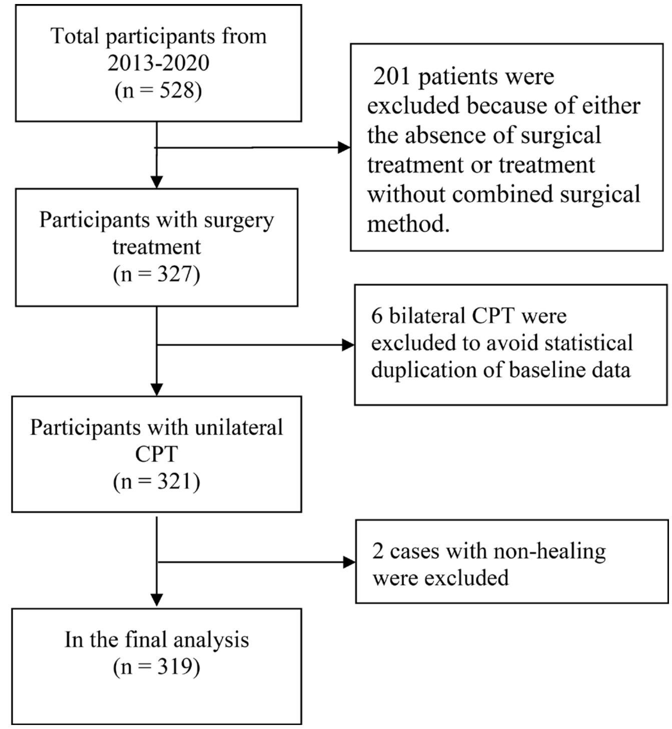

Figure 1 The flow chart of the study sample selection.

MATERIALS AND METHODS

Patient and public involvement

Patients and/or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this research.

Study design

When patients are admitted to the hospital, they or their parents usually sign an informed consent form, which includes a clause that their child's medical history and clinical data will be used for retrospective or prospective clinical studies while ensuring that their privacy is not disclosed. We retrospectively reviewed all 528 children with CPT who were treated at our institution from 1 January 2013 to 31 December 2020.

The inclusion criteria were as follows: (1) availability of follow-up data of more than 18 months, (2) diagnosed with CPT and received combined surgical treatment,[18] (3) CPT being unilateral, (4) operations being performed in the Hunan Children's Hospital and (5) a complete dataset existed for each patient. The exclusion criteria were as follows: (1) received surgical treatment at another hospital, (2) comorbidities such as tumours requiring surgical treatment and (3) had any known bone disease or other chronic diseases (eg, coeliac disease, thyroid disorder and systemic toxicity of glucocorticoids).

According to the inclusion and exclusion criteria, 201 patients were excluded because of either the absence of surgical treatment or treatment without combined surgical method. To avoid statistical duplication of the baseline data, six cases with bilateral CPT were excluded. To obtain more precise statistical results, two non-healing cases were excluded. Finally, 319 patients were included in this study. The flow chart of sample selection is shown in figure 1. We finally included 319 paediatric patients with Crawford type IV CPT who underwent surgical treatment.[19] Three orthopaedic surgeons retrospectively reviewed all clinical and imaging examination data of these patients. Medical records were reviewed for demographic information, age at fracture, age of patients undergoing surgery, concurrent fibular pseudarthrosis, CPT location, cystic changes in the fibula, surgical method and complication of ankle valgus. Follow-up was defined as 1 year after the last bone union.

Surgical methods

We divided the 319 cases into two groups based on the surgical method used: cross-union group and non-cross-union group. The 'cross-union' procedure was performed as described in previous literature,[16 20] which involves union of the tibia and fibula through autogenous iliac wrapped bone transplantation. Moreover, depending on the fibular condition (intact or not), the cross-union group was treated with either of the two surgical techniques, 3-in-1 and 4-in-1.[18 21] Currently, there are no guidelines for the treatment of fibular pseudarthrosis.[22] The type of surgical treatment used for CPT was based on the presence or absence of concurrent fibular pseudarthrosis; if the fibula was intact, then 3-in-1 technique was performed, and if the fibula was incomplete, then 4-in-1 technique was performed. Regardless of the group, intramedullary rod fixation was performed through the ankle. All cases included in this study were treated by the same surgical group.

