How Much Clinical and Functional Impairment do Children Treated With Knee Rotationplasty Experience in Adulthood?

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Abstract

Background Rotationplasty may be indicated for some children with osteosarcoma in the distal femur or proximal tibia; in properly selected patients, it may offer functional advantages over transfemoral amputation and more durable results than a prosthesis. The clinical and functional outcomes reported for this procedure generally have been limited to studies with a mean followup of approximately 8 years in terms of Musculoskeletal Tumor Society Score (MSTS), physical examination, and gait analysis. However,

the effects of residual thigh-shank length on gait have not been explored to our knowledge.

Questions/purposes We asked: (1) Do differences in the length of the surgically treated residual thigh-shank relative to the contralateral thigh result in altered gait patterns? (2) What were the clinical and functional impairments and radiographic findings of patients who underwent rotationplasty and who survived to adulthood? (3) Do gait analysis findings in adults differ from previously reported findings in children in terms of relevant gait parameters such as maximal ground reaction forces and sagittal knee angles?

Methods From January 1986 to December 2009, 254 children (age range, 3–14 years) affected by high-grade bone sarcomas located in the distal half of the femur were surgically treated at our institute. Forty-two of these patients (16.5%) underwent rotationplasty. During this period, three adolescents older than 15 years were treated by rotationplasty owing to the tumor volume and extracompartmental involvement. In total, 45 patients underwent rotationplasty. From January 1986 to December 2000, rotationplasty generally was the preferred treatment for patients younger than 9 years with a high-grade bone sarcoma calling for an intra- or extraarticular resection of the distal femur, as long as the sciatic nerve could be spared. From January 2001, the procedure was not used as often. Of the 45 patients who underwent a rotationplasty, 14 died of disease at a mean of 37 months (31%); 31 patients (69%) were survivors at the time the study was done, 29 of whom were continuously disease free (64%) and two had no evidence of disease after a pulmonary metastasectomy (5%). These 31 patients were invited to participate in the study, and 25 of the 31 agreed to participate. There were 15 males and 10 females with a mean age of 23.8 years (SD, 7.5 years) and mean followup of 15 years (SD, 5.8 years). Clinical assessment included the MSTS score (total score ranges between 0 and 30 with 0

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indicating poor results and 30 indicating good results), obtained by clinical assessment and patient interview, measurements of the residual thigh-shank length and of the contralateral thigh, of the lengths of the surgically treated and contralateral feet, and of active ROM of the rotated and contralateral ankles. Of the 25 patients, 22 (88%) agreed to have lower limb radiographs and 16 (64%) agreed to perform gait analysis.

Results The residual thigh-shank was, on average, 5.8% longer than the contralateral thigh. Differences in the length of the residual thigh-shank relative to the contralateral thigh resulted in altered gait patterns. Patients with longer residual thigh-shank length had greater pseudoknee flexion during stance and swing. Patients with shorter residual thigh-shank length walked with a gait similar to that of controls. The mean MSTS score was 25 (SD, 2). With respect to the contralateral foot, the surgically treated foot was 10% shorter, the talus 11% shorter in the long axis and 7.6% in the short axis and the calcaneus was 2.7% shorter in the long axis and 8.6% in the short axis. Radiologic arthritis was present in most patients at the tibiotalar, subtalar, and talonavicular joints. As adults, our patients showed improved gait parameters compared with previously reported findings for children undergoing rotationplasty. Vertical ground reaction force during midstance was reduced by 6% and knee ROM during the gait cycle was increased by 24.6°.

Conclusions The residual thigh-shank length influences the gait performance, such that patients with smaller discrepancies between the surgically treated and contralateral sides had the best walking performance. The MSTS score at a mean of 15 years after knee rotationplasty confirmed the results reported in the shorter-term for function and pain. The foot on the surgically treated side was smaller than the contralateral foot, and degenerative changes were present, which could contribute to impaired function. Gait performance, in terms of ground reaction forces and knee ROM, was improved in our adult patients although a difference in loading was still present between the surgically treated and contralateral limbs. Based on these findings, surgeons should endeavor to have the center axis of rotation of the contralateral knee and pseudoknee at skeletal maturity. An excessive residual thigh-shank length in adult patients could require contralateral lengthening to improve functional results.

