We present a retrospective study of patients suffering from a variety of benign tumours in whom external fixators were used to treat deformity and limb-length discrepancy, and for the reconstruction of bone defects. A total of 43 limbs in 31 patients (12 male and 19 female) with a mean age of 14 years (2 to 54) were treated.

The diagnosis was Ollier’s disease in 12 limbs, fibrous dysplasia in 11, osteochondroma in eight, giant cell tumour in five, osteofibrous dysplasia in five and non-ossifying fibroma in two. The lesions were treated in the tibia in 19 limbs, in the femur in 16, and in the forearm in eight. The Ilizarov frame was used in 25 limbs, the Taylor Spatial Frame in seven, the Orthofix fixator in six, the Monotube in four and the Heidelberg fixator in one. The mean follow-up was 72 months (22 to 221).

The mean external fixation period was 168 days (71 to 352). The mean external fixation index was 42 days/cm (22.2 to 102.0) in the 22 patients who required limb lengthening. The mean correction angle for those with angular deformity was 23˚ (7˚ to 45˚).

At final follow-up all patients had returned to normal activities. Four patients required a second operation for recurrent deformity of further limb lengthening. Local recurrence occurred in one patient, requiring further surgery.

The management of limb deformity, shortening and bone defects in the treatment of benign tumours is a major challenge. The conventional methods of correcting deformity and limb-length inequality that were used before the development of external fixation and distraction osteogenesis, such as shortening, single or multiple osteotomy or epiphysiodesis to slow the growth of the longer limb, were limited in their indications, unpredictable, and often unsatisfactory.

The methods of treating bone defects include free autograft, vascularised bone graft, allograft, artificial bone substitutes and prostheses. However, these methods have several disadvantages and a high incidence of complication. Long-term results are often unsatisfactory, especially after resection of extensive or juxta-articular tumours. Distraction osteogenesis was introduced by Ilizarov, and has been widely used to treat traumatic bone loss, nonunion, osteomyelitis, malunion and limb-length discrepancy, and to correct deformity. It has the advantages of biomechanical stability, a bloodless technique, regeneration of new bone, and gradual lengthening of the soft tissues. However, there are few studies of its use in the treatment of benign bone tumours. In this study we describe our experience of the use of external fixators to correct deformity, for limb lengthening and for the reconstruction of bone defects in the management of benign bone tumours.

**Patients and Methods**

External fixation techniques were used in 43 limbs in 31 patients, 12 male and 19 female. Their mean age at surgery was 14 years (2 to 54). After physical examination, the affected limb was examined by plain radiography, CT, and MRI as necessary.

The histological diagnosis was Ollier’s disease in 12 limbs (four patients), fibrous dysplasia in 11 (eight patients), osteochondroma or osteochondromatosis of the forearm in eight (eight patients), giant cell tumour (GCT) in five (five patients), osteofibrous dysplasia in five (four patients) and non-ossifying fibroma in two (two patients). An Ilizarov fixator (Smith and Nephew plc, Memphis, Tennessee) was used in 25 limbs, a Taylor spatial frame (Smith and Nephew plc) in seven, an Orthofix (Orthofix Inc., Verona, Italy) in six, a Monotube (Stryker Osteosynthesis, Kiel, Germany) in four and a Heidelberg external fixator (Smith and Nephew plc) in one. Prophylactic anti-
bacteriostats were given to all patients for three days post-operatively. Distraction at the osteotomy site was started seven days post-operatively at a rate of 0.25 mm every six hours in those patients who needed lengthening, with radiographs every two weeks. A rehabilitation programme of muscle and joint exercises was begun immediately after surgery.