Covariates

In this study, independent variable was the presence of concurrent fibular pseudarthrosis, and dependent variable was the occurrence of postoperative ankle valgus. The degree of ankle valgus was assessed using the tibiotalar angle measured at the intersection of the anatomic axis of the tibia and the superior articular surface of the talar dome. Patients with >4° ankle valgus were defined as having ankle valgus deformity. The covariates were as follows: sex, body mass index (BMI), fracture age, age of patients undergoing surgery, NF-1, surgery method, LLD, CPT location and cystic change in the fibula. We considered 2 cm LLD as a turning point and an indication for treatment of patients with a leg length discrepancy of ≥2 cm.[23]

Statistical analysis

Significant differences between different groups were assessed using the χ^2 test and one-way analysis of variance or Kruskal-Wallis's H test. The association between the risk of ankle valgus in patients and the presence of concurrent fibular pseudarthrosis was estimated using multivariable logistic regression models. Stratified multivariable logistic regression was then used to perform subgroup analyses. Statistical significance was set at p<0.05. All calculations were performed using the R software, V.3.4.3 (http://www.R-project.org; The R Foundation) and Empower software (www.empowerstats.com; X&Y Solutions, Boston, Massachusetts, USA).

RESULTS

Demographic and clinical data of the patients are summarised in table 1. Of the 319 patients undergoing surgery, 141 (44.20%) were treated with 4-in-1, and 178 (55.80%) were treated with 3-in-1 technique. A total of 140 (43.89%) developed ankle valgus deformity, and 104 (50.24%) of 207 patients with concurrent fibular pseudarthrosis developed ankle valgus deformity compared with 36 (32.14%) of 112 patients without concurrent fibular pseudarthrosis (p=0.002).

Table 2 shows the results of bivariate analysis. Concurrent fibular pseudarthrosis, cystic change in the fibula, age of patient undergoing surgery and LLD were identified as risk factors for ankle valgus (p<0.05). We also conducted multivariable logistic regression analyses for independent correlation of NF-1, CPT location, fibular cystic change, age of patient undergoing surgery and LLD with the risk of ankle valgus (online supplemental table S1). The results showed that only NF-1 was a significant independent risk factor for ankle valgus after adjusting for other covariates.

Table 3 shows the relationship between concurrent fibular pseudarthrosis and ankle valgus in different models. The results from unadjusted, minimally adjusted and fully adjusted analyses are shown simultaneously according to the recommendation of the Strengthening the Reporting of Observational Studies in Epidemiology statement.[24] In the fully adjusted model, OR for developing ankle valgus deformity in patients with concurrent fibular pseudarthrosis was 2.326 (95% CI 1.345 to 4.022) compared with those without concurrent fibular pseudarthrosis.

Table 4 shows the results of subgroup analyses stratified by NF-1, CPT location, age of patient undergoing surgery and LLD. In patients with CPT location of distal 1/3 (OR 2.195, 95% CI 1.154 to 4.175), age <3 years of patient undergoing surgery (OR 2.485, 95% CI 1.188 to 5.200), LLD <2 cm (OR 2.478, 95% CI 1.225 to 5.015) and NF-1 positive (OR 2.836, 95% CI 1.517 to 5.303), the risk of ankle valgus further increased with the presence of concurrent fibular pseudarthrosis.

DISCUSSION

Considering that CPT is a rare disease,[1] this study used a large sample size to comprehensively investigate whether concurrent fibular pseudarthrosis affects the risk of postoperative ankle valgus deformity. Our study demonstrated that after adjusting for potential risk factors of developing ankle valgus deformity, patients with CPT having preoperative concurrent fibular pseudarthrosis had a significantly higher risk of postoperative ankle valgus than those without concurrent fibular pseudarthrosis. In particular, this risk further increased with CPT location at the distal one-third of the tibia, age <3 years of patient undergoing surgery, LLD <2 cm and occurrence of NF-1.