Level of Evidence Level IV, therapeutic study.

Introduction

Rotationplasty may be indicated for children with osteosarcoma in the distal femur or proximal tibia if there is no tumor infiltration of the sciatic nerve and ROM of the

ipsilateral ankle is preserved [9]. Compared with transfemoral amputation, rotationplasty offers better clinical results by preserving the ankle as a new knee with flexion-extension capability [19, 20, 23]. Several studies have documented good functional outcomes after rotationplasty, in patients with with followup as long as a mean of 8 years after surgery, and who were evaluated clinically by means of the Musculoskeletal Tumor Society (MSTS) score and other measures [1, 10–18, 22, 24], and instrumentally by means of gait analysis [5, 8, 15, 22, 26]. After rotationplasty, patients were observed to have fewer restrictions for daily activities and sport exercises, even at a competitive level [17, 24], and for hobbies compared with patients with endoprostheses [14]. The mid- and long-term quality of life and social relationships of patients after rotationplasty are comparable to those of the general population [28]. In a study by Forni et al. [7], the older patients tended to adapt mentally better than the younger patients.

However, morphologic changes of the rotated foot associated with use of a prosthesis for a loading condition in the socket have been reported [10, 12, 16]. Patients often report soft tissue callosities and irritation around the rotated foot likely attributable to malpositioning and prosthesis stress, which could result in pain and loss of function. Furthermore, based on our clinical experience, not only the rotated foot, but also the residual thigh-shank length (ie, discrepancy between the residual thigh-shank and the contralateral thigh), seem to affect functional outcome, especially walking performance. This aspect, to our knowledge, has not been reported before.

We therefore asked the following: (1) Do differences in the length of the surgically treated residual thigh-shank relative to the contralateral thigh result in altered gait pattern? (2) What were the clinical and functional impairments and radiographic findings of patients who underwent rotationplasty and who survived into adulthood? (3) Do gait analysis findings in adulthood differ from previously reported findings in childhood in terms of relevant gait parameters such as maximal ground reaction forces and sagittal knee angles?

Patients and Methods

This is a retrospective study that was approved by our institutional review board.

Between January 1986 and December 2009, 254 children (age range, 3–14 years) affected by high-grade bone sarcoma located in the distal half of the femur were surgically treated at our institute. Forty-two (16.5%) of these patients underwent rotationplasty. During the period analyzed, three adolescents older than 15 years were treated by rotationplasty because of tumor volume and extracompartmental

involvement. The whole series was comprised of 45 patients who underwent rotationplasty. From 1986 to 2000, rotationplasty generally was the preferred treatment for patients younger than 9 years with a high-grade bone sarcoma calling for an intra- or extraarticular resection of the distal femur, as long as the sciatic nerve could be spared. During the last 10 years of the study period, the procedure was not used as often.

During the 23-year study period, in addition to the 42 rotationplasties, we also performed four hip disarticulations, six transfemoral amputations, 28 knee arthrodeses with various techniques, implantation of 133 prostheses and 15 osteoarticular bone allografts, and 26 distal intercalary biological reconstructions. Arthrodeses were performed only during the first decade (1986–1995) and abandoned thereafter because of frequent complications and lack of acceptance of a stiff knee by the young patients. During the period from 2001 to 2009, only four children had a rotationplasty, whereas 74 prostheses were implanted during that time; progressive development of expandable prostheses partially or totally customized with noninvasive expansion modalities changed the indication (Fig. 1). In our practice, rotationplasty remains our preferred treatment if an intraarticular distal femoral resection is used in a child younger than 5 years. We also continue to perform this procedure in older children or adults if the tumor involves

Fig. 1 The trend in surgical indications at our institution for high-grade sarcomas of the distal femur in children younger than 14 years (1986–2009) is shown.

the whole anterior compartment of the thigh or there is substantial neurovascular involvement.

