VASCULARISED FIBULAR GRAFT FOR CONGENITAL PSEUDARTHROSIS OF THE TIBIA
LONG-TERM RESULTS

YOSHI UCHIDA, TETSUO KOJIMA, YOICHI SUGIOKA

From Kyushu University, Fukuoka, Japan

Five children with congenital pseudarthrosis of the tibia treated by free vascularised fibular grafts were followed up until skeletal maturity. The ipsilateral fibula was used in four cases, the contralateral fibula in one. All our cases achieved bone union, but leg length discrepancy, atrophy of the foot and ankle stiffness were frequent complications, due perhaps to the many previous operations. Vascularised fibular grafting might achieve better results if it were done as the primary procedure.

Treatment of congenital pseudarthrosis of the tibia has proved a challenge for many orthopaedic surgeons. Bypass grafts (McFarland 1951), dual onlay bone grafts (Boyd and Sage 1958), fragmentation and intramedullary rod fixation (Sofield and Millar 1959) have all been employed. However, the long-term results of these methods have not proved satisfactory, with many patients requiring repeated procedures before bone union is achieved, and a high incidence of amputation (Murray and Lovell 1982; Crossett et al 1989). Encouraging results using pulsed electromagnetic fields have been reported (Bassett, Caulo and Kort 1981; Sutcliffe and Goldberg 1982), but a long time is required before union is achieved, and union may then prove tenuous. The technique is often ineffective if there is a large gap between the fragments.

The experimental use of vascularised bone grafts was first described by Östrup and Fredrickson (1974), and Taylor, Miller and Ham (1975) who were the first to use a vascularised fibular graft in clinical practice. Its use in the treatment of congenital pseudarthrosis of the tibia was first described by Judet et al 1978, and good results have since been reported by a number of investigators (Chen, Yu and Wang 1979; Weiland and Daniel 1980; Pho et al 1985; Gordon, Weulker and Jergesen 1986; Dormans et al 1990). To date, however, long-term results have not been published. As noted by Boyd and Sage (1958) and Van Nes (1966), the final assessment of treatment should await skeletal maturity. In this study, we report the results in five patients treated by vascularised fibular grafts who were followed until skeletal maturity.

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Y. Uchida, MD, Assistant Professor
T. Kojima, MD, Orthopaedic Surgeon
Y. Sugioka, MD, Professor and Chairman of Orthopaedic Surgery
Department of Orthopaedic Surgery, Faculty of Medicine, Kyushu University 3-1-1, Maidashi, Higashi-ku, Fukuoka 812, Japan.

Correspondence should be sent to Dr Y. Uchida.

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Diagram showing the operative technique for a free vascularised fibular graft. The vascular anastomoses are performed between the peroneal and the anterior tibial vessels.
Table I. Details of five patients treated for congenital pseudarthrosis of the tibia

<table>
<thead>
<tr>
<th>Case</th>
<th>Sex/age at operation</th>
<th>Side</th>
<th>Number of previous operations</th>
<th>Side of donor fibula</th>
<th>Time to union (mth)</th>
<th>Age at follow-up (yr)</th>
<th>Tibial length discrepancy (cm) Postoperative</th>
<th>Follow-up</th>
<th>Anterior bowing of tibia (degrees) Complications</th>
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<tr>
<td>1</td>
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<td>L†</td>
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<td>L</td>
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<td>F/8</td>
<td>L‡</td>
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<td>L</td>
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<tr>
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<td>F/8</td>
<td>L*</td>
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<tr>
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<td>M/8</td>
<td>R†</td>
<td>4</td>
<td>L</td>
<td>5</td>
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</tr>
<tr>
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<td>F/11</td>
<td>R*</td>
<td>6</td>
<td>R</td>
<td>9</td>
<td>19</td>
<td>14.2</td>
<td>15.6</td>
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</table>

* these cases had both café au lait spots and pseudarthrosis of the ipsilateral fibula  † had postoperative angiogram

Case 3. Pre-operative radiographs show a large tibial defect (a). Seven weeks after operation, early callus formation is visible (b). Nine years after operation, at age 17 years (c).

PATIENTS AND METHODS

Between 1980 and 1981, four girls and one boy with congenital pseudarthrosis of the tibia were treated by vascularised fibular grafts. In all, the diagnosis had been made soon after birth. Four patients had multiple café au lait spots. The age at operation ranged from seven to 11 years (mean, 8.4), and the average follow-up was 9.2 years (8 to 10).

All patients had undergone multiple operations previously, without achieving tibial union; pseudarthrosis of the fibula was also present in four. Most patients had leg length discrepancy, foot atrophy and ankle stiffness prior to vascularised fibular transplantation (Table I).

Operative technique. Before operation, the vascular anatomy was determined by angiography. Dissection of the vascularised fibula was performed by the technique described by Chen et al (1979), except that we retained very little muscle around the fibula. The ipsilateral fibula was used in four patients, and the contralateral fibula in the one patient whose ipsilateral fibula had been harvested subperiosteally at a previous operation. Our first choice was the ipsilateral fibula proximal to the pseudarthrosis site. The thick fibrous tissue around the tibial pseudarthrosis was resected completely but resection of the sclerotic bone ends was minimal. After correction of angular deformity, a slot was created to receive the fibular graft which was secured by several screws. End-to-end anastomosis was performed between the anterior tibial and the peroneal vessels. Bone chips were placed around the pseudarthrosis site using the bone resected to make the slot (Fig. 1). In case 3, there was a large gap after resection of the pseudarthrosis, so the vascularised fibula was interposed between the tibial ends, as described by Pho et al (1985) and shown in Figure 2). The lengths of the fibular grafts ranged from 6 to 9 cm (mean, 7.6).

