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A novel approach to juxta-articular aggressive and recurrent giant cell tumours: resection arthrodesis using bone transport over an intramedullary nail

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Abstract Aggressive juxta-articular giant cell tumours of the lower limbs occurring in young patients are a challenge to the average orthopaedic surgeon. Although it is the treatment of choice for these tumours, wide resection creates a problem for the reconstruction of large bone gaps. We describe our results after resection arthrodesis of such tumours using the technique of bone transport over a long intramedullary nail in 27 patients. This is the first and largest study of its kind in the management of giant cell tumours in the literature. All our patients fared well with this mode of treatment, and none had recurrence or major complications.

Résumé Les tumeurs à cellules géantes juxta articulaires des membres inférieurs surviennent chez de jeunes patients et leur traitement est difficile. Une résection large est le traitement de choix de ces tumeurs. Cette résection pose le problème de la reconstruction osseuse. Nous décrivons nos résultats après résection arthrodesis de telles tumeurs en utilisant la technique du transport osseux le long d'un clou centro médullaire, ce traitement a été utilisé chez 27 patients. Il s'agit de la première étude de ce type. Ces patients ont guéri avec ce traitement. Il n'y a pas eu de récurrence de cette tumeur ni de complications majeures.

Introduction

Giant cell tumour of the bone (GCT) is one of the most common primary bone tumours in young adults [3, 19, 27]. Local extensions, regional and systemic tumour implantation and malignant transformation with widespread metastases are among the reported manifestations of these neoplasms that are indicative of their aggressive and unpredictable nature [25, 33]. The ideal form of treatment

for lesions that arise near major joints remains unclear. Although extended curettage with adjuvant and local reconstruction is the standard treatment of uncomplicated giant cell tumours, it carries high recurrence rates in aggressive or recurrent GCT. There has been a great deal of development in the surgical management of the aggressive, recurrent or peri-articular giant cell tumour towards limb salvage. In these situations, wide resection of the tumour has been accepted as the standard treatment. But it has always been a challenge to reconstruct the resected gap, especially across joints [16].

This challenge to provide long-lasting survival and function of the limb after reconstruction is now being met with biological solutions using living bone. The ideal reconstruction should have biological affinity, resistance to infection, sufficient biomechanical strength and durability. Since 1996 we have been using distraction osteogenesis (callotasis) over an intramedullary nail, which can regenerate bone of sufficient strength for reconstruction. This is the largest study in the literature in which the patients have undergone bone transport over a nail for resection arthrodesis in giant cell tumour surgery. We report our findings.

Materials and methods

Twenty-seven patients with established diagnoses of giant cell tumour around the knee and ankle from 1996–2002 who underwent resection arthrodesis using bone transport with Ilizarov fixator over a small diameter interlocked intramedullary nail were included in the study (Table 1). The total number of giant cell tumours treated during this period was 138, and nearly 20% of the patients with giant cell tumours had resection arthrodesis. There were 19 males and 8 females. The mean operative age of the patients was 35.5 ± 7.1 years (24–52 years). Twelve lesions were in the tibia and 15 in the femur. Twenty-two lesions were around the knee joint and five around the ankle. Eleven patients underwent this procedure primarily for stage 3 aggressive tumours after an incision biopsy or FNAC

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(fine-needle aspiration cytology), seven following pathologically displaced intra-articular fracture, seven for recurrence after primary surgery and two for soft tissue extension. Of the seven patients with recurrence of the tumour, five had undergone more than one operation before presentation, and the details of the treatment were not known. Two patients treated primarily at our institution had aggressive lesions recurring within 6 months of operation. Preoperatively, all the patients had magnetic resonance imaging of the lesion as a part of the surgical staging (Fig. 1). All the patients who underwent the resection arthrodesis had stage 3 tumours.