Of the 45 patients who underwent rotationplasty, 14 died of disease at a mean of 37 months (31%), 31 (69%) were surviving at the time this study was done, 29 were continuously disease free (64%), and two had no evidence of disease after a pulmonary metastasectomy (5%). These patients were invited to participate to the study. Twenty five of these patients agreed to participate. They were 15 males and 10 females, with a mean age of 23.8 years (SD, 7.5 years) and mean followup of 15 years (SD, 5.8 years).

Of the 25 patients who agreed to participate in the assessment, three declined radiographs to avoid additional radiation, and nine could not perform gait analysis because they could not stay in Bologna for the time required for the analysis. Thus, our final patient group included 25 patients who underwent clinical assessment, 22 who underwent radiographic study, and 16 who underwent gait analysis. We found no general differences between patients who completed a full assessment and those who did not (Table 1).

No other specific inclusion or exclusion criteria were adopted, and geographic proximity to the center played a relevant role in the duration of a patient's stay in Bologna.

Twenty-five patients with a mean followup of 15 years (SD, 5.8 years; range, 3–22 years) after surgery agreed to participate in this study. The mean age of the patients was 23.8 years (SD, 7.5 years) at the time of evaluation and 9.3 years (SD, 3.1 years) at the time of surgery. Twenty-two patients had osteosarcoma and three had Ewing's sarcoma. All the patients completed chemotherapy and had no signs of recurrence.

Variables, Outcome Measures, Data Sources, and Bias

To answer the question whether the length of the surgically treated residual thigh-shank relative to the contralateral thigh resulted in an altered gait pattern, a physical examination was performed on the patients. Residual thigh-shank length was taken as the distance from the anterosuperior

Table 1. Patient characteristics

Variable	Total (n = 25)	With radiography (n = 22)	With gait analysis (n = 16)
Followup (years)	15 (6 [3–22])	15 (6 [3–21])	16 (5 [7–22])
MSTS score	24 (2 [20–26])	24 (2 [20–26])	24 (6 [21–26])
Length difference between the residual thigh-shank and the contralateral thigh (cm)	3 (4 [–10 to 8])	3 (4 [–10 to 8])	3 (4 [–10 to 8])

Mean (SD [range]); MSTS = Musculoskeletal Tumor Society

Fig. 2 An example of radiographic foot measurement is presented. The calcaneus long axis (Line A) is measured from the upper limit of the anterior joint surface to the tuberosity, passing the midpoint of line B, the calcaneus short axis. The talus long axis (line C) is measured from the center of the anterior joint surface to the posterior limit of the talus, and the talus short axis (line D) (the diameter of the head).

iliac spine to the ipsilateral external malleolus (former medial malleolus), and contralateral thigh length was the distance from the contralateral anterosuperior iliac spine to the lateral femur epicondyle.

To investigate the clinical and functional impairments of patients who underwent rotationplasty and who survived into adulthood, the MSTS score [6] (total score ranges between 0 and 30 [100%] with 0 indicating poor and 30 indicating excellent results), was obtained by clinical assessment and patient interview. Furthermore, possible foot deformities related to the use of the prosthesis were explored by means of radiography in 22 patients, for both sides in an unloaded condition. Bone density, joint space narrowing, and presence of osteophytosis and osteosclerosis for the tibiotalar, subtalar, and talonavicular joints were qualitatively assessed. The sizes of the talus and calcaneus were measured (long and short axes) (Fig. 2) for each bone by using image processing software (ImageJ Version 1.46r; National Institutes of Health, Bethesda, MD, USA). Measurements of foot length, active ankle ROM for both sides, and the presence of foot problems (eg, callosities, bursitis, presence of pain) were recorded. The foot length was taken from the posterior edge of the heel to the anterior edge of the hallux.