A long leg plaster cast was applied for about three
Case 2. Pre-operative radiograph showing severe angulation at the pseudarthrosis (a). Ten weeks after operation (b). Ten months after operation, solid union is achieved. Union of the fibular pseudarthrosis has also occurred (c). Radiographs taken at the final follow-up at age 18 years (d).

Case 4. Pre-operative radiograph (a). Radiograph three weeks after operation (b). Radiograph taken at final follow-up at age 18 years (c). At age 12, four years after the operation, a valgus deformity of the ankle on the contralateral side had developed (d) and a corrective osteotomy was necessary.
months and a patellar-tendon-bearing brace was worn thereafter, until solid bone union and recanalisation had occurred.

In three patients, angiography was performed about two months after operation and the patency of the anastomoses was confirmed.

RESULTS

Bone union was eventually achieved in all patients without further surgery. The time required for union ranged from five to nine months. In cases 1 and 4 stress fractures developed after grafting, but these united after a period of splinting. In cases 1 and 2, the fibular pseudarthrosis also united, within nine months, without specific surgical intervention (Fig. 3). In case number 4, in which the healthy contralateral fibula was harvested, a valgus deformity of the ankle developed on that side and corrective osteotomy had to be performed four years later. The ankle below the pseudarthrosis never deformed into valgus (Fig. 4).

Tibial length discrepancy, measured radiographically at follow-up ranged from 0.5 to 15.6 cm (mean, 7.4). This was almost the same as the postoperative discrepancy in three patients, but in cases 3 and 5, in which the distal tibial physis was destroyed by previous operations, the discrepancy had progressively increased.

Stiffness of the ankle and atrophy of the foot were seen in all patients. Foot length discrepancy varied from 1.5 to 5.2 cm (mean, 3.2). In case 5, in which the leg length discrepancy was 16 cm, a long leg brace is still required. In case 3, who had a 10 cm discrepancy, a short leg brace with a built-up heel is being used. The other three patients do not wear a brace. In three patients, an anterior convex deformity of the tibia was noted at final follow-up.

DISCUSSION

The aetiology of congenital pseudarthrosis of the tibia is still unknown. It is frequently associated with neurofibromatosis, but this type of tissue has not been demonstrated histologically in the thick fibrous tissue surrounding the pseudarthrosis (Aegerter 1950; Brown, Osebold and Ponseti 1977). When conventional free bone grafts are used the graft is frequently resorbed, and Boyd (1982) has reported that the fibromatosus tissue around the pseudarthrosis has osteolytic activity, which is most pronounced in the young child and decreases with age. Umber, Moss and Coleman (1982) noted that the older the patient the more likely it was that union would be achieved. However, if union is not achieved early in life, limb length discrepancy and foot deformities develop. Morrissey et al (1979) did not achieve a satisfactory result in any patient in whom union had not occurred by the age of six years.

With vascularised bone grafts resorption does not occur and successful operations can be performed in young children. Unfortunately, every child in this series already had several operations, and leg length discrepancy and foot atrophy had already developed. Had a vascularised graft been performed as the initial operation, better results might have been obtained.

In most previous reports, the contralateral healthy fibula has been used. This can lead to the development of a valgus deformity of the ankle (Hsu et al 1972; Wiltsie 1972) and therefore necessitates distal tibiofibular synostosis (Weiland and Daniel 1980; Gordon et al 1986). Furthermore, many parents are hesitant to consent to the use of the healthy fibula. We believe that the ipsilateral fibula is the best donor site. If a healthy segment proximal to the pseudarthrosis is used, there is no problem, and we were able to achieve solid fusion in all patients. No patient developed a valgus ankle on the affected side, though distal tibiofibular synostosis was not performed. Our experience contrasts with that of Pho et al (1985) who reported a high incidence of valgus deformity of the ankle of the affected leg. This may be explained by the fact that most patients in our series already had a stiff ankle and had worn a protective brace for several years. It may, therefore, be wise to perform distal tibiofibular synostosis of the affected foot at the time of the vascularised bone graft if this is performed as the primary operation.

In most previous reports, the entire sclerotic portion of the tibia has been resected and the vascularised fibula has been inserted into the medullary canal of the tibia proximally and distally (Weiland and Daniel 1980; Pho et al 1985). Dormans et al (1990) have recently reported their results using this technique and, in half the patients, union was delayed at the distal site and an additional bone graft was necessary. Though we remove all the thick, fibrous tissue around the pseudarthrosis, resection of the sclerotic bone ends is sufficient to correct the angular deformity. If union occurs between the two ends of the graft and the decorticated tibia proximally and distally, then the sclerotic bone ends will later unite. In two of four patients with pseudarthrosis of the fibula, this united without a specific surgical intervention after successful tibial union. We conclude that radical resection of the sclerotic bone ends is not necessary.

With conventional bone grafting, recurrent fractures are common after successful union, and repeated bone grafting is often necessary (Boyd and Sage 1958; Van Nes 1966). Two of our patients suffered stress fractures, but these readily united. Union is more solid after vascularised bone grafts than with conventional methods and we recommend the procedure as the primary treatment of choice in congenital pseudarthrosis of the tibia.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.
REFERENCES


