Vascularised fibular graft for the treatment of congenital pseudarthrosis of the tibia: long-term complications in the donor leg

Raquel B. Iamaguchi · Patricia M. M. B. Fucs ·
Antonio Carlos da Costa · Ivan Chakkour

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Abstract Treatment of congenital pseudarthrosis of the tibia is still controversial, and vascularised fibula graft is a reliable method for consolidation, although complications can occur in the donor leg after fibula grafting. This study evaluates 16 patients with congenital pseudarthrosis of the tibia (CPT) treated with contralateral fibular graft, with regard to complications in the donor leg, and assesses the influence of distal tibiofibular joint arthrodesis in these complications. All patients with CPT were consecutively submitted to surgical treatment with contralateral fibular graft. The mean follow-up was 94 months, varying from 44 to 162 months. The long-term effects in the donor leg were determined, and 12 cases with distal tibiofibular arthrodesis (group 1) were compared with a group without arthrodesis (group 2). Half of the patients had proximal migration of the lateral malleolus. Eight patients had ankle valgus. Group 1 had an average valgus tilt angle of 5.8°, while group 2 had an average angle of 1.5°. There was no significant difference between the patients with or without distal tibiofibular joint arthrodesis. The patients with a distal fibula remnant smaller than 5 cm had greater valgus tilt angle measurements. Distal tibiofibular arthrodesis was not effective in preventing all the complications in the

donor leg; however, it can lessen the severity of the complications. Our results show that a distal fibular remnant greater than 5 cm should be retained to prevent ankle valgus.

Introduction

Congenital pseudarthrosis of the tibia (CPT) is a rare disease with no consensus as to the best treatment. In the past, surgical solutions yielded negative results, and sometimes even culminated in amputation. Even with the evolution of treatment, many techniques are described, the most common being the Ilizarov external fixator through the distraction osteogenesis method with increased angiogenesis [2, 3, 10], intramedullary nailing with or without bone graft [4], and vascularised fibula graft have all been advocated for primary, or secondary surgery [3, 7, 17, 19, 21]. Advances in molecular biology have resulted in some recent studies that use recombinant human bone morphogenetic proteins for the treatment of CPT [12, 16].

Vascularised fibular graft from the contralateral leg is already common practice, with good results reported [3, 7, 17, 19, 21]. Although many complications can occur in the graft donor leg, few reports specifically relating to the donor leg have been published. Among the early complications is devascularisation of the donor leg due to a *peronea magna* artery [13, 18]. Of the late complications, valgus ankle deformity is the most common in the growing child, and some authors advise distal tibiofibular joint arthrodesis or reconstruction of fibula using a tibial autograft as surgical prevention procedures [1, 8, 11, 15].

The aim of this study was to evaluate the severity of ankle valgus and the long-term complications in the donor leg in patients submitted to vascularised fibular graft for

R. B. Iamaguchi · P. M. M. B. Fucs · A. C. da Costa · I. Chakkour
Neuromuscular Clinic, Department of Orthopaedics and
Traumatology, Santa Casa Medical School and Hospitals,
São Paulo, Brazil

P. M. M. B. Fucs
e-mail: patriciaa@fucs.com.br

R. B. Iamaguchi (✉)
Rua Cerro Cora, 1540,
CEP 05061300 São Paulo, SP, Brazil
e-mail: biraquel@bol.com.br

CPT correction, and to analyse the association of ankle valgus with distal tibiofibular arthrodesis as a prevention method.

Methods

This study evaluated the donor leg of 16 patients with CPT submitted to surgical treatment with contralateral fibular graft. The study was approved by the institution's ethics committee. All of the patients' parents or care givers gave their informed consent prior to the child's inclusion in the study.

There were eight male and eight female patients, with the right leg affected in seven, and the left in nine. The patients were treated between 1994 and 2005, and were reviewed radiographically and clinically, with a mean follow-up of 94 months (range 44–162).

The mean age on first consultation with the department was 48 months (range 2–140 months). The mean age at operation was 72 months, ranging from 18 to 149 months.

Preoperative arteriographies were obtained in ten patients, and no vascular abnormalities were identified.