Finally, to see the differences between gait analysis findings in adulthood and previously reported findings in childhood, relevant gait parameters such as maximal ground reaction forces and sagittal angles at the knee were recorded in 16 patients. An eight-camera stereophotogrammetric system (Vicon® 612; Oxford Metrics, Oxford, UK) and two forceplates (Kistler, Winterthur, Switzerland) were used.

Statistical Analysis

To verify whether the length of the surgically treated limb influences spatiotemporal parameters of gait and knee kinematics, a hierarchical cluster analysis [2] was performed. Because the data set included 48 gait cycles (ie, three repetitions each of the 16 patients), a bias resulting from the patient “effect” was hypothesized. For this reason, a two-step clustering analysis was considered to be reliable and stable even for a data set with continuous variables. This analysis clusters the data set into many small subclusters, which then are collected in the desired number of clusters as explanatory variables with standardized Euclidian distance as a measure of the distance. Post hoc analysis of the clusters, corrected for multiple comparisons using Bonferroni’s method, was performed to identify the variables characterizing each cluster. Only significant variables for a probability less than 0.001 were considered.

Descriptive statistics were performed on anthropometric data and gait analysis results. The thigh length, foot length, and active ankle ROM were compared between limbs using a paired t-test. For gait analysis, numerous parameters were extracted from the patients’ gait variable time histories [3] in the surgically treated and contralateral limbs and compared with those from a control group of 20 healthy subjects (11 men and nine women; mean age, 28 years). One-way ANOVA and post hoc pairwise comparison by Scheffé’s test were used for parameters with homogeneous variances assessed by the Levene test; otherwise, the Mann-Whitney test with Bonferroni’s correction was performed. A significance level was set at 0.05 for all statistical comparisons. Data are presented throughout the text as mean and SD. All the statistical analyses were performed using SPSS 11.0 (SPSS Inc, Chicago, IL, USA).

Results

The residual thigh-shank was, on average, 5.8% longer than the contralateral thigh (mean, 52 cm [SD, 5 cm] vs 49 cm [SD, 3 cm]; $p < 0.001$). Two patients had a residual thigh-shank shorter (1 cm [2%] and 10 cm [18%]) than the contralateral thigh. The hierarchical cluster analysis performed to verify the effects of the residual thigh-shank length on gait identified two clusters (A = nine patients and B = seven patients) (Table 2), with no significant differences in age, followup, stance and swing time, normalized stride length, and walking speed. The residual thigh-shank (mean, 55 cm; SD, 4 cm; $p < 0.001$) and the contralateral thigh length (mean, 51 cm; SD, 2 cm; $p < 0.001$) characterized cluster A as significant variables ($p < 0.001$). The patients in cluster A also were characterized by pseudoknee

Table 2. Cluster analysis summary

Variable	Cluster A	Cluster B	Total	p values [†]	Control
Age (years)	24 (6)	25 (7)	24 (7)	NS/NS	NA
Age at intervention (years)	12 (9)	10 (2)	11 (7)	NS/NS	NA
Followup (months)	210 (49)	188 (69)	200 (59)	NS/NS	NA
Residual thigh-shank length (cm)	55 (4)*	48 (2)*	52 (5)	<0.001/<0.001	NA
Contralateral thigh length (cm)	51 (2)*	47 (1)*	49 (3)	<0.001/<0.001	NA
Length difference between the residual thigh-shank and the contralateral thigh (cm) [‡]	4 (5)	1 (1)*	3 (4)	NS/<0.001	NA
Stance duration (% stride)	59 (2)	59 (2)	59 (2)	NS/NS	60 (2)
Swing duration (% stride)	41 (2)	41 (2)	41 (2)	NS/NS	40 (2)
Stride length (cm)	145 (9)*	129 (9)*	138 (12)	0.03/<0.001	141 (9)
Stride length (% height)	84 (5)	84 (5)	84 (5)	NS/NS	82 (6)
Cycle duration (seconds)	1.2 (0.1)	1.2 (0.1)*	1.2 (0.1)	NS/0.001	1.1 (0.1)
Cadence (stride/minutes)	49 (3)	52 (3)*	50 (4)	NS/0.003	54 (4)
Walking speed (cm/seconds)	117 (12)	113 (6)	115 (10)	NS/NS	128 (11)
Walking speed (% height/seconds)	68 (7)	72 (4)*	70 (6)	NS/0.04	74 (7)
Knee flexion at initial contact (°)	12 (8)*	3 (4)	8 (8)	0.04/NS	5 (4)
Maximal knee flexion at loading response (°)	21 (12)	9 (8)*	16 (12)	0.05/<0.001	16 (6)
Minimal stance knee flexion (°)	13 (10)	4 (7)*	9 (10)	NS/0.008	4 (4)
Knee flexion at toe-off (°)	44 (10)	34 (8)*	39 (10)	NS/0.008	39.0 (6)
Maximal swing knee flexion (°)	79 (8)*	68 (3)*	74 (8)	0.002/<0.001	65 (6)
Knee flexion-extension range (°)	70 (11)	67 (3)	68 (8)	NS/0.05	65 (5)

