Tibial Growth Disturbance Following Distal Femoral Resection and Expandable Endoprosthetic Reconstruction

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Background: In growing children, an expandable endoprosthesis is commonly used after distal femoral resection to compensate for loss of the distal femoral physis. Our hypothesis was that such prostheses can affect proximal tibial growth, which would contribute to an overall leg-length discrepancy and cause angular deformity.

Methods: Twenty-three skeletally immature patients underwent the placement of a distal femoral expandable endoprosthesis between 1994 and 2012. Tibial length, femoral length, and mechanical axis were measured radiographically to determine the growth rate.

Results: No patient had radiographic evidence of injury to the proximal tibial physis at the time of surgery other than insertion of the tibial stem. Fifteen (65%) of the patients experienced less proximal tibial growth in the operative compared with the contralateral limb. In ten (43%) of the patients, the discrepancy progressively worsened, whereas in five (22%) of the patients, the discrepancy stabilized. Seven patients did not develop tibial length discrepancy, and one patient had overgrowth of the tibia. For the ten patients with progressive shortening, the proximal tibial physis grew an average of 4.0 mm less per year in the operative limb. Five (22%) of the patients had ≥ 20 mm of tibial length discrepancy at last follow-up. Three of these patients underwent contralateral tibial epiphysiodesis. Three patients required corrective surgery for angular deformity.

Conclusions: The tibial growth plate may not resume normal growth after implantation of a distal femoral prosthesis. Physeal bar resection, prosthesis revision, and contralateral tibial epiphysiodesis may be needed to address tibial growth abnormalities.

Level of Evidence: Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

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Primary sarcomas of bone often develop in skeletally immature patients. The occurrence of osteosarcoma, for example, peaks during adolescence and coincides with the pubertal growth spurt. Osteosarcoma is usually located in the metaphyseal region of long bones, with more than 50% of the cases occurring about the knee, thus creating potential problems with skeletal growth for many patients¹. With progress in chemotherapeutic treatment, limb salvage can now be performed in

most patients, and no significant difference has been demonstrated in terms of overall survival or disease-free survival between limb salvage and amputation^{2,3}.

Limb salvage in skeletally immature patients presents unique reconstructive challenges. Maintaining equal limb lengths at the completion of the growth period is the desired result. In children with normal bone health, the remaining growth of each segment of the lower extremity can be

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TABLE I Patient Data

No. of patients	23
Age at resection* (yr)	9.9 (6-13)
Sex (no. [%])	
Male	14 (61)
Female	9 (39)
Osteosarcoma diagnosis (no. [%])	23 (100)
Enneking stage ⁴⁸ (no. [%])	
IIa	1 (4)
IIb	18 (78)
III	4 (17)
Follow-up duration (yr)	
Mean	6.3
Std. dev.	4.6
Median	5.5
Range	0.7-18.8
Duration of skeletal growth with implant (yr)	
Mean	3.7
Std. dev.	2.1
Median	3.5
Range	0.5-8.3
Type of expandable implant (no. [%])	
Biomet	8 (35)
Repiphysis	14 (61)
Stanmore	1 (4)
Patient status at last follow-up (no. [%])	
No evidence of disease	20 (87)
Died of disease	3 (13)
Limb status at last follow-up (no. [%])	
Amputation for local recurrence	2 (9)
Expandable prosthesis	
Skeletally immature	4 (17)
Skeletally mature	4 (17)
Adult prosthesis	
Skeletally mature	13 (57)

*The value is presented as the mean with the range in parentheses.

estimated^{4,5}. It has been reported that growth of the distal femoral physis averages 13 mm per year in normal children (except for the last two years of growth, when it slows) and the proximal tibial physis, 9 mm per year (again, excluding the last two years)⁶.

If the distal femoral growth plate cannot be preserved, a discrepancy in leg length is expected at skeletal maturity. Surgical options allowing growth include distraction osteogenesis, vascularized physal transfer, and the use of an expandable endoprosthetic implant⁷. Alternative surgical procedures for growing children include amputation⁸ and ro-

tationplasty⁸⁻¹⁰. The optimal choice for young patients remains controversial¹⁰⁻¹³.

Expandable endoprosthetic implants have been used for more than thirty years¹⁴⁻¹⁶. Expansion is periodically performed when leg-length discrepancy develops, and the procedure is repeated as needed during growth to achieve equal overall leg length. Studies have shown successful lengthening of expandable implants^{10,17-20}. Despite the numerous associated complications^{18,21-23}, most patients seem to have minimal leg-length discrepancy at skeletal maturity^{10,24} and good functional outcomes^{10,22,23,25}. Emotional acceptance of this complex reconstruction is generally satisfactory¹⁰.