The surgical procedure consisted of harvesting the contralateral fibula using a skin monitor, according to the technique described by Taylor et al. [20] and modified by Gilbert [6]. An appropriate length of fibula was removed, according to pre-operative measurements, and rods were inserted proximally and distally in the host tibia and fixed by different methods, which have been investigated in another study. The patients were divided into two groups: arthrodesis of the distal tibiofibular joint was performed in the 12 patients of group 1 and was not performed in four patients of group 2. Arthrodesis of the distal tibiofibular joint was achieved in the donor leg, by cortical freshening of the distal metaphyseal tibia and fibula, fibular autograft obtained during the harvest of the fibula and a transfixating tibiofibular screw with the ankle in mild dorsiflexion (0–10 degrees). A below-knee plaster was applied in all patients for six weeks. Weight-bearing was allowed, according to the progression of the radiographic union. The length of the distal fibula remnant of the donor leg was measured.

No vascular complications arose in the donor leg after surgery.

Consolidation of tibiofibular arthrodesis was defined in consecutive radiographs, showing trabecular bone in between the two bones, and clinically without pain in the donor side. After consolidation, the following data were evaluated in the radiographs:

- Proximal migration of the lateral malleolus in relation to the tibia, according to Malhotra et al. [14], which measures the relation between the distal fibular growth plate and the distal tibial growth plate and is classified

as: normal, when the distal fibular growth plate is at the level of the talar plateau; mild, if it is between the top of the talus and the distal tibial growth plate; moderate, if the distal fibular growth plate is in line with the distal tibial growth plate; and severe, if the distal fibular growth plate is proximal to the distal tibial growth plate

- Valgus tilt angle (angle between the line perpendicular to the long axis of the tibia with the distal joint surface of the tibia)
- Cortical thickening of the tibia (as compensation after fibula resection)
- Arthrosis in the distal tibiofibular joint

In the follow-up, the patients were questioned about pain, and gait was evaluated.

To study the association between the qualitative variables, contingency tables were constructed. For the quantitative variables, analysis of variance was used, in which the homogeneity of variance tested by the Bartlett test was rejected ($P < 0.05$), and the test was dismissed. The Mann-Whitney test was applied for comparison between the two groups. The Kruskal-Wallis test was used for more than two groups. Statistical significance was considered when $P < 0.05$. The software programs Epi-Info (version 3.3.2) and SPSS Statistics 17 were used. When we could not use these tests because of the low number of cases, the non-parametric tests and boxplot or scatter/dot graphs were used with SPSS Statistics data editor 17.

Results

The remaining distal fibula lengths varied from 2.0 cm to 7.3 cm (mean 4.4 cm) (Table 1). Eight patients developed an increase of valgus tilt angle, and seven had distal tibiofibular arthrodesis. For the patients with arthrodesis, the mean length was 4.0 cm and for the patients without arthrodesis, the mean length was 4.8 cm. No difference was found between the groups. There were no differences between degrees of proximal migration of the lateral malleolus and size of the distal fibular remnant ($P = 0.27$). There was an inverse correlation between the distal fibular remnant length and valgus tilt angle; although Pearson's correlation was moderate (-0.54), it was nonetheless statistically significant ($P = 0.03$) (Fig. 1).

Group 1 had an average age of 67 months and group 2 had an average age of 40 months. Age had no statistically significant influence on complications in the donor leg in either group. Nor was there any difference between degrees of proximal migration of the lateral malleolus and age ($P = 0.13$). However, an inverse correlation was observed between age and valgus tilt angle, with Pearson's correlation -0.24 ($P = 0.36$) (Fig. 2).