There are several treatments available for CPT to achieve a long-term bone union.[11 20 25] However, little attention has been paid to ankle valgus deformity that occurs during the postoperative period. Further, this is considered by many researchers as a possible cause of postoperative refracture.[6 14–16] Therefore, it is necessary to identify high-risk patients and correct modifiable risk factors to minimise the risk of ankle valgus. In this study,

Table 1 Demographic and clinical data of patients included in the study

Fibular pseudarthrosis	Total (319)	Present (207)	Absent (112)	P value
BMI (kg/m^2), mean±SD	16.76±3.19	16.74±3.09	16.80±3.38	0.862
Sex, n (%)				
Male	202 (63.32)	139 (67.15)	63 (56.25)	0.054
Female	117 (36.68)	68 (32.85)	49 (43.75)	
Fracture age (years), mean±SD	1.68±2.01	1.57±2.11	1.45±1.59	0.596
Age of patient undergoing surgery (years), mean±SD	3.84±3.31	3.71±3.34	3.15±3.40	0.156
LLD (cm), mean±SD	2.58±2.91	2.72±3.14	1.52±2.16	<0.001
Surgery method, n (%)				
Non-cross union	141 (44.20)	79 (44.10)	62 (44.30)	0.164
Cross union	178 (55.80)	100 (55.90)	78 (55.70)	
CPT location, n (%)				
Proximal 2/3	85 (26.65)	44 (39.29)	41 (19.81)	<0.001
Distal 1/3	234 (73.35)	166 (80.19)	68 (60.71)	
NF-1, n (%)				
Yes	231 (72.41)	147 (71.01)	84 (75.00)	0.447
No	88 (27.59)	60 (28.99)	28 (25.00)	
Fibular cystic change, n (%)				
Yes	23 (7.21)	4 (1.93)	19 (16.96)	<0.001
No	296 (92.79)	203 (98.07)	93 (83.04)	
Ankle valgus, n (%)				
Yes	140 (43.89)	104 (50.24)	36 (32.14)	0.002
No	179 (56.11)	103 (49.76)	76 (67.86)	

Mean±SD for continuous variables: p value was calculated using one-way ANOVA (normal distribution) and Kruskal-Wallis H test (skewed distribution). n (%) for categorical variables: p value was calculated using χ^2 test.
ANOVA, analysis of variance; BMI, body mass index; CPT, congenital pseudarthrosis of the tibia; LLD, limb-length discrepancy; NF-1, type 1 neurofibromatosis.

we observed that concurrent fibular pseudarthrosis and NF-1 were significant independent risk factors for developing ankle valgus after bone union, which is consistent with the results of previous studies.[6 15 26–28] However, none of these studies discussed the association between concurrent fibular pseudarthrosis and risk of ankle valgus.

Previous studies have reported the presence of concurrent fibular pseudarthrosis in more than half of total cases.[29 30] Our study yielded consistent results. Studies of normal growth have reported distal migration of the fibula relative to the tibia and that the contribution to longitudinal growth from the proximal fibular epiphysis is greater than that from the distal fibular epiphysis (61% vs 39%).[31–33] However, histological studies indicated the presence of fibrous hamartoma tissue and an increase in the thickness of abnormal periosteum at the site of pseudarthrosis.[34 35] We speculated that concurrent fibular pseudarthrosis is a poor prognostic factor for the growth of the tibia and fibula after bone union and leads to ankle valgus. The effect on the subsequent growth of the fibula was particularly pronounced in the distal third of the tibia. Our subgroup analyses further supported the hypothesis that CPT located in the distal third tibia had a higher risk of ankle valgus in patients with concurrent fibular pseudarthrosis.

The choice of age at the index surgery for patients with CPT is still controversial. Various studies have shown that age <3 years is an unfavourable prognostic factor for CPT treatment and that these patients have a higher rate of bone nonunion.[14 15 36 37] However, our previous study showed that there was no need to defer surgery time until the child was older than 3 years.[38] Other studies have similarly supported the safety and effectiveness of surgical intervention in young patients aged 1–3 years.[6 39] Our study further showed that age <3 years at the index surgery had no significant effect on the risk of developing ankle valgus. This result suggests that there is no need to delay surgery to the age of 3–10 years. Notably, our subgroup analysis suggested a higher risk of postoperative ankle valgus in children under 3 years of age with concurrent fibular pseudarthrosis. Therefore, we recommend that age at the index surgery for patients with concurrent fibular pseudarthrosis could be deferred.