Operative technique

The primary procedure was directed towards *en-block* resection of the tumour and stabilisation of the limb together with maintenance of limb length (1–2 cm shorter than the normal limb in cases of knee arthrodesis) using a small-diameter interlocking intramedullary nail crossing the joint (Fig. 2). The fibula was resected primarily in a few of our initial cases in order to facilitate smooth bone transport. Later on, this was substituted by the dislocation of the superior tibio-fibular joint. In the second sitting, once the wound had healed, a simple two-ring Ilizarov apparatus was applied along with single- or two-level percutaneous corticotomy (Fig. 3). Unifocal or bifocal distraction over the nail was begun approximately 7 to 10 days after the second operation at a rate of 0.5 mm twice daily or 0.25 mm four times daily at each of the distraction sites. In case of bifocal distraction, the bone transport was attempted at the rate of 2 mm per day by moving both the rings by 1 mm towards each other. The distraction at the corticotomy site was confirmed using radiographs after 5 days of distraction. This was then continued until the docking of both of the fragments (Fig. 4). The patients were allowed toe-touch weight-bearing crutch walking as soon

as the pain subsided. The patients were discharged once they could manage distraction by themselves. Patients were followed up every 4 weeks with radiographs to assess the bone transport and quality of the regenerate. Any complications encountered were identified at the earliest stage and treated aggressively.

When callus formation was poor, distraction was delayed or compression and distraction of a moving segment (the accordion manoeuvre) was applied. Colour Doppler study was performed at regular intervals to monitor the blood flow in the regenerate in seven cases where this was doubtful. Colour Doppler sonography seems to have the capability to predict whether the development of fracture callus will be normal or delayed [4]. When the regenerate was consolidating prematurely, the distraction rate was increased to 1.5 mm per day.

Once the bone fragments were docked, the Ilizarov fixator was removed. Plate osteosynthesis along with autologous ipsilateral iliac crest cancellous bone grafting was performed at the docking site. Full weight bearing was allowed only after the docking site had united in at least three of the four cortices with trabeculae crossing the bone ends.

Three indices were used to evaluate the results: an **external fixation index** obtained by dividing the entire duration of external fixation by the length of the bone regeneration. The **distraction index** was obtained by dividing the duration of distraction by the length of the bone regeneration. All of our patients had the **maturation index** of zero as a result of fixator removal and plate osteosynthesis of transported bone segments once they were docked. The function of the affected limb was assessed at the final follow-up according to Enneking [9].

Results

The mean length of the bone defect was 11.65 ± 3.2 cm (range 6.4–18.6). The mean external fixation index was 8.1 ± 1.71 days/cm (range 6.1–11.9), and the distraction index was 8.59 ± 1.76 days/cm (range 6.3–12.3). Twenty-

Fig. 1 Preoperative anteroposterior and lateral view radiograph of giant cell tumour of the lower end of the femur with intra-articular pathological fracture

Fig. 2 Wide resection of the aggressive GCT after FNAC and stabilisation with a long, small diameter interlocking intramedullary nail

Table 1 Details of the 27 patients with juxta-articular aggressive giant cell tumours treated by resection arthrodesis with bone transport over an intramedullary nail

