Vascularised bone grafting for fibrous dysplasia of the upper limb

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We describe our experience with vascularised bone grafting for the treatment of fibrous dysplasia of the upper limb in eight patients, five men and three women, aged between 17 and 36 years. The site was in the humerus in six and the radius in two. Persistent pain, progression of the lesion and pathological fracture with delayed union were the indications for surgical intervention.

We used a vascularised fibular graft after curettage of the lesion. Function and radiological progress were serially monitored. Early radiological union of the graft occurred at periods ranging from 8 to 14 weeks. The mean period for reconstitution of the diameter of the bone was 14 months (12 to 18) predominantly through inductive formation of bone around the vascularised graft, which was a prominent feature in all patients. There were no recurrences and none of the grafts sustained a fracture or failed to unite. After operation function was excellent in three patients and good in five. Vascularised bone grafts provide a safe and reliable means of ensuring good continuity of bone with little risk of recurrence and failure.

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Fibrous dysplasia is a relatively common, benign skeletal disorder of adolescents and young adults in which the normal medullary canal is replaced and weakened by fibrous tissue.¹ A single bone may be involved, either

wholly or partially, or several may be affected. Management is designed to prevent pain, limit deformity and treat pathological fractures.

Fractures in bones affected with fibrous dysplasia often do not require operations since they generally heal without difficulty.² Callus formed at the affected site, however, is dysplastic³ and patients are thus prone to repeated fractures and deformity (Fig. 1a). This can be prevented by prolonged immobilisation in a cast but may lead to other problems such as stiffness of the joint and muscle atrophy. Progressive deformity, failure of union and persistent pain are considered to be indications for surgery.⁴ Surgical treatment is challenging. Curettage and cancellous bone grafting have been used to replace the dysplastic fibro-osseous tissue with normal bone, but this is associated with a high rate of recurrence.⁵ Internal splintage with metal implants has been used to prevent deformity but they are prone to fatigue failure. The use of cortical autografts and allografts⁶⁻⁸ has likewise been employed.

In many clinical situations vascularised bone grafts which promote early remodelling and hypertrophy have been used to bridge large skeletal defects.⁹ Their benefit in fibrous dysplasia has been described anecdotally¹⁰ and there have been no reports specifically detailing their use in fibrous dysplasia of the upper limb. We describe eight patients with fibrous dysplasia of the upper limb in whom surgical intervention was necessary because of progressive disease and persistent pain, and to prevent deformity. In each we used a vascularised fibular graft to bridge the bone defect after aggressive curettage of the lesion.

Patients and Methods

There were five men and three women aged between 17 and 36 years (Table I). The humerus was affected in six and the radius in two. Persistent pain was associated with delayed healing of a fracture in five, with recurrence after initial primary treatment by curettage and grafting in two and with formation of a secondary aneurysmal bone cyst in one.

Operative technique. We used an extensile approach to expose the affected segment of bone and performed aggressive curettage using a motorised burr. Curettage was considered to be complete when the medullary cavity was

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Fig. 1

Figure 1a – Anteroposterior radiograph of the left humerus before operation. There is expansion, ground-glass opacification and cortical thinning of the proximal diaphysis, indicative of fibrous dysplasia. An unhealed fracture is present proximally with a rim of adjacent callus and there is a second healed fracture in the mid-portion of the lesion. Figure 1b – An early postoperative radiograph. Only a thin incomplete shell of the original cortex remains. The vascular fibular graft is secured proximally and distally by screws. Figure 1c – Three months after surgery there is good radiological union. Figure 1d – Two years after surgery inductive formation of bone around the graft leads to approximation of the normal calibre. The proximal screws have been removed.

completely clear of dysplastic bone and only a thin shell of cortex remained (Fig. 1b). The length of the affected bone ranged from 11 to 20 cm. In each case, an appropriate length of fibula, removed with the peroneal vessels, was used to bridge the defect. The technique of taking a vascularised fibular graft has been well described.^{11,12} With the patient supine an incision is made over the lateral aspect of the fibula. The muscles of the peroneal and anterior compartment are reflected off the length of the fibula. Distally, the peroneal vessels are exposed after incising and then transecting the interosseous membrane proximal to the tibiofibular syndesmosis. A distal osteotomy of the fibula facilitates its mobilisation along with the peroneal vessels in a proximal direction. The peroneal vessels are dissected proximally to provide a pedicle 5 to 6 cm long and a fibular bone segment is resected which is usually 1 to 2 cm longer than the intended defect of the bone. The additional length of graft is telescoped into the host bone for extra stabilisation, and is secured using either screws or wire loops. To ensure a reliable junction, end-to-side vascular anastomoses of the graft artery are made to the brachial artery in lesions of the humerus and to the radial artery in those of the radius

or ulna. Since the vessels are healthy, mobilisation and anastomosis are easily accomplished without the use of interpositional vein grafts or other techniques. The cephalic vein is used for venous anastomosis to the venae comitantes of the peroneal artery.