The site of the pseudarthrosis also plays a crucial role. Distal fractures closer to the ankle are more

Table 2 Bivariate analysis of the risk factors of ankle valgus

Ankle valgus	Statistics, n (%)	OR (95% CI)	P value
Sex			
Female	117 (36.677)	Reference	
Male	202 (63.323)	0.914 (0.578 to 1.445)	0.699
Fracture age (years)			
<1	163 (51.097)	Reference	
≥1	156 (48.903)	1.326 (0.851 to 2.066)	0.212
Age of patient undergoing surgery (years)			
<3	190 (59.561)	Reference	
≥3	129 (40.439)	2.266 (1.435 to 3.579)	0.001
Surgery method			
Non-cross union	141 (44.2)	Reference	
Cross union	178 (55.8)	1.0 (0.6 to 1.6)	0.978
NF-1			
No	88 (27.586)	Reference	
Yes	231 (72.414)	1.537 (0.927 to 2.547)	0.096
CPT location			
Proximal 2/3	85 (26.646)	Reference	
Distal 1/3	234 (73.354)	1.626 (0.973 to 2.718)	0.063
Fibular cystic change			
No	296 (92.790)	Reference	
Yes	23 (7.210)	0.331 (0.120 to 0.916)	0.033
LLD (cm)			
<2	204 (63.950)	Reference	
≥2	115 (36.050)	2.114 (1.328 to 3.364)	0.002
BMI (kg/m^2)			
<20	296 (92.790)	Reference	
≥20	23 (7.210)	1.432 (0.612 to 3.349)	0.408
Concurrent fibular pseudarthrosis			
No	112 (35.110)	Reference	
Yes	207 (64.890)	2.132 (1.318 to 3.449)	0.002

BMI, body mass index; CPT, congenital pseudarthrosis of the tibia; LLD, limb-length discrepancy; NF-1, type 1 neurofibromatosis.

difficult to fix and associated with greater complications.[11] In addition, CPT located in the distal third undoubtedly has a more pronounced negative effect on the specific growth pattern of the tibia and fibula.[33] This may cause faster growth in the tibia than the fibula, leading to ankle valgus deformity. The results of our subgroup analysis further validated this conclusion, suggesting a higher risk of postoperative ankle

Table 3 Relationship between concurrent fibular pseudarthrosis and ankle valgus in different models

Concurrent fibular pseudarthrosis	Unadjusted model β, OR (95% CI)	Minimally adjusted model β, OR (95% CI)	Fully adjusted model β, OR (95% CI)
No	Reference	Reference	Reference
Yes	2.132 (1.318 to 3.449)	2.270 (1.370 to 3.759)	2.326 (1.345 to 4.022)
P value	0.00205	0.00145	0.00252

Unadjusted model: adjusted for none.
Minimally adjusted model: adjusted for sex, BMI, fracture age, and age of patient undergoing surgery.
Fully adjusted model: adjusted for sex, BMI, fracture age, age of patient undergoing surgery, surgery method, NF-1, LLD, CPT location and fibular cystic change.
BMI, body mass index; CPT, congenital pseudarthrosis of the tibia; LLD, limb-length discrepancy; NF-1, type 1 neurofibromatosis.

Table 4 Effect modification of patients with concurrent fibular pseudarthrosis on the risk of ankle valgus compared with those without concurrent fibular pseudarthrosis, stratified by NF-1, CPT location, age of patient undergoing surgery and LLD

	OR (95% CI)	P value
CPT location		
Proximal 2/3	1.580 (0.563 to 4.431)	0.3850
Distal 1/3	2.195 (1.154 to 4.175)	0.0166
Age of patient undergoing surgery (years)		
<3	2.485 (1.188 to 5.200)	0.0157
≥3	1.392 (0.604 to 3.208)	0.4370
LLD (cm)		
<2	2.478 (1.225 to 5.015)	0.0116
≥2	1.035 (0.394 to 2.723)	0.9441
NF-1		
No	0.981 (0.335 to 2.871)	0.9725
Yes	2.836 (1.517 to 5.303)	0.0011