Table 1 Patient data

Case	Gender	Age (months)	Proximal migration lateral malleolus	Arthrodesis	Valgus tilt angle	Fibular remnant (cm)	Follow-up (months)
W.S.R.	Male	31	Mild	Yes (group 1)	12°	3.7	130
J.B.G.	Female	56	Mild	No (group 2)	0	3.7	128
L.S.G.	Male	129	Normal	Yes (group 1)	5°	7.3	51
J.A.A.R.	Female	144	Normal	No (group 2)	0	6.5	78
V.A.G.	Male	81	Normal	Yes (group 1)	25°	2.0	132
N.A.S.S.	Female	65	Normal	Yes (group 1)	0	5.6	97
E.A.B.	Female	61	Moderate	No (group 2)	6°	3.0	122
T.C.S.	Female	20	Mild	Yes (group 1)	3°	5.3	157
G.M.V.L.	Female	58	Mild	Yes (group 1)	0	5.4	61
G.F.A.	Female	149	Normal	Yes (group 1)	0	5.6	71
L.G.S.	Male	93	Normal	Yes (group 1)	0	5.3	54
A.B.C.A.	Female	69	Mild	Yes (group 1)	5°	3.0	57
L.E.J.O.	Male	39	Normal	Yes (group 1)	0	2.8	44
M.W.J.J.	Male	27	Moderate	Yes (group 1)	16°	3.2	83
C.A.G.	Male	90	Mild	No (group 2)	0	3.8	90
P.R.A.S.	Male	48	Normal	Yes (group 1)	3°	4.7	162

In the post-operative radiograph examinations, eight patients (50%) showed proximal migration of the lateral malleolus. In group 1, there were four cases classified as mild and one moderate. In group two, there were two cases classified as mild and one as moderate. There were no cases classified as severe in either group. In group 1, there were seven cases of increase in valgus tilt angle, with a mean of 5.75° (max. 25° and min. 0°) for the group. In group two, there was one case with an angle of 6°.

No patient had cortical thickening of the tibia nor was there arthrosis in the distal tibiofibular joint, and there was no complaint of pain (Fig. 3).

The screw used for distal tibiofibular arthrodesis had to be removed in one case due to screw breakage and in five cases because of loosening of the screw after growth of the legs. In three cases of loosening the patients also had pain over the head of the screw (Fig. 4).

Fig. 1 Correlation between the distal fibular remnant length and valgus tilt angle **Fig. 2** Correlation between age and valgus tilt angle

Fig. 3 Case W.S.R., male, 31 months at vascularised fibular graft, ten-year follow-up. **a** Preoperative radiographs. **b** Immediate postoperative. **c** Consolidation of distal tibiofibular arthrodesis with mild proximal

migration of lateral malleolus and 12° valgus tilt angle. **d** Ten-year postoperative radiographs showing mild leg length discrepancy

During clinical evaluation, we observed no cases of pain or disturbance of gait due to limited range of motion in the ankle (Fig. 5).

Discussion

Few studies address the complications in the donor site of fibular graft in CPT patients. Some early complications are described, such as infection, sensitivity deficits and devascularisation due to arteria peronea magna. Rosson and Singh [18], in a retrospective study in 2000, reviewed 93 arteriographs of patients submitted to fibula grafting, and they observed a peronea magna artery in 5.3% (a higher prevalence than that reported in the literature, according to the authors). Lutz et al. [13] observed this phenomenon in 0.9%, and they do not recommend the use of routine arteriography. In this study, carried out in a public hospital, it was possible to obtain arteriographs for ten patients, with normal results in all of them. We agree with Lutz et al. [13] that this investigation should be indicated only when there is a suspicion of vascular alteration.

There are many possible complications of fibula graft harvesting in patients with or without open physis [1, 8, 11]. Babhulkar et al. [1], in 1995, studied 104 patients subjected to fibula resection for bone grafting and they observed that 60 patients had complaints, the majority involving muscular weakness in the extensor hallucis longus (isolated or associated with other tendons), inability to run, numbness of the foot, and occasional cramps. Six of their patients developed ankle instability and pain, and required reconstruction. Our patients, however, were all asymptomatic with regard to the donor leg.

The main complication of fibula graft harvesting in patients with open physis is valgus deformity of the ankle [1, 8, 11]. Langeskiöld [11], in 1967, observed this and suggested distal tibiofibular arthrodesis with iliac graft as a preventative measure.