In all patients there was excellent flow and active bleeding from the ends of the fibular graft and surface after completion of the vascular suturing. We did not encounter any difficulties in achieving an anastomosis and undertook no specialised investigations to assess vascular patency or the viability of the graft after surgery. Movement of the upper limb was encouraged as soon as postoperative pain allowed. This was strictly supervised on an outpatient basis. Particular emphasis was placed on movements of the shoulder and function of the hand.

Follow-up. The mean follow-up was 5.6 years (4 to 9). Clinical evaluation of the function of the hand and upper limb was carried out jointly by experienced physiotherapists and occupational therapists using a score which integrated elements of pain, stability, deformity and overall functional ability.¹³ Radiography was used to monitor union of the grafts, bone hypertrophy and recurrence.

Table I. Details regarding the location of the lesion, the indication for surgery, and outcome in eight patients treated by vascularised bone grafting

Case	Gender	Age (yr)	Site	Previous treatment	Duration of treatment or interval between previous treatment and vascularised bone grafting in months	Indication for surgery	Follow-up (yr)	Outcome
1	M	17	Metadiaphysis of the humerus	Brace	6	Delayed healing/persistent pain	9	Excellent
2	M	21	Metadiaphysis of the humerus	Curettage and cancellous bone grafting	24	Recurrence/refracture/persistent pain	5	Excellent
3	F	30	Radius (whole)	Brace	4	ABC* formation/persistent pain	4	Excellent
4	F	28	Radius (whole)	Cast	5	Delayed healing/persistent pain	5	Excellent
5	M	36	Metadiaphysis of the humerus	Shoulder immobiliser	3	Delayed healing/persistent pain	6	Excellent
6	M	17	Diaphysis of the humerus	Curettage and bone grafting, plating	24	Delayed healing/persistent pain	4	Excellent
7	F	23	Metaphysis of the humerus	Curettage and bone grafting	24	Recurrence/persistent pain	7	Good
8	M	30	Humerus (whole)	Brace	6	Delayed healing/persistent pain	6	Excellent

* aneurysmal bone cyst

Results

Function was assessed three months after the operation. In three patients it was excellent and in the remaining five good (Table I). All patients regained a complete range of movement of the shoulder and forearm within six weeks.

Early radiological union of the graft occurred at periods ranging from 8 to 14 weeks (Fig. 1c). Inductive formation and consolidation of bone around the vascularised graft were prominent features in all patients (Fig. 1d), and there was some hypertrophy of the graft and remodelling. The mean period for reconstruction of the diameter of the original bone was 14 months (12 to 18) predominantly by inductive formation of bone. There were no recurrences and none of the grafts sustained fractures or failed to unite.

Discussion

Fibrous dysplasia is a benign, pathological condition characterised histologically by poorly orientated osseous trabeculae, weakened by replacement with fibrous tissue. Not all the lesions require surgical intervention, and most of those in the upper limb are amenable to non-surgical management.¹⁴ In our patients clinical and radiological progression despite previous treatment, and anxiety about immobilisation of the upper limb prompted us to use a vascularised bone graft. Surgical intervention ideally should ensure that there is adequate clearance, predictable restoration of bony continuity, maintenance of the function of the limb and a low incidence of recurrence. Persistence of pain, progression of the lesion and failure of union are established indications for surgical intervention.⁴ Although small focal sites may be treated successfully using cancellous bone grafts after curettage, local recurrence remains a problem. Conventional cancellous bone grafts are

