Rotationplasty type BI *versus* type BIIIa in children under the age of ten years

SHOULD THE KNEE BE PRESERVED?

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Type BI rotationplasty is currently indicated for children with tumours of the proximal femur whereas type BIIIa rotationplasty is reserved for those in which the entire femur has to be removed. Our aim was to compare these two types of rotationplasty and determine whether the knee should be preserved in children with tumours of the proximal femur. We compared the post-operative complications, oncological outcome, range of movement, Enneking score and radiographs of six children, who had undergone type BI rotationplasty with those of 12 who had undergone type BIIIa rotationplasty.

Patients with type BI rotationplasty had a mean Enneking score of 21.6 compared with 24.4 in those with type BIIIa rotationplasty, and worse mean results in all of the parameters investigated. We conclude that type BI rotationplasty has a worse functional outcome and more complications than type BIIIa rotationplasty in children under the age of ten years.

Rotationplasty can be used as an alternative to amputation in patients with malignant bone tumours of the proximal, distal or entire femur and proximal tibia.^{1,2} A further indication is malignant bone tumours in children under ten years of age as an alternative to an extendible endoprosthesis.³ It can also be performed as a second-line surgical procedure after failure, e.g. because of infection, of a limb-salvage procedure.^{4,6}

Rotationplasty was first performed by Borggreve in 1927 on a patient with a fused knee and consequent limb-length inequality secondary to tuberculosis.^{4,7} In 1950, Van Nes⁸ used rotationplasty in patients with a congenital defect of the femur. Its application in the treatment of malignant bone tumours, however, was first described by Kristen, Knahr and Salzer in 1975⁹ and Salzer et al in 1981¹⁰ who used it as an alternative to above-knee amputation in a patient with osteosarcoma of the distal femur.

In 1986 a modification of the procedure by Winkelmann^{1,3} allowed it to be used in patients with malignant tumours of the proximal femur with or without involvement of the hip as well as of the lower pelvis.

A further modification by Winkelmann described its use in patients with skip metastases or extensive femoral tumours, in which the whole femur had to be removed.^{2,3,6,11,12} He classified rotationplasty into two main types as follows: type A, knee rotationplasties and

type B, hip rotationplasties.^{3,13} Type A is subclassified into type AI for tumours of the distal femur and type AII for those of the proximal tibia. Type B is subclassified into types BI, BII and BIII. Type BI is indicated for malignant tumours of the proximal femur without involvement of the hip and involves replacement of the latter by the ipsilateral knee, which is rotated through 180° and fixed to the pelvis where it functions as a hinge joint. Type BII hip rotationplasty is indicated for malignant tumours of the proximal femur with involvement of the lower pelvis, in which a lower internal hemipelvectomy is required. Finally, type BIII is performed in tumours which require complete resection of the femur and is further subdivided into type BIIIa for children and type BIIIb for adults. In type BIIIa, the hip is reconstructed by placing the rotated, lateral tibial condyle into the acetabulum after disarticulation of the knee and complete removal of the affected femur. In children, the lateral tibial condyle progressively remodels to resemble a new 'femoral head'. In adults, such remodelling does not occur and therefore an arthroplasty is used, hence type BIIIb rotationplasty.

We describe the long-term results of type BI and type BIIIa rotationplasty in children up to ten years of age. The main aim was to compare these two types of rotationplasty and determine which was the more appropriate for children and whether preservation of the knee was worthwhile.

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Fig. 1a

Fig. 1b

Case 16. Anteroposterior (AP) radiographs of a type BIIIa rotationplasty in a 10.6-year-old boy a) seven months and b) four years and seven months after operation. The entire proximal tibia has remodelled to assume the shape of a normal proximal femur with a more favourable distribution of load and an improved weight-bearing capacity. Tibial remodelling is impressive in this patient, considering that he was already 10.6 years old at operation.

Patients and Methods

Between August 1984 and December 2001, 18 patients, eight girls and ten boys, underwent rotationplasty for malignant femoral tumours (Table I). The normal upper age limit for type BIIIa rotationplasty is ten years, but we included one patient (case 16) who had type BIIIa rotationplasty at the age of 10.6 years (Fig. 1).