González-Herranz et al. [8], in 2003, studied 23 children who underwent to 24 fibula resections. In half of the cases, the tibiofibular joint was stabilised. After a mean 6.2-year follow-up, the complications observed were distal migration of the fibula head in 18 cases, cortical thickening of the tibia in five cases, an increase in the tibiotalar joint angle in

Fig. 4 Case V.A.G., male, 81 months at vascularised fibular graft, 11-year follow-up. **a** Preoperative radiographs. **b** Postoperative 1 month. **c** Eight-years postoperative. **d** Eleven years postoperative

Fig. 5 Case T.C.S., female, 20 months old at vascularised fibular graft, 13-year follow-up. **a** Two years postoperative. **b** Thirteen-year postoperative radiographs. **c** and **d** Thirteen-year postoperative clinical evaluation

11 cases, and proximal migration of the lateral malleolus in 13 cases. The increase in the tibiotalar joint angle was associated with proximal migration of the lateral malleolus. The authors demonstrate that fibula resection is not entirely harmless for children.

Many authors recommend arthrodesis or transfixation with a screw of the distal tibiofibular joint in patients with open physis, in order to avoid the valgus deformity of the ankle and proximal migration of the lateral malleolus [5, 7, 9, 11, 15, 17, 22]. Although we frequently observed proximal migration of the lateral malleolus, we could not associate this with clinical complications. Fragnière et al. [5], in 2003, described 20 children with open physes submitted to vascularised fibula grafting, dividing them in three groups: group A, with no prevention; group B, prevention with a transfixation tibiofibular quadricortical screw; and group C, prevention with fibula reconstruction. They observed a lower incidence of valgus deformity of the ankle in the patients with the distal tibiofibular screw fixation. The authors describe age as the only variable associated with the deformity. Omokawa et al. [15] studied 13 patients treated with vascularised fibula grafts in childhood, and observed that tibiofibular synostosis, using a modified Langenskiöld's method, prevented increase in valgus tilt angle compared to those patients without prevention. In our study, although we had a higher degree of valgus tilt angle in group 1, there was no association between distal tibiofibular arthrodesis and valgus deformity or proximal migration of the lateral malleolus, possibly due to the small number of patients. Successful arthrodesis was achieved in all group 1 patients, but it did not effectively prevent ankle valgus deformity in seven patients. However, well-known sequelae of fibular resection, such as severe ankle valgus deformity or proximal migration of the lateral malleolus, were not observed in patients with tibiofibular arthrodesis.

Babhulkar et al. [1] suggest that a distal fibular remnant should be preserved with a length of more than 8 cm, although no patient data was provided to support this recommendation. It was not possible to retain an 8 cm-long remnant in our patients, due to their young age and the need for longer grafts. The distal fibulae that developed valgus deformities were shorter on average, and an inverse correlation was observed between the distal fibular remnant length and valgus tilt angle, with a moderate Pearson's correlation which was statistically significant ($P=0.03$). We recommend that a distal fibular remnant equal or longer than 5 cm should be preserved, as the mean valgus tilt angle in such cases was 1.1 degrees, while the mean valgus for cases with a distal fibular remnant shorter than 5 cm was 7.4 degrees.

Arthrodesis requires a longer surgical time and can also cause complications with the screws, such as breakage and local pain. Despite these complications, we recommend arthrodesis of the distal tibiofibular joint for the prevention of ankle valgus because we observed only mild complications, and no cases of severe ankle valgus requiring surgical correction in these patients.

As these patients presented a rare disease and were submitted to a microsurgical procedure on the affected leg, they are being still followed-up in our clinic, and the final results will be seen after all of them have completed the growth period.

Conclusion

Proximal migration of the lateral malleolus or ankle valgus accounted for the majority of complications in patients undergoing vascularised fibular graft as a treatment for CPT. Distal tibiofibular arthrodesis was not effective in preventing all complications in the donor leg; however, it can prevent the more severe complications. We recommend

that a distal fibular remnant greater than 5 cm be retained to prevent ankle valgus.

Conflicts of interest There were no conflicts of interest concerning this study.