Different types of expandable distal femoral endoprostheses have been described^{16,18-20,24,26-28}. In all of the designs, the tibial baseplate is a stemmed implant, which can be cemented²⁹, noncemented with a smooth metallic surface^{23,28}, or a sliding stem inside a polyethylene tube^{15,30}. All implants require perforation of the tibial growth plate, and meticulous technique is necessary to limit damage to the physis.

Previous authors have observed that an open physis can continue to grow after transgression by noncemented^{15,26,30,31} or cemented stems²⁹. Cement fracture can be observed in certain cases as the tibia continues to grow²⁹. It has also been noted that smooth pin fixation across the physis for fractures is compatible with continued growth³². However, few authors have critically examined how much growth occurs at the proximal physis after penetration by a stem.

The purpose of the present study was to evaluate and quantify tibial growth following distal femoral resection and placement of an expandable endoprosthesis. Our hypothesis was that the proximal tibial growth plate may not resume normal growth after insertion of a stem. Growth retardation may contribute to overall leg-length discrepancy, joint line asymmetry, and angular deformity.

Materials and Methods

Between 1994 and 2012, thirty-one skeletally immature patients underwent wide excision of the distal part of the femur and reconstruction with an expandable prosthesis. All patients had a distal femoral primary sarcoma. All surgeries were performed at a single institution. Twenty-three patients were found to have adequate data to be included in this retrospective cohort study (Table I); seven patients without bilateral scanograms or teleroentgenograms and one patient who had undergone bilateral distal femoral replacement were excluded. Patients had a minimum of twelve months of follow-up unless death occurred prior to twelve months (one patient). Demographic, surgical, and radiographic data were reviewed for the twenty-three patients. The study was approved by the institutional review board.

All twenty-three patients had a diagnosis of high-grade osteosarcoma and received both preoperative and postoperative chemotherapy. Three expandable implants were used (Table I), with the choice of implant made according to surgeon preference. The custom Biomet design employed a cemented tibial stem across the proximal tibial physis. The Repiphysis (Wright Medical Technology) and Stanmore Implants (Juvenile Tumour System [JTS]) designs utilized a smooth stem. During preparation of the tibia, care was taken to remove only the articular cartilage, preserving the physis and its vascular supply through the ring of LaCroix. The tibial canal was minimally reamed for the tibial stem, limiting the area of penetration to the central portion of the growth plate. Following surgery, all patients were allowed full weight-bearing and range-of-motion exercises on the first postoperative day.

TABLE II Association Between Progressive Tibial Discrepancy (TD) and Patient Characteristics

Covariate	Progressive TD, N = 10	Nonprogressive TD, N = 13	P Value*
Mean age at resection (yr)	9	10.6	NS
Duration of growth with implant (yr)	4.5	3.1	NS
Sex (no. [%])			NS
Male	6 (60)	8 (62)	
Female	4 (40)	5 (38)	
Average expansion length (mm)	65.8	28.8	0.009
Average no. of expansions	6.8	2.2	0.007
Prosthesis type (no.)			NS
Biomet	3	5	
Repiphysis	7	7	
Stanmore	0	1	

*NS = not significant.

Tibial growth was assessed by full-length standing anteroposterior radiographs of the lower extremity with the patella pointing anteriorly (teleroentgenogram)³³ or with scanograms, which utilize three radiographic

exposures centered on the hip, knee, and ankle joints. With this technique, the patient remains supine next to a ruler, with both patellae pointing toward the ceiling³⁴. Radiographs of inadequate quality, especially those with improper

Fig. 1

Tibial, femoral, and overall leg-length discrepancies are shown for all twenty-three patients. The patients are arranged in order of decreasing tibial length discrepancy. Negative values indicate shortening of the operative leg compared with the contralateral leg. Lengthening of the femur to compensate for tibial shortening was a common finding (nine patients with ≥ 1.0 cm of femoral overlengthening). SM = skeletal maturity, DOD = died of disease, E = epiphysiodesis, and A = amputation.