The patients were divided into six groups according to the histological diagnosis (Table I). Those in the first group had Ollier’s disease in 12 limbs (six femora and six tibiae). The problems in this group were leg-length discrepancy, deformity and pathological fracture. Treatment involved limb lengthening and correction of deformity (Fig. 1), and fixation of pathological fractures followed by lengthening and correction of deformity. We used an Ilizarov frame in eight limbs, an Orthofix in two, a Taylor spatial frame in one and a Heidelberg fixator in one. The second group had fibrous dysplasia with lesions in 11 limbs (eight femora and three tibiae); the clinical problems were pathological fracture and deformity. We used an Ilizarov frame in eight limbs, an Orthofix in two, a Taylor spatial frame in one and an Orthofix in two. A single or double level osteotomy was used depending on the degree of deformity. The third group consisted of osteochondroma or multiple osteochondromata of the forearm in eight limbs. The clinical problems were deformity and shortening of the affected bone (Fig. 3). A Monotube (Stryker Osteosynthesis) was used in four limbs, an Orthofix in two, and an Ilizarov in two. The fourth group included subarticular GCT of the proximal tibia with a large soft-tissue extension in five limbs. Partial en bloc resection combined with curettage at the subarticular area was undertaken. The tumour bed was treated with phenol and ethanol three times. The subarticular defect was then immediately reconstructed using a cylinder of bone from the diaphysis and iliac bone graft followed by bone transport to fill the newly-created diaphyseal defect using an Ilizarov frame (Fig. 4). The fifth group included five limbs with osteofibrous dysplasia of the tibia. After en bloc marginal resection, bone transport was performed to reconstruct the defect and correct the deformity when necessary (Fig. 5). The last group consisted of two limbs with non-ossifying fibroma of the distal femur, which was treated by curettage, bone grafting and correction of deformity using the Ilizarov fixator (Fig. 6). In one patient, the deformity was corrected at the site of pathological fracture.

Correction of deformity was performed in eight limbs (one in the Ollier’s disease group, five in the fibrous dysplasia group, two in the non-ossifying fibroma group), limb lengthening in five (three in the Ollier’s disease group and two in the forearm exostosis group). Limb lengthening was combined with correction of deformity in 20 (eight in the Ollier’s disease group, six in the fibrous dysplasia group, six in the forearm exostosis group), and bone transport in ten (five in the GCT group, five in the osteofibrous dysplasia group). The mean follow-up was 72 months (22 to 221). The affected sites were 19 tibiae, 16 femora and eight forearms. The out-

<table>
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<th>Table I. Patient demographics</th>
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<tr>
<td><strong>Number of</strong>&lt;br&gt;<strong>limbs</strong></td>
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<tr>
<td>Ollier’s disease</td>
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<td>Fibrous dysplasia</td>
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<td>Multiple exostosis</td>
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<td>Osteofibrous dysplasia</td>
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<td>Non-ossifying fibroma</td>
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* TSF, Taylor spatial frame<br>† HDB, Heidelberg fixator
comes were evaluated using the external fixation time and index, obtained by dividing the external fixation time by the length distracted and limb function.

Results

The mean external fixation time was 168 days (71 to 352); 175 in the Ollier’s disease group (102 to 352), 121 days in the fibrous dysplasia group (70 to 155), 131 days in the forearm exostosis group (84 to 155), 234 days in the GCT group (161 to 284), 256 days in the osteofibrous dysplasia group (74 to 349), and 114 days in the non-ossifying fibroma group (102 to 126). The mean external fixation index was 42 days/cm (22.2 to 102.0) in 35 limbs subjected to distraction osteogenesis. It was 42.4 days/cm (22.2 to 80.0) in the Ollier’s disease group, 51.1 days/cm (34.8 to 69.0) in the fibrous dysplasia group, 53.9 days/cm (31.9 to 102.0) in the forearm exostosis group, 40.8 days/cm (35.8 to 43.7) in the GCT group, and 38.7 days/cm (24.5 to 63.5) in the osteofibrous dysplasia group.

The mean length of distraction was 4.3 cm (1.5 to 7.2). The mean correction angle in the 28 limbs who underwent deformity correction was 23˚ (7˚ to 45˚). It was 24.8˚ (20˚ to 31˚) in the Ollier’s disease group, 25.8˚ (11˚ to 45˚) in the fibrous dysplasia group, 16.1˚ (7˚ to 25˚) in the forearm exostosis group, and 7˚ (7˚ to 7˚) in the non-ossifying fibroma group.