References

- Babhulkar SS, Pande KC, Babhulkar S (1995) Ankle instability after fibular resection. *J Bone Joint Surg Br* 77(2):258–261
- Borovecki F, Pecina-Slaus N, Vukicevic (2007) Biological mechanics of bone and cartilage remodeling—genomic perspective. *Int Orthop* 31(6):799–805
- Delgado-Martínez AD, Rodríguez-Merchán EC, Olsen B (1996) Congenital pseudarthrosis of the tibia. *Int Orthop* 20(3):192–199
- Dobbs MB, Rich MM, Gordon JE, Szymanski DA, Schoenecker PL (2005) Use of an intramedullary rod for the treatment of congenital pseudarthrosis of the tibia. Surgical technique. *J Bone Joint Surg Am* 87 Suppl 1(Pt 1):33–40
- Fragnière B, Wicart P, Mascard E, Dudousset J (2003) Prevention of ankle valgus after vascularized fibular grafts in children. *Clin Orthop Relat Res* 408:245–251
- Gilbert A (1979) Vascularized transfer of the fibular shaft. *Int J Microsurg* 1:100–109
- Gilbert A, Brockman R (1995) Congenital pseudarthrosis of the tibia. Long-term followup of 29 cases treated by microvascular bone transfer. *Clin Orthop Relat Res* 314:37–44
- González-Herranz P, del Río A, Burgos J, López-Mondejar JA, Rapariz JM (2003) Valgus deformity after fibular resection in children. *J Pediatr Orthop* 23(1):55–59
- Kanaya K, Wada T, Kura H, Yamashita T, Usui M, Ishii S (2002) Valgus deformity of the ankle following harvesting of a vascularized fibular graft in children. *J Reconstr Microsurg* 18(2):91–96
- Kristiansen LP, Steen H, Terjesen T (2003) Residual challenges after healing of congenital pseudarthrosis in the tibia. *Clin Orthop Relat Res* 414:228–237
- Langenskiöld A (1967) Pseudarthrosis of the fibula and progressive valgus deformity of the ankle in children: treatment by fusion of the distal tibial and fibular metaphyses. Review of three cases. *J Bone Joint Surg Am* 49(3):463–470
- Lee FY, Sinicropi SM, Lee FS, Vitale MG, Roye DP Jr, Choi IH (2006) Treatment of congenital pseudarthrosis of the tibia with recombinant human bone morphogenetic protein-7 (rhBMP-7). *J Bone Joint Surg Am* 88(3):627–633
- Lutz BS, Wei FC, Ng SH, Chen IH, Chen SH (1999) Routine donor leg angiography before vascularized free fibula transplantation is not necessary: a prospective study in 120 clinical cases. *Plast Reconstr Surg* 103(1):121–127
- Malhotra D, Puri R, Owen R (1984) Valgus deformity of the ankle in children with spina bifida aperta. *J Bone Joint Surg Br* 66(3):381–385
- Omokawa S, Tamai S, Takakura Y, Yajima H, Kawanishi K (1996) A long-term study of the donor-site ankle after vascularized fibula grafts in children. *Microsurgery* 17(3):162–166
- Pecina M, Vukicevic S (2007) Biological aspects of bone, cartilage and tendon regeneration. *Int Orthop* 31(6):719–720
- Pho RW, Levack B, Satku K, Patradul A (1985) Free vascularized fibular graft in the treatment of congenital pseudarthrosis of the tibia. *J Bone Joint Surg Br* 67(1):64–70
- Rosson GD, Singh NK (2005) Devascularizing complications of free fibula harvest: peronea arteria magna. *J Reconstr Microsurg* 21(8):533–538
- Sakamoto A, Yoshida T, Uchida Y, Kojima T, Kubota H, Iwamoto Y (2008) Long-term follow-up on the use vascularized fibular graft for the treatment of congenital pseudarthrosis of the tibia. *J Orthop Surg Res* 3:13
- Taylor GI, Miller GD, Ham FJ (1975) The free vascularized bone graft. A clinical extension of microvascular techniques. *Plast Reconstr Surg* 55(5):533–544
- Toh S, Harata S, Tsubo K, Inoue S, Narita S (2001) Combining free vascularized fibula graft and Ilizarov external fixator: recent approaches to congenital pseudarthrosis of the tibia. *J Reconstr Microsurg* 17(7):497–508, discussion 509
- Wiltse LL (1972) Valgus deformity of the ankle: a sequel to acquired or congenital abnormalities of the fibula. *J Bone Joint Surg Am* 54(3):595–606