Mean (SD); * values with significant difference; [†]cluster A versus total/cluster B versus total; [‡]positive value indicates a longer residual thigh-shank.

NS = nonsignificant; NA = not applicable.

flexion at initial contact (mean, 12°; SD, 8°; $p = 0.04$), during loading response (mean, 21° SD; 12°; $p = 0.05$), during swing (mean, 79°; SD, 7.7°; $p = 0.002$), and by stride length (mean, 145 cm; SD, 9 cm; $p = 0.03$). Cluster B was characterized by residual thigh-shank (mean, 48 cm; SD, 2 cm; $p < 0.001$), contralateral thigh length (mean, 47 cm; SD, 1 cm; $p < 0.001$), discrepancy between the residual thigh-shank and contralateral thigh (mean, 1 cm; SD, 1 cm; $p < 0.001$), knee flexion during loading response (mean, 9°; SD, 8°; $p < 0.001$), during terminal stance (mean, 4°; SD, 7°; $p = 0.008$), at toe-off (mean, 34°; SD, 8°; $p = 0.008$), and during swing (mean, 68°; SD, 3°; $p < 0.001$), stride length (mean, 129 cm; SD, 9 cm; $p < 0.001$), cycle duration (mean, 1.2 seconds; SD, 0.1 second; $p < 0.001$), cadence (mean, 52 strides/minute; SD, 3 strides/minute; $p = 0.03$), and walking speed (mean, 72% height/second; SD, 4% height/second; $p = 0.04$).

The mean value of the MSTTS score used for clinical functional assessment was 25 (SD, 2; 82%).

Pain had a mean value of 4.6 (SD, 0.7), function had a mean value of 4.6 (SD, 0.5), emotional acceptance had a mean value of 3.4 (SD, 0.8), support had a mean value of 5 (SD, 0), walking ability had a mean value of 4.9 (SD, 0.3),

and gait had a mean value of 4.4 (SD, 0.9) (Fig. 3). Sixteen patients could perform sports activities (two at a competitive level, swimming and cycling) and the other patients had restriction only in sports activities. The surgically treated foot was a mean of 10% (range, 0.5–5 cm) shorter than the contralateral limb. Active ankle dorsiflexion ROM of the surgically treated side was 105% smaller (difference, 7.2°; SD, 4.8°) than in the contralateral ankle, whereas active plantar flexion ROM was not different, with the numbers available, from the contralateral ankle. The talus was 11% smaller (mean difference, 0.9 cm; SD, 0.6 cm) on the surgically treated side than on the contralateral side in the long axis, and 7.6% (mean difference, 0.4 cm; SD, 0.3 cm) on the short axis. The calcaneus was 2.7% smaller (mean difference, 0.2 cm; SD, 0.3 cm) on the surgically treated side than on the contralateral side in the long axis and 8.6% (mean difference, 0.3 cm; SD, 0.3 cm) on the short axis (Table 3). For prosthetic use, six of the 25 patients had callosities at sites around the rotated foot such as the external edge of the foot, malleolar region, digits, and dorsal tarsus. One had bursitis at the base of the first metatarsal. No patient reported having foot pain. All 22 patients who had radiographic evaluations showed joint

Fig. 3 The mean and SD values of the individual items of Musculoskeletal Tumor Society (MSTS) score for the patients after knee rotationplasty are shown.