Case	Age	Sex	Site	Indication for arthrodesis	Length of distraction (mm)	External fixator index (days/cm)	Distraction index (days/cm)	Distraction	Complications	Last follow-up (months)	Function
1	44	F	Femur	Stage 3	130	6.4	7.6	Bifocal		84	Excellent
2	24	M	Upper tibia	Fracture, stage 3	112	8.3	8.7	Bifocal		48	Good
3	28	M	Femur	Recurrence, stage 3	146	6.8	7.1	Bifocal	Skin invagination	86	Excellent
4	27	F	Upper tibia	Stage 3	86	7.3	8.1	Bifocal	Pes equinus	94	Fair
5	24	M	Lower tibia	Stage 3	94	10.2	10.5	Unifocal		58	Excellent
6	36	F	Femur	Fracture, stage 3	126	7.3	7.9	Bifocal		78	Excellent
7	38	M	Femur	Recurrence, stage 3	148	7.6	8.3	Bifocal		56	Good
8	33	F	Femur	Stage 3	124	7.9	8.3	Bifocal		70	Good
9	36	M	Femur	Fracture, stage 3	186	6.3	6.7	Bifocal		84	Fair
10	43	M	Lower tibia	Stage 3	118	11.4	12.3	Unifocal		88	Good
11	29	M	Upper tibia	Fracture, stage 3	114	6.1	6.3	Bifocal		65	Good
12	39	M	Femur	Recurrence, stage 3	136	8.1	8.5	Bifocal		71	Excellent
13	33	F	Femur	Soft tissue extension, stage 3	164	7.2	7.4	Bifocal	Deep infection	74	Fair
14	32	M	Upper tibia	Recurrence, stage 3	136	7.7	7.8	Bifocal	Pes equinus	65	Excellent
15	29	M	Femur	Stage 3	98	6.9	7.3	Bifocal		66	Good
16	42	F	Lower tibia	Fracture, stage 3	64	11.9	12.3	Unifocal		69	Good
17	48	M	Upper tibia	Fracture, stage 3	78	8.4	8.8	Bifocal		58	Excellent
18	52	M	Femur	Stage 3	94	9.8	10.3	Bifocal	Subluxation of head of fibula with transient foot drop	54	Fair
19	36	F	Lower tibia	Recurrence, stage 3	66	11.4	11.9	Unifocal		59	Excellent
20	37	F	Femur	Fracture, stage 3	154	7.3	7.7	Bifocal	Skin invagination	55	Fair
21	37	M	Femur	Soft tissue extension, stage 3	146	6.7	6.9	Bifocal		51	Excellent
22	38	M	Upper tibia	Stage 3	106	6.9	7.5	Bifocal		53	Excellent
23	29	M	Lower tibia	Recurrence, stage 3	88	11.3	11.7	Unifocal		49	Good
24	26	M	Femur	Stage 3	84	7.8	8.1	Bifocal		49	Excellent
25	41	M	Upper tibia	Stage 3	77	6.7	6.9	Bifocal	Premature consolidation	50	Fair
26	38	M	Femur	Stage 3	110	7.5	7.9	Bifocal		53	Good
27	39	M	Femur	Recurrence, stage 3	159	8.7	9.2	Bifocal		55	Excellent

Fig. 3 Double corticotomy and two-ring Ilizarov fixator application at the second stage surgery

Fig. 4 Third stage surgery after docking with plate osteosynthesis and bone grafting

two patients with tumour resection around the knee had bifocal distraction while the five with ankle arthrodesis had unifocal distraction. The average delay before toe touch crutch walking was 5.4 days (range 4–7), 5.3 months (range 4.8–6.4) before partial weight bearing with a single crutch and 9.8 months (range 8.6–10.4) for unaided full

weight bearing. The mean period of follow-up for the patients was 64.5 ± 13.73 months (range 48–94). The function of the affected limb was rated excellent in 12 patients, good in 9 and fair in 6 as per the Enneking criteria. No local recurrence of tumour was seen. All patients are currently free from disease, and none have had metastases (Fig. 5).

Seven complications occurred in six patients, including two skin invaginations, two mild pes equinus, one deep infection, one premature consolidation, and one subluxation of the head of the fibula with transient foot drop. Skin invaginations were treated by resection of elongated and infolded skin and suturing the edges at the time of docking. Pes equinus was managed by physiotherapy after removal of the Ilizarov apparatus. The only case of deep sepsis developed from a superficial wire infection when the bone transport was complete. It was treated by removal of the Ilizarov, irrigation, and the administration of intravenous antibiotics. The case of premature consolidation of regenerated bone was managed by a further percutaneous osteotomy, resulting in excellent bone regeneration. The patient with foot drop along with subluxation of the head of the fibula was managed by stopping the distraction for 3 weeks while the neuropraxia recovered. There were no fatigue fractures, and none of the patients required intramedullary nail removal.

Discussion

The aim of treatment in giant cell tumours is to preserve the joint and only in a few of the selected cases was sacrificing the joint necessary. The survival rates after the wide resection of these locally aggressive tumours are very encouraging. The morbidity and mortality after successful surgical wide resection of these tumours are negligible. The options for reconstruction after resection of such an

Fig. 5 Five-year follow-up radiographs of the same patient with solid healthy bone

aggressive or recurrent bone tumour about the knee and ankle include the use of a custom tumour endoprosthesis, an osteoarticular allograft or an allograft-endoprosthesis composite, or an arthrodesis with intercalary bone grafting.

Endoprosthetic replacement after resection of the tumour provides an early improvement in function [20]. However, the use of custom-made constrained tumour endoprostheses in this situation is limited by the exorbitant cost and high functional demands of the patients. Allograft-endoprosthesis composites are an alternative to endoprosthetic reconstruction. But the high incidence of complications such as fracture, deformity and infection, makes the outcome unpredictable [5, 12, 15].