Six patients were treated by type BI rotationplasty combined with pre- and post-operative chemotherapy. Their mean age at operation was 7 years and 6 months (4.9 to 9.2) and the mean follow-up was 6 years and 5 months (2.8 to 12.4). One of these patients had an osteosarcoma (stage

III), with pulmonary metastases and the other five had a Ewing's sarcoma (stage IIb and stage III in two). The tumour occupied the proximal femur in all six patients. Three patients (cases 1, 2 and 6) suffered pre-operative pathological fractures and one (case 6) had plate osteosynthesis. Finally, three patients (cases 1, 5 and 6) received radiotherapy in addition to chemotherapy.

Twelve patients were treated by type BIIIa rotationplasty combined with chemotherapy. The mean age at operation was 5 years and 9 months (2.8 to 10.6) and their mean follow-up 4 years and 10 months (0.3 to 9.8). Five had an osteosarcoma (stage IIb in four and stage III in one), six had a Ewing's sarcoma (stage IIb) and one a primitive neuroectodermal tumour (stage IIb). The tumour involved the entire femur in three patients, the proximal and diaphyseal femur in two, the proximal femur with a distal skip metastasis in one, the distal femur in one, diaphyseal femur in one, the distal femur with a proximal skip metastasis in one and the proximal femur in three. One patient (case 9) simultaneously suffered two pathological fractures of the proximal and distal right femur. Finally, one patient with a Ewing's sarcoma (case 13) suffered two pre-operative pathological fractures after radio- and chemotherapy with consequent femoral malrotation, limb shortening and palsy of the sciatic nerve.

The parameters used to assess the outcome were post-operative complications, oncological outcome, range of movement and functional score according to Hillmann et al.¹² and Enneking et al.¹⁴ In addition, anteroposterior (AP) and lateral radiographs were used to estimate the post-operative progress of the reconstructed joints.

The range of movement is that at the rotated knee in patients with type BI rotationplasty and recorded as degrees of flexion, extension, abduction, adduction and internal and external rotation at the 'neohip' in patients with type BIIIa. The range of movement was available for all the patients who underwent type BI rotationplasty, but it could not be measured in one patient with type BIIIa rotationplasty (case 15), who had an above-knee amputation because of post-operative complications. We have therefore compared the mean range of movement of all patients with type BI with that of 11 with type BIIIa rotationplasty.

Radiological assessment of patients with type BI rotationplasty was based on the extent of the following degenerative changes in the reconstructed hip: joint-space narrowing, and the presence of osteophytes, subchondral sclerosis and cysts. Radiographs from patients with type BIIIa rotationplasty were further assessed with respect to the extent of remodelling of the tibial head and its degree of containment within the acetabulum.

The limited number of patients who undergo these two types of rotationplasty, made our sample size too small to allow a detailed statistical analysis. However, we have included all the patients who underwent these two types of rotationplasty in two University Hospitals over the last 20 years.

Results

Post-operative complications. These are detailed in Table II. Complications occurred in three of the six patients, who underwent type BI rotationplasty. In one (case 2) there was loosening of the osteosynthesis, thrombosis of the venous anastomosis and haematoma and the patient required revision osteosynthesis, re-anastomosis and drainage of the haematoma on the first post-operative day. The osteosynthesis was removed ten months later. In another (case 5) there was a haematoma and embolism of the femoral artery requiring embolectomy on the day of the operation. The third patient (case 4) presented with delayed wound-healing and required debridement and repeat suturing.

Post-operative complications occurred in four of 12 patients who had undergone type BIIIa rotationplasty. One (case 11) developed a haematoma with temporary total paresis and sensory loss over the affected limb, which eventually recovered. The haematoma was drained on the first post-operative day. Subsequently, the patient developed shortening of 6.5 cm of the rotated limb which did not affect the clinical result. The second patient (case 9) had post-operative limb shortening of 2 cm, which was of trivial clinical importance. The third patient (case 14) required a second procedure to shorten the proximal rotated fibula shortly after the initial operation. This patient also suffered a fracture of the proximal tibia two years after the operation. The fourth patient (case 15) developed a haematoma and compartment syndrome, requiring decompression on the first post-operative day. Despite this an above-knee amputation with sparing of the proximal tibia was required on the 12th post-operative day for progressive muscle necrosis.