Table 3. Anthropometric evaluation

Foot measures	Surgically treated	Contralateral	p value*
Foot length (cm)	22 (2 [19–29])	24 (2 [21–30])	<0.001
Foot dorsiflexion ROM (°)	0 (5 [–10 to 0])	7 (7 [–10 to 10])	<0.001
Foot plantar flexion ROM (°)	59 (8 [40–70])	61 (5 [50–60])	NS
Calcaneus long axis (cm) [†]	8 (1 [7–9])	9 (1 [7–1])	<0.001
Calcaneus short axis (cm) [†]	4 (0 [4, 5])	5 (1 [4–6])	<0.001
Talus long axis (cm) [†]	6 (1 [4–6])	6 (1 [5–7])	0.004
Talus short axis (cm) [†]	3 (0 [2, 3])	3 (0 [2–4])	<0.001

Mean, SD, and range are presented; * paired t test; [†]n = 22; NS = nonsignificant.

space narrowing on the surgically treated foot (nine patients for the tibiotalar joint, 17 patients for the subtalar joint, 21 patients for the talonavicular joint) and osteopenic appearance of the foot bones. In the unloaded condition, the metatarsophalangeal joints were observed to have a different spatial orientation over the feet, oriented in plantar flexion on the surgically treated side in most of the patients, but positioned in dorsiflexion at the contralateral side (Fig. 4).

The mean walking speed of the patients (mean, 1.2 m/second; SD, 0.1 m/second) was slower than that of the controls (mean, 1.3 m/second; SD, 0.1 m/second; $p < 0.001$) as was cadence (mean for surgically treated side, 50 strides/minute [SD, 4 strides/minute]; mean for control subjects, 54 strides/minute [SD, 4 strides/minute], $p < 0.001$), whereas the stride length was not different, with the numbers available, between the groups (mean for surgically treated side, 1.4 m [SD, 0.1 m]; mean for the contralateral side, 1.4 m [SD, 0.1 m]; mean for controls, 1.4 m [SD, 0.1 m]). Analysis of the ground reaction forces

(Table 4) showed that the first peak of the vertical component was significantly greater ($p = 0.03$) on the contralateral side of the patients (mean, 113% body weight [BW]; SD, 11% BW) than in the controls (mean, 108% BW; SD, 11% BW) whereas the second peak was reduced ($p < 0.001$) on the surgically treated side (mean, 103% BW; SD, 9% BW) with respect to controls (mean, 118% BW; SD, 9% BW). The minimum of the vertical component at midstance was not different, with the numbers available, between the patients and the controls (mean for surgically treated side, 79% BW [SD, 5% BW]; mean for contralateral side, 79% BW [SD, 6% BW]; mean for controls, 78% BW, [SD, 6% BW]). Patients exhibited greater flexion of the pseudoknee at initial contact (mean for surgically treated, 8° [SD, 8°]; mean for controls, 5°, [SD, 4°]; $p < 0.01$), at terminal stance (mean for surgically treated, 9° [SD, 10°]; mean for controls, 4° [SD, 4°], $p = 0.006$) and during swing (mean for surgically treated, 74° [SD, 8°]; mean for controls, 75° [SD, 6°]; $p < 0.001$). The contralateral knee in patients treated surgically extended more

than that of the control subjects at initial contact (mean, -2° [SD, 4°]; mean for controls, 5° [SD, 4°]; $p < 0.001$), at terminal stance (mean, $^\circ$ [SD, 5°]; mean for controls, 4°

[SD, 4°]; $p < 0.001$), and during swing (mean, 63° SD, 5°); mean for controls, 75° [SD, 6°]; $p = 0.03$) (Table 4).