For selected patients, an arthrodesis is the better option because of their age, body weight and desired level of activity. An arthrodesis offers the potential advantages of being a stable, durable and predictable reconstruction that enables unrestricted activity, but at the expense of losing the knee motion [1, 8].

Several authors have reported on the technique of resection arthrodesis using bone grafts. The defect has been reconstructed with the use of various combinations of autogenous grafts from the fibula, tibia or iliac crest [10]. In these studies, a high rate of non-oncological complications (40%) like non-union, fatigue fracture of the graft, loss of fixation and inadequate soft-tissue coverage have been reported. These problems, along with the limited availability of autogenous bone, have led to experimental investigations into the use of allografts for arthrodesis either alone or in combination with autogenous bone grafts [2, 11, 21–23, 28]. The use of allograft arthrodesis also has its own share of complications [7]. The results of vascularised bone transfer are more favourable, but there are limitations in biomechanical strength, length and availability [14, 32].

The Ilizarov method is widely used in tibial fracture, non-union, traumatic bone defects, osteomyelitis and deformity, but there are very few reports of its use in the management of bone tumours [6, 13, 17, 18]. The living bone regenerated in distraction osteogenesis will eventually provide sufficient biomechanical strength, length and durability [24, 26, 30, 31]. Merle d'Aubigne was the first to use an intramedullary nail in the resection arthrodesis of the knee [29]. The use of the intramedullary nail has been described in a variety of disorders requiring arthrodesis, but the combination of the nail and Ilizarov has not been described in the literature.

Tsuchiya et al. in 1996 reported their results after Ilizarov distraction osteogenesis of sub-articular bone gap following resection of giant cell tumours in the proximal tibia in five patients [31]. They also performed bone grafting at the docking site soon after positioning the bone fragments. The mean length of the bone gap in their patients was 5.7 cm and the mean duration of external fixation was 233 days. The same workers in 1997 reported their results of limb salvage after distraction osteogenesis in 19 patients, of whom 10 had bone transport, 3 had shortening distraction and 6 had distraction osteogenesis over an intramedullary nail [30]. The mean length of the

defect was 8.4 cm and the mean external fixator index was significantly lower in the distraction histogenesis than that of the intramedullary nail group of patients. Only five of their patients in the bone transport group had giant cell tumours.

We have used distraction osteogenesis over a nail across the joints to achieve arthrodesis to reconstruct defects in the metaphysis and epiphysis after wide excision of giant cell tumours. The addition of an interlocking intramedullary nail is beneficial as a temporary measure to maintain the limb length. One to two centimetres of shortening was planned for all the knee arthrodesis patients in order to facilitate ground clearance while walking. The thin, stress-resistant intramedullary nail is appropriate for combined use to preserve the intramedullary blood supply and allow space for the insertion of wires. The interlocked intramedullary nail affords better axial and rotational control of the bone fragments during bone transport, even with the use of two Ilizarov rings. None of our patients had malalignment.

With the use of bifocal distraction aiming to achieve trifocal osteosynthesis in resection arthrodesis around the knee, the transport rate nearly doubled. It was interesting to note that in this situation, although the speed of distraction was the same at both of the corticotomy sites, the shorter fragment moved faster than the longer fragment. This could be explained partly by virtue of the soft tissue attachment to the larger bony component, which hinders distraction. Immediately after distraction was completed, the fixator was removed, thus reducing the fixator time drastically. Keeping the number of rings to a minimum (two) and the distraction time and fixator time to the least possible, it was encouraging to see that only one of our patients had significant pin tract infection.

Plate osteosynthesis with cancellous bone grafting ensures faster and definite union at the docking site. This allows Ilizarov removal even before the regenerate has consolidated. The consolidation of the regenerate usually takes twice the time of distraction. The ipsilateral iliac crest bone graft was used in all patients with minimal or no donor site morbidity. None of our patients had non-union at the docking site after plating and bone grafting. After the plate osteosynthesis, long before consolidation of the regenerate, the patient was allowed gradual progression to partial weight-bearing walking with a single crutch. This early guarded weight bearing probably helps faster consolidation of the regenerate.

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