Above models adjusted for: sex, BMI, fracture age, age of patient undergoing surgery, surgery method, NF-1, LLD, CPT location, and fibular cystic change. In each case, the model was not adjusted for stratification variable itself.
BMI, body mass index; CPT, congenital pseudarthrosis of the tibia; LLD, limb-length discrepancy; NF-1, type 1 neurofibromatosis.

valgus in patients with CPT and concurrent fibular pseudarthrosis located in the distal third. LLD is considered an independent factor influencing ankle valgus in patients with CPT and concurrent fibular pseudarthrosis. Based on previous evidence that LLD resulted in more work of the ankle joint, inadequate distribution of mechanical loads and gait kinematics asymmetries,[40 41] we speculated that strong association between LLD and ankle valgus may be related to the altered mechanical distribution and gait patterns at the ankle. Neurofibromatosis may have a detrimental effect on pseudarthrosis prognosis. Our results also indicate that NF-1 is a negative independent factor for ankle valgus. Furthermore, in our subgroup analysis, the incidence of ankle valgus was significantly higher in NF-1-positive patients with concurrent fibular pseudarthrosis than in those without concurrent fibular pseudarthrosis. However, many studies have shown that the role of NF-1 in the prognosis of CPT is uncertain, similar to its impact on the development of complications.[42–44] However, our results suggest a different conclusion: postoperative ankle protection in these patients needs to be performed more carefully to avoid the risk of ankle valgus deformity.

Our study has some limitations. The observational design of the cross-sectional study did not allow us to determine the time correlation of fibular pseudarthrosis with the risk of postoperative ankle valgus. Therefore, a longitudinal study is warranted. Moreover, we did not find any difference in the risk of ankle valgus with respect to the location of fibula pseudarthrosis. The sample size at the proximal region was too small for statistical analysis. In addition, biases caused by other potential confounding factors were not excluded. For example, the data of the preoperative course were missing because many parents could not record it accurately; however, we did not adjust for this potential confounding factor in this study. Some patients experienced a refracture during the healing process; whether this has an effect on the ankle joint is yet to be determined. It is mandatory to perform an evaluation at skeletal maturity due to growth abnormalities of the distal tibia and fibula; however, this cross-sectional study lacks long-term follow-up results. Our another ongoing randomised controlled trial may be able to address these limitations.

CONCLUSION

Conclusively, patients with CPT and concurrent fibular pseudarthrosis have an increased risk of ankle valgus compared with those without concurrent fibular pseudarthrosis. This risk further increased with CPT location at the distal third, age <3 years of patient undergoing surgery, LLD <2 cm and occurrence of NF-1 disorder.

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Contributors GY conceived and designed the study; analysed and interpreted the data; wrote the paper. HY: conceived and designed the study; review and record data; analysed and interpreted the data; wrote the paper. ZL: conceived and designed the study; analysed and interpreted the data; wrote the paper. HM: analysed and interpreted the data; review and record data. KL: analysed and interpreted the data; review and record data. GZ: analysed and interpreted the data; review and record data. QT: conceived and designed the study; analysed and interpreted the data. GY responsible for the overall content.

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Competing interests None declared.

Patient and public involvement Patients and/or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this research.

Patient consent for publication Consent obtained from parent(s)/guardian(s).

Ethics approval This study was approved by Institutional Review Board of the Hunan Children's Hospital permission and informed consent from all included patients, and all informed consents were obtained from all included patients' parents or legal guardian. Participants gave informed consent to participate in the study before taking part.