dependent on the local healing response for their remodelling. This process of 'creeping substitution'¹⁵ involves resorption of the bone graft and newly-formed host bone. In fibrous dysplasia, cancellous bone grafts undergo resorption and replacement with the same type of poorly-formed woven bone, reminiscent of the underlying pathological process and it is not surprising therefore that curettage and grafting of cancellous bone do not prevent recurrence of the lesion and may lead to fracture and deformity. Even aggressive extraperiosteal resection¹⁶ may not eliminate these problems. Unlike conventional cancellous bone grafts, vascular grafts are unaffected by the dysplastic process. Remodelling of the graft, incorporation and hypertrophy are independent of the local dysplastic tissue and depend on the immediate restoration of the vascular supply after anastomosis.¹⁷ We therefore deliberately preserved an osteoperiosteal tube after curettage within which the vascularised bone graft was placed. The osteoinductive influence of the vascularised fibula within this tube was a notable feature in all our patients, and accounted for most of the bony reconstitution, rather than true hypertrophy of the fibula. It may be argued that an osteoperiosteal tube filled with cancellous grafts may yield similar results, provided that stability is maintained. Vascularised fibular grafting, however, has distinct advantages providing immediate secure splintage and sufficient stability to allow early movement. In our series, healing was rapid and solid bony continuity was established, bypassing the local dysplastic process completely, thus providing immediate and reliable structural support without the need for external stabilisation or immobilisation. Minimal internal fixation was required to secure the ends of the graft. Early mobilisation allowed early restoration of function and provided the physiological stimulus for the osteoinductive and remodelling influence of the graft on surrounding tissues.

We suggest that if surgical treatment is contemplated for fibrous dysplasia of the upper limb, vascularised bone grafts provide a reliable option for bony continuity, preventing recurrence and allowing satisfactory restoration of the function of the limb within a short period of time.

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References

1. **Lichtenstein L, Jaffe HL.** Fibrous dysplasia of bone: a condition affecting one, several or many bones, the graver cases of which may present abnormal pigmentation of skin, premature sexual development, hyperthyroidism or still other extraskkeletal abnormalities. *Arch Pathol* 1942;33:777-816.
2. **Grabias SL, Campbell CJ.** Fibrous dysplasia. *Orthop Clin North Am* 1977;8:771-83.
3. **Enneking WF.** *Clinical musculoskeletal pathology*. Third revised ed. Gainesville: University of Florida Press, 1990:266-7.
4. **Harris WH, Dudley HR Jr, Barry RJ.** The natural history of fibrous dysplasia: an orthopaedic, pathological and roentgenographic study. *J Bone Joint Surg [Am]* 1962;44-A:207-33.
5. **Guille JT, Kumar SJ, MacEwen GD.** Fibrous dysplasia of the proximal part of the femur: long-term results of curettage and bone-grafting and mechanical realignment. *J Bone Joint Surg [Am]* 1998; 80-A:648-58.
6. **Bryant DD IIIrd, Grant RE, Tang D.** Fibular strut grafting for fibrous dysplasia of the femoral neck. *J Natl Med Assoc* 1992; 84:893-7.
7. **Enneking WF, Gearen PF.** Fibrous dysplasia of the femoral neck: treatment by cortical bone-grafting. *J Bone Joint Surg [Am]* 1986; 68-A:1415-22.
8. **Shih HN, Chen YJ, Huang TJ, Hsu KY, Hsu RW.** Treatment of fibrous dysplasia involving the proximal femur. *Orthopedics* 1998; 21:1263-6.
9. **Weiland AJ, Moore JR, Daniel RK.** Vascularized bone autografts: experience with 41 cases. *Clin Orthop* 1983;174:87-95.
10. **Tomczak RL, Johnson RE, Hamilton J.** The treatment of monostotic fibrous dysplasia of the first metatarsal with free vascularized fibular bone graft. *J Foot Ankle Surg* 1993;32:604-10.
11. **Taylor GI, Miller GD, Ham FJ.** The free vascularised bone graft: a clinical extension of microvascular techniques. *Plast Reconstr Surg* 1975;55:533-44.
12. **Gilbert A.** Free vascularized bone grafts. *Int Surg* 1981;66:27-31.
13. **Enneking WF, Dunham W, Gebhardt MC, Malawar M, Pritchard DJ.** A system for the functional evaluation of reconstructive procedures after surgical treatment of tumours of the musculoskeletal system. *Clin Orthop* 1993;286:241-6.
14. **Stephenson RB, London MD, Hankin FM, Kaufer HJ.** Fibrous dysplasia: an analysis of options for treatment. *J Bone Joint Surg [Am]* 1987;69-A:400-9.
15. **Burchardt H, Enneking WF.** Transplantation of bone. *Surg Clin North Am* 1978;58:403-7.
16. **Henry A.** Monostotic fibrous dysplasia. *J Bone Joint Surg [Br]* 1969; 51-B:300-6.
17. **Weiland AJ, Phillips TW, Randolph MA.** Bone grafts: a radiologic, histologic and biomechanical model comparing autografts, allografts, and free vascularized bone grafts. *Plast Reconstr Surg* 1984; 74:368-79.