TABLE III Patient Lengthening Data

	Age* (yr)	Sex	Total Follow-up Duration (mo.)	Growth Period† (mo.)	Final Tibial Length‡ (mm)	Progressive Tibial Length Discrepancy	Tibial Length Discrepancy§ (mm)
1	9	F	62	60	341	Yes	-25
2	13	M	173	41	385	Yes	-25
3	10	M	44	44	380	Yes	-25
4	7	F	68	73	315	Yes	-25
5	6	M	108	100	350	Yes	-20
6	12	F	225	51	370	No	-15
7	9	F	37	32	340	Yes	-15
8	8	F	94	64	315	Yes	-15
9	10	M	33	15	350	No	-10
10	8	M	195	83	360	No	-10
11	8	M	78	43	300	Yes	-10
12	11	F	69	56	320	No	-10
13	9	M	51	33	365	Yes	-5
14	11	F	39	32	335	No	-5
15	11	M	66	54	385	Yes	-5
16	13	M	80	34	383	No	0
17	7	F	40	9	280	No	0
18	11	F	8	5	340	No	0
19	10	M	97	81	350	No	0
20	8	M	79	56	370	No	0
21	13	M	39	31	350	No	0
22	12	M	56	35	380	No	0
23	12	M	12	12	330	No	10

*At index operation. †Postoperative period when scanogram or teleoroentgenogram data were available. ‡Last measured length of the tibia of the operative side. §Length of the operative side minus the nonoperative side. A negative value indicates that the operative side is shorter. #R = Repiphysis, B = Biomet, and S = Stanmore.

rotation of the leg and asymmetrical patellar positions, were excluded. Tibial length, femoral length, and overall leg length were measured by one of the authors (A.A.), with the final tibial and leg-length discrepancies verified by the senior author (P.P.L.). Overall leg length was measured from the top of the femoral head to the center of the tibial plafond³⁵. The tibial length of the operative leg was measured from the center of the medial prosthetic plateau, including the polyethylene bearing, to the center of the tibial plafond. The tibial length of the contralateral limb was measured from the center of the medial plateau to the center of the tibial plafond. Femoral length was obtained by subtracting the tibial length from the overall leg length.

The mechanical axis was determined as measured between the center of rotation of the femoral head, the center of the intercondylar notch (at the axle of the prosthesis), and the center of the tibial plafond³⁶. Serial measurements were performed on the operative leg and on the contralateral extremity to assess mechanical axis variation with growth. Indirect signs of partial physeal growth arrest included displacement of the tibial stem in the canal and cortical remodeling around the stem.

The growth period with implant was defined as the duration of time between the date of surgical resection and the date when the distal femoral and proximal tibial growth plates of the nonoperative leg were closed as evident on radiographs. For patients who underwent an amputation for local recurrence or who died of disease, the end of the measureable growth period was considered the time of the last scanogram before amputation or death. The last available

scanogram or teleoroentgenogram was used for the remaining skeletally immature patients. One patient who did not undergo expansion because of local recurrence was included because there were data on tibial growth for the duration of the implant.

For statistical analysis, the mean, median, range, and standard deviation were calculated for continuous variables. The Wilcoxon rank-sum test was used to examine the association between tibial growth and continuous variables. The Fisher exact test was used to examine the association between tibial growth disturbance and categorical variables. A p value of <0.05 was considered significant. SAS software (version 9.3, SAS Institute) was used for all analyses.

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Results

At the time of the initial surgery, patients were a mean age of 9.9 years (Table I). The mean period of skeletal growth between initial surgery and the last scanogram or teleoroentgenogram was 3.7 years (Table I). Three patients developed local recurrence and were treated with either resection of the

TABLE III (continued)

Total Leg-Length Discrepancy§ (mm)	Total Femoral Expansion (mm)	No. of Expansions	Prosthesis#	Comment
7	60	6	B	
0	50	4	B	
-20	100	8	R	
-25	89	9	R	Epiphysiodesis and correction of recurvatum
5	90	12	R	Epiphysiodesis and physeal bar excision
8	20	1	B	
-10	35	2	R	Epiphysiodesis
-10	120	18	R	
5	0	0	R	Amputation for recurrence; died of disease
0	100	8	B	
-15	34	2	R	
-25	20	1	B	
5	30	3	B	Died of disease
5	23	2	R	
-15	50	4	R	
0	20	1	B	
0	30	2	R	Hip disarticulation for recurrence
0	10	1	R	Died of disease
-10	47	3	R	
-15	70	6	B	
-15	0	0	R	
15	23	1	R	
05	12	2	S	

recurrence (one patient) or amputation (two patients). Seven patients developed lung metastasis. At last follow-up, four of these patients were alive without disease, and three had died. The other sixteen patients were alive without disease.

Examination of the early postoperative radiographs showed that the proximal tibial osteotomy was correctly made in all cases without injury to the proximal tibial physis. All patients exhibited some growth at the proximal tibial physis after surgery. A total of ninety-six expansions were performed (mean, 4.2 per patient). The average total expansion length per patient (and standard deviation [SD]) was 44.9 ± 34.7 mm (range, 0 to 120 mm).