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REFERENCES

1 Hefti F, Bollini G, Dungl P, *et al*. Congenital pseudarthrosis of the tibia: history, etiology, classification, and epidemiologic data. *J Pediatr Orthop B* 2000;9:11–5.
2 Li Z, Yu H, Huang Y, *et al*. Analysis of risk factors affecting Union and refracture after combined surgery for congenital pseudarthrosis of the tibia: a retrospective study of 255 cases. *Orphanet J Rare Dis* 2022;17:245.
3 Zhou Y, Tan Q, Liu K, *et al*. Epidemiological and clinical characteristics of congenital pseudarthrosis of the tibia in china. *Front Pediatr* 2022;10:943917.
4 Ohnishi I, Sato W, Matsuyama J, *et al*. Treatment of congenital pseudarthrosis of the tibia: a multicenter study in Japan. *J Pediatr Orthop* 2005;25:219–24.
5 Khan T, Joseph B. Controversies in the management of congenital pseudarthrosis of the tibia and fibula. *Bone Joint J* 2013;95-B:1027–34.
6 Zargarbashi R, Bagherpour A, Keshavarz-Fathi M, *et al*. Prognosis of congenital pseudarthrosis of the tibia: effect of site of tibial pseudarthrososis and fibular involvement. *J Pediatr Orthop* 18, 2021.
7 Boero S, Catagni M, Donzelli O, *et al*. Congenital pseudarthrosis of the tibia associated with neurofibromatosis-1: treatment with ilizarov's device. *J Pediatr Orthop* 1997;17:675–84.
8 Van Royen K, Brems H, Legius E, *et al*. Prevalence of neurofibromatosis type 1 in congenital pseudarthrosis of the tibia. *Eur J Pediatr* 2016;175:1193–8.
9 Boyd HB. Pathology and natural history of congenital pseudarthrosis of the tibia. *Clinical Orthopaedics and Related Research* 1982;166(amp;NA):5
10 Shah H, Rousset M, Canavese F. Congenital pseudarthrosis of the tibia: management and complications. *Indian J Orthop* 2012;46:616–26.
11 Paley D. Congenital pseudarthrosis of the tibia: biological and biomechanical considerations to achieve Union and prevent refracture. *J Child Orthop* 2019;13:120–33.
12 Davids JR, Valadie AL, Ferguson RL, *et al*. Surgical management of ankle valgus in children: use of a transphyseal medial malleolar screw. *J Pediatr Orthop* 1997;17:3–8.
13 Dias LS. Valgus deformity of the ankle joint: pathogenesis of fibular shortening. *J Pediatr Orthop* 1985;5:176–80.
14 Dobbs MB, Rich MM, Gordon JE, *et al*. Use of an intramedullary rod for treatment of congenital pseudarthrosis of the tibia. A long-term follow-up study. *J Bone Joint Surg Am* 2004;86:1186–97.
15 Vanderstappen J, Lammens J, Berger P, *et al*. Ilizarov bone transport as a treatment of congenital pseudarthrosis of the tibia: a long-term follow-up study. *J Child Orthop* 2015;9:319–24.
16 Choi IH, Lee SJ, Moon HJ, *et al*. " 4-in-1 osteosynthesis " for atrophic-type congenital pseudarthrosis of the tibia. *J Pediatr Orthop* 2011;31:697–704.
17 Deng H, Mei H, Wang E, *et al*. The association between fibular status and frontal plane tibial alignment post-union in congenital pseudarthrosis of the tibia. *J Child Orthop* 2021;15:261–9.
18 Zhu G-H, Mei H-B, He R-G, *et al*. Combination of intramedullary rod, wrapping bone grafting and ilizarov's fixator for the treatment of Crawford type IV congenital pseudarthrosis of the tibia: mid-term follow up of 56 cases. *BMC Musculoskelet Disord* 2016;17:443.
19 Crawford AH, Bagamery N. Osseous manifestations of neurofibromatosis in childhood. *J Pediatr Orthop* 1986;6:72–88.
20 Liu Y, Yang G, Liu K, *et al*. Combined surgery with 3-in-1 osteosynthesis in congenital pseudarthrosis of the tibia with intact fibula. *Orphanet J Rare Dis* 2020;15:62.
21 Yan A, Mei H-B, Liu K, *et al*. Wrapping grafting for congenital pseudarthrosis of the tibia: a preliminary report. *Medicine (Baltimore)* 2017;96:e8835.
22 Wang KK, Vuillermin CB, Eisenberg KA, *et al*. Congenital pseudarthrosis of the fibula: controlling ankle valgus in a heterogenous condition. *J Pediatr Orthop* 2020;40:e647–55.