Oncological outcome. One patient treated by type BI rotationplasty (case 6) had a stage-III Ewing's sarcoma and died two years and eight months after the operation from pulmonary and bone metastases. Two more patients (cases 1 and 4) had pulmonary metastases but are in remission. Thus, five of six patients who had type B1 rotationplasty had no evidence of recurrence of the tumour and were free from disease at follow-up. One of the 12 patients who had type BIIIa rotationplasty (case 18) had stage III osteosarcoma and died from metastases in the lung and proximal tibia approximately three months after operation. The remaining 11 patients had no evidence of recurrence and were free from disease at follow-up.

Range of movement. The mean flexion and extension of the reconstructed hip in patients with type BI rotationplasty was 55.8° (40 to 70) and 0°, respectively, compared with a mean 'neohip' flexion of 82.7° (60 to 110) and extension of 0° in those with type BIIIa rotationplasty (Table II). Furthermore, patients with type BI rotationplasty did not have any abduction, adduction or internal or external rotation at the reconstructed hip. By contrast, type BIIIa patients had a mean abduction and adduction of 25.5° (15 to 35) and 20° (10 to 35) and a mean internal and external rotation of 18.4° (10 to 40) and 21.2° (10 to 50), respectively.

Fig. 2a

Fig. 2b

Case 5. AP radiographs of a boy aged four years and nine months with type BI rotationplasty, taken post-operatively and at follow-up nine years later showing a) good fusion of the osteosynthesis without any axis deviation and b) a hyperadduction deformity because of ossification of the medial aspect of the distal femoral growth plate, presumably caused by an altered distribution of load.

Enneking score. The mean Enneking score in the patients who underwent type BI rotationplasty was 20.2 of 30 (13 to 24) (Table II). One patient (case 6) was in a poor physical condition because of post-operative metastases and eventually died. If this patient was excluded the mean Enneking score in these patients rose to 21.6 of 30 (19 to 24), which represented 72% of normal function.

The mean Enneking score among the type BIIIa rotationplasty patients was 23.9 of 30 (19 to 27). However, assuming we excluded the patient who presented with walking difficulties because of pre-operative palsy of sciatic nerve (case 13) which was irrelevant to the type of rotationplasty offered, the mean Enneking score for these patients then rose to 24.4 of 30 (23 to 27), which denoted 81.3% of normal function (Table II). Enneking scores were not assigned to two patients (cases 15 and 18) because amputation was necessary in one (case 15) and the other (case 18) died from metastases shortly after the operation.

Radiological results. Moderate joint-space narrowing and subchondral sclerosis were seen in two of the six patients treated by type BI rotationplasty (cases 1 and 3). One

Fig. 3

Case 6. AP pelvic radiograph taken two years and eight months after a type BI rotationplasty in a boy aged eight years and seven months. There is stable osteosynthesis with no signs of axis deviation or loosening and a well-preserved knee without any evidence of degenerative joint disease.

Fig. 4

Case 7. AP pelvic radiograph of an 11-year-old girl who had a type BIIIa rotationplasty at the age of five years. Six years after operation, excessive remodelling has transformed the tibial head into the semblance of a normal femoral head and has remarkably improved its containment in the acetabulum.

patient (case 5) developed progressive hyperadduction of the distal femur (Fig. 2), while the radiographs of the remaining three patients (cases 2, 4 and 6) showed moderately demineralised bones with no signs of degenerative joint disease (Fig. 3).

Of the patients treated by type BIIIa rotationplasty, five had excellent radiological results (Fig. 4) with no evidence

of degenerative disease and remarkable tibial remodelling and containment in the acetabulum (cases 7 to 9, 12 and 17). Another (case 16), who was aged 10.6 years at operation showed remarkably good remodelling of the 'neohip' (Fig. 1). Less impressive tibial remodelling was seen in six patients (cases 10 to 14 and 16). Two patients (cases 15 and 18) were not included in the assessment since one (case 15) had an above-knee amputation and the other (case 18) died from metastases three months after the operation.