At last follow-up, fifteen (65%) of the patients exhibited tibial length shortening (Fig. 1). For the entire group of twenty-three patients, the mean reduction in tibial length was 9.1 mm, which represented a reduction of 2.5 mm per year of growth. In ten (43%) of the patients, the discrepancy progressively worsened during growth, and for these patients, the mean reduction in tibial length was 17 mm, which represented a reduction of 4.0 mm per year of growth. In five patients, a discrepancy developed after surgery but stabilized with time. The number of lengthening procedures and total expansion length were factors associated with progressive tibial discrepancy (Table II).

Five patients, four of whom were skeletally mature, had ≥ 20 mm of tibial length discrepancy at last follow-up (Fig. 2).

Three (13%) of the patients underwent a contralateral tibial epiphysiodesis. The final overall leg-length discrepancy among skeletally mature patients was >20 mm in two patients (Table III). One patient with a Biomet prosthesis developed varus deformity and shortening of the tibia. Revision of the prosthesis corrected the varus but extensive scarring precluded complete correction of leg-length discrepancy. The second patient had a Repiphysis implant and underwent overlengthening of the femur and contralateral epiphysiodesis to minimize leg-length discrepancy. Lengthening of the femoral component was a technique used in several patients to compensate for the tibial length lost (Fig. 1).

Three patients developed angular deformity at the proximal part of the tibia. Progressive mechanical varus developed in two of these patients. The first patient underwent anteromedial physeal bar resection (Fig. 3) and revision of the tibial baseplate to correct a 10° varus deformity as well as recurvatum at the knee. He also underwent a contralateral epiphysiodesis two years later to stop the progression of his tibial length discrepancy that reached 20 mm. The second patient underwent a tibial component revision with a longer tibial stem for a 9° varus deformity. The third patient did not have varus or valgus deformity, but he developed anterior tibial physeal growth arrest and recurvatum, which was

Fig. 2

Tibial and total leg-length discrepancy are illustrated for the seventeen patients who reached skeletal maturity. LLD = leg-length discrepancy.

Fig. 3

Successive radiographs of the same patient showing anterior and medial physeal growth arrest. Progressive varus and recurvatum is noted along with remodeling of the posterior cortex around the stem.

manifested radiographically by posterior migration of the tip of the tibial stem and cortical remodeling around the stem. This patient underwent revision for mechanical failure of the expandable component. The tibial component was exchanged for an adult-type stem, and the deformity was corrected at the time of revision.

Discussion

The findings of this study support our hypothesis that the proximal tibial physis often does not continue to grow normally after insertion of a stem through the physis. Tibial length discrepancy was a common problem that developed in approximately two-thirds of the patients.

Our study had several limitations, including a relatively small number of patients and the retrospective nature of the experimental design with the potential for detection bias. In addition, there is a degree of imprecision in determining on radiographs when physeal closure is complete. The end of growth may have occurred at some point between two scanograms, and the true duration of the growth period may have been slightly less than as measured. The use of radiographs to accurately determine tibial length is also a limitation. If a flexion contracture of the knee develops after lengthening, the true tibial length may be underestimated on radiographs because of foreshortening. We relied on the clinic notes to indicate whether there was any flexion contracture. In general, lengthenings were not performed if patients had not regained full extension and flexion to 90°.

The degree of tibial growth retardation varied considerably. In seven patients, the proximal tibial physis grew at the same rate as that in the unaffected limb. One patient had slight overgrowth of the tibia. In five patients, there was growth retardation initially, but the amount of discrepancy seemed to stabilize with time. For the remaining ten patients, the difference in tibial length between limbs worsened with time. Assuming that the discrepancy originated only from the proximal tibial physis, growth retardation reached 4 mm per year in the latter subset of patients.

Our results are consistent with those of Cool et al.¹⁵, who used Harris growth arrest lines to determine the proximal and distal tibial growth-plate contributions to overall tibial growth. These authors reported that the proximal tibial physis grew at 69% of the rate of the contralateral leg after insertion of a tibial baseplate and stem¹⁵. They noted great variability in the growth rate of the proximal physis, ranging from 18% to 136% of the contralateral side. The distal tibial physis resumed normal growth without compensating for the diminished growth of the proximal tibial physis¹⁵.