All 31 patients had returned to normal daily activities without pain by the final follow-up. There were ten complications. The most common was pin track infection in six patients (19.4%), which was treated successfully with oral antibiotics. Only one patient (3.2%) needed intravenous antibiotics. Two patients (6.5%) developed an equinus deformity that was treated successfully with physiotherapy after removal of the fixator. Early consolidation of the regenerate bone occurred in two patients (6.5%) requiring a repeat subcutaneous osteotomy. A second operation for limb lengthening or correction of deformity was required in three patients with Ollier’s disease and one with an osteochondroma of the forearm. Local tumour recurrence occurred in one patient with GCT. This was treated by curettage and phenol and ethanol followed by iliac bone grafting.
Discussion

A number of surgical treatments have been reported for correction of deformity, limb equalisation and reconstruction in patients with bone tumours. In this series of patients with benign bone tumours we were able to solve most of the problems using external fixators.

Multiple enchondromatosis (Ollier’s disease) is a common intraosseous benign cartilaginous tumour that develops in close proximity to the growth plate. It can cause deformity and limb-length discrepancy, and has a potential risk of malignant change to chondrosarcoma. Conventional treatment is curettage and bone grafting, which may result in severe deformities requiring repeated osteotomy. It is often difficult to obtain stabilisation and normal bone growth by autogenous bone grafting. Jesus-Garcia et al described the use of the Ilizarov technique in ten patients with Ollier’s disease. They reported excellent results and claimed that the technique led to conversion of the abnormal cartilage to histologically mature bone in all their patients. However, at the final follow-up in our series, the regenerate was usually (ten of 12 segments) partially replaced by enchondromatous tissue. Patients with Ollier’s disease may require limb lengthening and correction of deformity at a young age because of progressive deformity. As a result, a second operation was necessary in three of four patients in this series.

In fibrous dysplasia, curettage of the lesion and bone grafting may be effective for monostotic lesions but not for polyostotic fibrous dysplasia. Guille Kumar and MacEwen reported that none of the lesions in their series was eradicated or diminished in size, and that all bone grafts were absorbed. Curettage and bone grafting is not suitable in patients with deformity and pathological fracture. Corrective osteotomy with plate-and-screw fixation is a relatively simple method but it is difficult to achieve sufficient fixation with screws in weakened bone, and fracture may occur because of stress shielding at the distal end of the plate. It may be difficult to obtain satisfactory limb alignment and to fit the plate to the contour of the deformed bone. The other method that can be used, particularly for a shepherd’s crook’s deformity of the femur, is multiple...
osteotomies and Zickel nail fixation. Freeman et al\textsuperscript{16} recommended this technique because it provided intramedullary stabilisation, reduced the stress concentration distally and, when combined with osteotomy, can correct deformity. However, Guille et al\textsuperscript{19} reported a patient with fibrous dysplasia treated with a Zickel nail who at long-term follow-up demonstrated thinning of the medial cortex, probably caused by stress shielding resulting from the intramedullary nail. In the treatment of fibrous dysplasia in our series we used either single or double osteotomies depending on the axis of the deformity, followed by gradual correction with external fixation. Correct alignment of the mechanical axis of the lower limb following correction was maintained in all patients and was effective in preventing recurrent deformity and fracture.

Previously recommended methods of treatment of osteochondroma of the forearm include excision alone, excision with radial shortening and resection of the displaced radial head, and excision with ulnar lengthening and radial hemiepiphyseal stapling.\textsuperscript{21,22} Excision of the exostosis has not been effective in controlling the progression of deformity, nor has resection of the head of radius proved satisfactory.\textsuperscript{23} Radial hemiepiphyseal stapling, used alone or with ulnar lengthening, has been effective but causes unacceptable shortening of the forearm and the final result is unpredictable.\textsuperscript{24} In this series, the main problems included ulnar deviation of the hand, dislocation of the radial head and deformity of the radius. We undertook resection of the lesion, ulnar lengthening using an external fixator, and corrective osteotomy of the radius with plate-and-screw fixation, with satisfactory results. One of the eight patients needed a second operation for recurrent ulnar deformity and shortening.