Discussion

The improved prognosis of patients with malignant bone tumours achieved by the use of neoadjuvant chemotherapy underlines the importance of successful limb-salvage surgery.¹⁵ In children less than ten years of age, limb-salvage procedures using extendible endoprostheses carry the risk of numerous complications and re-operations.¹⁶ Rotationplasty has the advantage of being a single operation which does not usually require revision procedures and is associated with remarkably good function.^{2,11,17} It is therefore a good treatment option in children with malignant bone tumours of the lower limb.

Winkelmann¹⁻³ developed type BI rotationplasty for tumours of the proximal femur and type BIIIa rotationplasty for those involving the entire femur in children. The idea underlying the use of type BI rotationplasty was that preservation of the knee could lead to improved post-operative function. However, the use of type BIIIa rotationplasty gives excellent functional results. The plasticity of the cartilaginous tibial plateau in children allows it to remodel and function like a femoral head.^{2,11} In our study, patients with a type BIIIa rotationplasty had a better range of movement at the reconstructed hip and a higher Enneking score, 81.3% compared with 72% in patients with type BI rotationplasty. The Enneking score was devised for use in adults but we felt it was the best score to use although it has not been validated in children. The lack of abduction, adduction and rotation in the hip as a result of fixation of the knee to the pelvis, restricted patients with a type BI rotationplasty in their activities of daily life. This did not apply to patients with type BIIIa rotationplasty, who had a useful range of abduction, adduction and rotation at the 'neohip'. Furthermore, type BIIIa rotationplasty appeared to be associated with fewer post-operative complications.

An additional problem of type BI rotationplasty is the difficulty of fixing the femur to the pelvis. Loosening of the osteosynthesis with consequent axis deviation can often lead to an increased stay in hospital and further re-admissions. In one patient (case 5) a hyperadduction deformity of the distal femur resulted in poor function for which a corrective osteotomy is planned. By contrast, no axis deviation was seen in the patients with type BIIIa rotationplasty.

The limb-length discrepancy which was sometimes seen in type BIIIa rotationplasty (cases 9 and 11) did not appear to be of clinical importance and was easily compensated for by the prosthesis.

Table I. Details of the patients

Case	Gender	Type of operation	Age at operation (yrs)	Diagnosis*	Localisation†	Pre-operative complications‡	Radiotherapy	Chemotherapy	Follow-up (mths)
1	M	BI	9.2	ES	Prox femur R	Fx	Yes	Yes	111
2	F	BI	6.9	ES	Prox femur L	Fx	No	Yes	19
3	F	BI	9.2	ES	Prox femur R	No	No	Yes	148
4	M	BI	6.10	OS	Prox femur L	No	No	Yes	45
5	M	BI	4.9	ES	Prox femur R	No	Yes	Yes	105
6	M	BI	8.7	ES	Prox femur R	Fx and osteosynthesis	Yes	Yes	32
7	F	BIIIa	5.1	OS	Dist femur L	No	No	Yes	105
8	F	BIIIa	4.1	ES	Entire femur R	No	No	Yes	74
9	F	BIIIa	3.5	ES	Entire femur R	Fx§	No	Yes	71
10	M	BIIIa	4.3	PNET	Prox and diaph femur R	No	No	Yes	54
11	F	BIIIa	9.8	OS	Entire femur R	No	No	Yes	57
12	M	BIIIa	2.8	ES	Prox femur and dist skip metastases L	No	No	Yes	116
13	M	BIIIa	8.0	ES	Prox femur R	Fx¶	Yes	Yes	73
14	F	BIIIa	3.1	ES	Prox femur R	No	No	Yes	30
15	M	BIIIa	3.11	ES	Prox femur L	No	No	Yes	13
16	M	BIIIa	10.6	OS	Prox and diaph femur R	No	No	Yes	65
17	M	BIIIa	6.0	OS	Dist and mid femur	No	No	Yes	32
18	F	BIIIa	9.8	OS	Dist femur and prox skip metastases R	No	No	Yes	3