23 Vogt B, Gosheger G, Wirth T, *et al*. Leg length discrepancy- treatment indications and strategies. *Dtsch Arztebl Int* 2020;117:405–11.
24 von Elm E, Altman DG, Egger M, *et al*. The strengthening the reporting of observational studies in epidemiology (STROBE) statement: guidelines for reporting observational studies. *Int J Surg* 2014;12:1495–9.
25 Dobbs MB, Rich MM, Gordon JE, *et al*. Use of an intramedullary rod for the treatment of congenital pseudarthrosis of the tibia. surgical technique. *J Bone Joint Surg Am* 2005;87 Suppl 1(Pt 1):33–40.
26 El-Gammal TA, El-Sayed A, Kotb MM, *et al*. Crawford type IV congenital pseudarthrosis of the tibia: treatment with vascularized fibular grafting and outcome at skeletal maturity. *J Pediatr Orthop* 2021;41:164–70.
27 Song MH, Park MS, Yoo WJ, *et al*. Femoral overgrowth in children with congenital pseudarthrosis of the tibia. *BMC Musculoskelet Disord* 2016;17:274:274.:.
28 Iamaguchi RB, Fucs PMMB, Carlos da Costa A, *et al*. Congenital pseudoarthrosis of the tibia – results of treatment by free fibular transfer and associated procedures – preliminary study. *J Pediatr Orthop B* 2011;20:323–9.
29 Keret D, Bollini G, Dungl P, *et al*. The fibula in congenital pseudoarthrosis of the tibia: the EPOS multicenter study. *Journal of Pediatric Orthopaedics, Part B* 2000;9:69–74.
30 Pannier S. Congenital pseudarthrosis of the tibia. *Orthop Traumatol Surg Res* 2011;97:750–61.
31 Kärrholm J, Hansson LI, Selvik G. Changes in tibiofibular relationships due to growth disturbances after ankle fractures in children. *The Journal of Bone & Joint Surgery* 1984;66:1198–210.
32 Pritchett JW. Growth and growth prediction of the fibula. *Clinical Orthopaedics and Related Research* 1997;334:251
33 Frick SL, Shoemaker S, Mubarak SJ. Altered fibular growth patterns after tibiofibular Synostosis in children. *J Bone Joint Surg Am* 2001;83:247–54.
34 El-Hoss J, Sullivan K, Cheng T, *et al*. A murine model of neurofibromatosis type 1 tibial pseudarthrosis featuring proliferative fibrous tissue and osteoclast-like cells. *J Bone Miner Res* 2012;27:68–78.
35 Cho TJ, Seo JB, Lee HR, *et al*. Biologic characteristics of fibrous hamartoma from congenital pseudarthrosis of the tibia associated with neurofibromatosis type 1. *J Bone Joint Surg Am* 2008;90:2735–44.
36 Kesireddy N, Kheireldin RK, Lu A, *et al*. Current treatment of congenital pseudarthrosis of the tibia: a systematic review and meta-analysis. *J Pediatr Orthop B* 2018;27:541–50.
37 Wientroub S, Grill F. Congenital pseudarthrosis of the tibia: part 1. European pediatric orthopaedic Society multicenter study of congenital pseudoarthrosis. *J Pediatr Orthop B* 2000;9:1–2.
38 Liu Y, Mei H, Zhu G, *et al*. Congenital pseudarthrosis of the tibia in children: should we defer surgery until 3 years old? *J Pediatr Orthop B* 2018;27:17–25.
39 Joseph B, Somaraju VVJ, Shetty SK. Management of congenital pseudarthrosis of the tibia in children under 3 years of age: effect of early surgery on Union of the pseudarthrosis and growth of the limb. *J Pediatr Orthop* 2003;23:740–6.
40 Zeitoune G, Nadal J, Batista LA, *et al*. Prediction of mild anatomical leg length discrepancy based on gait kinematics and linear regression model. *Gait Posture* 2019;67:117–21.
41 Aiona M, Do KP, Emara K, *et al*. Gait patterns in children with limb length discrepancy. *J Pediatr Orthop* 2015;35:280–4.
42 Feldman DS, Jordan C, Fonseca L. Orthopaedic manifestations of neurofibromatosis type 1. *J Am Acad Orthop Surg* 2010;18:346–57.
43 Horn J, Steen H, Terjesen T. Epidemiology and treatment outcome of congenital pseudarthrosis of the tibia. *J Child Orthop* 2013;7:157–66.
44 Borzunov DY, Chevardin AY, Mitrofanov AI. Management of congenital pseudarthrosis of the tibia with the ilizarov method in a paediatric population: influence of aetiological factors. *Int Orthop* 2016;40:331–9.