A number of treatments have been advocated for GCT of bone, including curettage and grafting with autogenous bone graft, allograft or synthetic bone substitutes, either graft alone or combined with adjuvant therapy, such as cryotherapy or the application of phenol after curettage.\textsuperscript{25-29} Autograft may be used successfully for a small defect, but for managing a large defect it has many disadvantages, such as donor site morbidity, lack of sufficient graft and the high rate of graft fracture in a long defect.\textsuperscript{20} Vascularised bone graft techniques also have some disadvantages, such as the sacrifice of normal tissue, technical difficulty, prolonged surgery, and a longer period to achieve hypertrophic change.\textsuperscript{31} Allograft has many disadvantages, such as transmission of disease, rejection, late fracture, a high infection rate, bone resorption, and a long period of immobilisation.\textsuperscript{32} Synthetic bone substitutes such as hydroxyapatite (HA) may be used in small defects, but because of the extended time necessary to achieve bony union, HA should not be used as a grafting material for extensive bone defects or for major weight-bearing areas.\textsuperscript{33,34} Treatment of GCT with curettage, cauteryisation and methylmethacrylate is now widely accepted in most orthopaedic oncology centres, but has disadvantages, such as the thermal effect on articular cartilage, degenerative arthritis and the fact that methylmethacrylate is not biological.\textsuperscript{35} Persson et al\textsuperscript{36} reported good results using this technique with only two recurrences in 50 patients. However, Wada et al\textsuperscript{37} reported that all their patients developed a radiolucent zone up to 2.5 mm wide at
the bone-cement interface during the first six months as a result of thermal necrosis following aggressive curettage and acrylic cement reconstruction for GCT. Mjøberg et al.38 also reported a radiolucent zone in six patients. However, because of the high rate of recurrence reported in the literature after treatment of GCT, it is likely that it is the adequacy of tumour resection rather than the use of adjuvant therapy that determines the risk of recurrence.39 The use of external fixation and distraction osteogenesis allowed us to successfully treat GCTs of the proximal tibia with an associated extensive soft-tissue mass. We were able to reconstruct a large subarticular defect after tumour resection with normal regenerate bone using bone transport, and to preserve normal function of the knee joint. No patient has so far developed osteoarthritic changes in the knee.

Osteofibrous dysplasia is a rare tumour-like condition that almost exclusively affects the tibia in children with a possible link to adamantinoma.40 Its management depends on the age of the patient, the size of the lesion and the severity of the symptoms. Small lesions with no bowing of the tibia or recurrent fracture should be treated conservatively.41 However, patients with repeated fractures, recurrence, deformity or pain should be treated surgically. Treatment by curettage and bone graft has been associated with a high rate of recurrence, especially in young children.42 Kempson43 proposed en bloc excision for osteofibrous dysplasia because of its close relationship to adamantinoma. In this series, we successfully treated five limbs with symptomatic osteofibrous dysplasia by en bloc marginal excision and bone transport using an Ilizarov fixator.

Non-ossifying fibroma produces eccentric osteolytic lesions in the diaphyseal-metaphyseal junction of long bones, commonly the femur and the tibia. It most commonly affects children.44 Most non-ossifying fibromas heal without treatment, but some large lesions cause persistent pain, deformity or pathological fracture. Treatment by simple curettage and bone grafting has been associated with a high rate of recurrence, a limited supply of transplantable tissue in children, and morbidity at the donor site.45 In the two patients in our series, there has so far been no tumour recurrence after thorough curettage and correction of the deformity by osteotomy and external fixator.

In conclusion, we believe that external fixation is an effective technique for treating deformity, limb-length discrepancy and bone defects produced by benign bone tumours. It offers a good alternative to other conventional methods of management. There are disadvantages to the technique, such as pin track infection, the bulk of the frame, the long treatment period and the need for repeated adjustment of the frame. The Ilizarov fixator or Taylor spatial frame are more suitable than unilateral fixators for treating deformity and limb-length discrepancy simultaneously.

References

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