The mechanism of growth-plate malfunction is not clear. One theory is that it results from vascular insult to the physeal blood supply and the ring of LaCroix during surgery, while another theory is that it results from physeal bar formation near the cement or stem of the implant. Additional contributing factors include surgical technique, implant design, body weight, age at the time of surgery, and activity level. Our statistical analysis did not reveal a significant correlation with these factors, but the small number of patients may have precluded finding an effect. The question of whether design parameters might affect growth is important. In addition to smooth versus cemented stems, stem diameter, stem length, fixed hinge versus rotating hinge (which requires a larger reaming diameter for the sleeve), and the ratio of the area of the stem to the cross-sectional area of the proximal tibial physis could all potentially be relevant.

An interesting question is whether growth disturbance develops during the period of postoperative chemotherapy, which may last nine to twelve months. Chemotherapy can temporarily cause growth retardation, and growth arrest lines are common findings. Unfortunately, we did not have sufficient data points within the postoperative chemotherapy period to

make an accurate assessment of physeal growth rate during this phase of treatment.

The number of lengthenings and the total expansion length of the femoral component were both associated with progressive tibial discrepancy. However, neither the age at initial resection nor the duration of growth seemed to be associated with increased risk in our cohort or in the study by Cool et al.¹⁵. One possible explanation is that the survival of patients was not the same, and very young patients who do not survive sufficiently long may not exhibit tibial length discrepancy. In some patients, growth retardation seemed to stabilize over time. These patients were closer to skeletal maturity and grew for a shorter period of time after surgery (Table II). Physeal growth naturally slows near the time of skeletal maturity, thereby lessening the potential for length discrepancy. Time to skeletal maturity might be as important a factor to consider as the absolute age at resection. Sex may be a related factor, as females reach skeletal maturity at a younger age than males. Clearly, more work with a greater number of patients is needed to address these issues.

In most series, expandable endoprostheses have been reported to achieve overall leg-length equivalence for the majority of patients at skeletal maturity^{22,24}. Those findings were confirmed by our study, and only two skeletally mature patients in our cohort had >20 mm of leg-length discrepancy. A discrepancy of <20 mm is minimally symptomatic because long-term adaptation occurs³⁷⁻⁴⁰.

There are several options for addressing tibial length discrepancy. One patient in our series underwent the placement of a tibial augment and thicker polyethylene bearing to increase tibial length. Care must be taken with this strategy because of the risk of patella baja, weakness, and decreased range of motion^{41,42}. Contralateral proximal tibial epiphysiodesis is appropriate for discrepancies of 2 to 5 cm, particularly in patients with substantial growth remaining³⁵. Femoral overlengthening can also help to offset tibial shortening. Although this was used in nine of our patients, it could have negative consequences. In addition to cosmetic appearance, there is the potential impact on function, including stiffness after repeated expansions and weakening of the quadriceps. Our data do not address these issues directly.

The current literature on expandable endoprostheses infrequently mentions secondary angular deformity^{16-20,23,24,26-28,43}. Cool et al. found no angular deformity in their study¹⁵. Henderson et al. reported that varus deformity developed in two of twenty-five cases, necessitating endoprosthetic revision⁴⁴.

Our study showed that angular deformity may occur in both sagittal and coronal planes. The pathogenesis may be related to asymmetric physeal growth and small physeal bars that are hidden by radiopaque implants. Once angular deformity occurs, asymmetric compressive forces may cause further growth inhibition according to the Heuter-Volkman principle^{45,46}. Excessive pressure on the medial physis may lead to progressive varus⁴⁷. The three cases of angular deformity in our series were associated with a Repiphysis implant, which employs a relatively short stem that often ends in the metaphysis. Surgical correction may require physeal bar resection and a longer stem with better press fit and perpendicular alignment in the diaphysis.

On the basis of our findings, approximately two-thirds of skeletally immature patients who undergo distal femoral resection and reconstruction with an expandable distal femoral endoprosthesis have diminished growth of the tibial physis, and growth loss can reach 4.0 mm per year if the deficit progresses with time. Impairment of tibial growth can contribute to leg-length discrepancy and result in overlengthening of the femur as a compensatory measure. There is a risk of angular deformity from partial physal closure that has not been emphasized in previous studies. Patients who undergo numerous expansions and who undergo a greater expansion of the component seem to be especially at risk for growth disturbance of the tibia. We recommend that, in addition to monitoring overall leg length, clinicians should also pay attention to individual femoral length, tibial length, and mechanical axis. These parameters should be assessed in a serial fashion and compared with assessments of the contralateral extremity. Patients with severe growth disturbance of the proximal tibial physis may require

additional surgery, including contralateral epiphysiodesis and tibial component revision. ■

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