* ES, Ewing's sarcoma; OS, osteosarcoma; PNET, primitive neuroectodermal tumour

† Prox, proximal; dist, distal; diaph, diaphyseal; R, right; L, left

‡ Fx, fracture

§ pathological fractures of proximal and distal femur

¶ two pathological fractures with femoral malrotation, palsy of the sciatic nerve and limb-length discrepancy of 8 cm

Table II. Post-operative function and results in the 18 patients

Case	Type of operation	Post-operative complications	Repeat operations	Disease status*	Hip			Enneking score ¹⁴
					Flexion/extension (°)	Abduction/adduction (°)	Int/ext rotation (°)	
1	BI	None	None	NED	45/0/0	0/0/0	0/0/0	23/30
2	BI	Loosening of osteosynthesis, haematoma and thrombosis	Re-osteosynthesis and re-anastomosis. Osteosynthesis removed	NED	40/0/0	0/0/0	0/0/0	19/30
3	BI	None	None	NED	50/0/0	0/0/0	0/0/0	23/30
4	BI	Wound-healing problem	Debridement and repeat suturing	NED	70/0/0	0/0/0	0/0/0	24/30
5	BI	Haematoma and embolism of the femoral artery	Embolectomy on the same day	NED	60/0/0	0/0/0	0/0/0	19/30
6	BI	None	None	DOD	70/0/0	0/0/0	0/0/0	13/30†
7	BIIIa	Tibial greenstick fracture	None	NED	75/0/0	35/0/15	10/0/20	24/30
8	BIIIa	None	None	NED	110/0/0	35/0/35	40/0/50	27/30
9	BIIIa	Post-operative shortening of the rotated limb by 2 cm	None	NED	80/0/0	30/0/30	10/0/25	25/30
10	BIIIa	None	None	NED	80/0/0	20/0/15	30/0/25	24/30
11	BIIIa	Haematoma with temporary total paresis and sensory loss Post-operative shortening of the rotated limb by 6.5 cm	Drainage of haematoma	NED	90/0/0	20/0/10	10/0/10	23/30
12	BIIIa	None	None	NED	100/0/0	35/0/30	20/0/25	25/30
13	BIIIa	None	None	NED	60/0/0	20/0/10	15/0/10	19/30
14	BIIIa	Fracture of the proximal tibia	Shortening of the proximal fibula	NED	85/0/0	26/0/15	19/0/18	24/30
15	BIIIa	Haematoma and compartment syndrome	Pressure relief procedure and above-knee amputation	NED	N/A	N/A	N/A	N/A
16	BIIIa	None	None	NED	80/0/0	20/0/20	20/0/20	24/30
17	BIIIa	None	None	NED	60/0/0	15/0/15	10/0/10	24/30
18	BIIIa	None	None	DOD	90/0/0	25/0/25	18/0/20	N/A

* NED, no evidence of disease; DOD, died of disease

† because of recurrence of tumour and deteriorated physical condition

Furthermore, in the two patients (cases 13 and 15) with the worst functional outcome of those who had undergone type BIIa rotationplasty the operative procedure did not affect their post-operative function. Indeed, compartment syndrome, which led to an above-knee amputation in one of these patients (case 15) could just as well have complicated type BI rotationplasty. The palsy of the sciatic nerve which was responsible for reduced function in one patient (case 13) was present pre-operatively and not a complication of the type BIIa rotationplasty. The compartment syndrome which led to an above-knee amputation in another patient (case 15) was a major complication affecting the functional result. Finally, no patient in either group had symptoms of degenerative joint disease.

These results raise the question as to whether the knee should be preserved in children with a tumour of the proximal femur. We believe that the functional results of type BIIa rotationplasty are superior to those of type BI rotationplasty in which the knee is preserved and justify the use of the former in tumours of the proximal femur. However, the limited number of patients in the study do not allow statistical analysis. We therefore report our findings and conclusion and hope that these will be confirmed by additional similar studies in the future.

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