Discussion

Knee reconstruction with an expandable prosthesis is performed today for most skeletally immature patients with a bone sarcoma around the knee. However, when an intraarticular distal femoral resection is used in children younger than 5 years, or when the tumor involves the whole anterior compartment of the thigh, or there is substantial neurovascular involvement in older children or adults, rotationplasty is the preferred choice as it may offer functional advantages over transfemoral amputation and more durable results than a prosthesis. Numerous studies have explored the clinical and functional outcomes in patients who had a knee rotationplasty, using the MSTs, clinical and radiographic assessment, and gait analysis (Table 5). However, the effect of length of the residual limb in adulthood on gait and on clinical and functional outcomes was not explored in any of the studies. Because some patients have reported problems in walking or in daily life owing to different alignment of the knees, we aimed mainly to explore objective relationships between the residual limb length at the end of growth, gait performance, and clinical and functional outcomes. We found that the residual-thigh shank length is associated with limb loading and knee kinematics during walking.

Our findings indicated that the residual thigh-shank in adulthood is on average 5.8 cm longer than the contralateral thigh. Considering the residual potential growth of the contralateral femur and expected smaller growth of the

Fig. 4A–B Radiographs of the foot and ankle of one of our patients (29 years old) who underwent rotationplasty at age 12 years are shown. **(A)** On the surgically treated side, narrower joint spaces, smaller bone sizes, an osteopenic appearance, and plantar flexed digits are seen compared with **(B)** the contralateral side.

Table 4. Ground reaction forces and knee kinematics

Variables	Surgically treated	Contralateral	Control	p value (versus control) [†]	
				Surgically treated	Contralateral
Ground reaction forces (% body weight)					
First peak in vertical force	106 (7 [89–119])	113 (11 [91–147])	108 (11 [83–134])	NS*	0.03
Second peak in vertical force	103 (9 [79–119])	114 (11 [93–138])	118 (9 [100–147])	<0.001	NS*
Minimum vertical force during midstance	79 (5 [69–92])	79 (6 [67–91])	78 (6 [60–91])	NS	NS
Maximum aft force	16 (5 [6–26])	21 (5 [13–37])	20 (5 [13–34])	<0.001	NS
Maximum fore force	17 (5 [7–25])	25 (5 [15–36])	24 (4 [17–35])	<0.001*	NS
Knee kinematics (°)					
Knee flexion at contact	8 (8 [–8 to 20])	–2 (4 [–9 to 9])	5 (4 [–1 to 12])	0.01*	<0.001
Maximum knee flexion at loading response	16 (12 [–6 to 33])	17 (7 [–1 to 27])	16 (6 [4–30])	NS*	NS*
Minimum stance knee flexion	9 (10 [–6 to 28])	1 (5 [–6 to 12])	4 (4 [–5 to 13])	0.006*	<0.001*
Maximum swing knee flexion	74 (8 [63–94])	63 (5 [47–69])	65 (6 [50–79])	<0.001*	0.03
Knee flexion–extension range	69 (8 [50–88])	68 (6 [48–82])	65 (5 [54–75])	0.04*	0.01

Presented as mean, SD, and range; * Mann-Whitney U-test with Bonferroni correction; [†]parameters with no sign indicate the use of one-way ANOVA with post hoc Scheffé's test; BW = body weight; NS = nonsignificant.

Table 5. Summary of nononcologic outcome studies for patients after knee rotationplasty

Study	Number	Followup (years)*	MSTS score†	Other main evaluations
Agarwal et al. [1]	27	3 (2–6) ‖	Not available¶	
Catani et al. [5]	10	2 (1–4)	Not available	Gait analysis
Fuchs et al. [8]	7	8 (4 to 16)	25 (84%, 22–27)	Activities, gait analysis
Gebert et al. [10]	21	14 (10–25)	Not available	Activities, foot condition, radiography (qualitative), MRI§
Ginsberg et al. [11]	4	Not available	28 (92%, 2)	Functional abilities, activities, HRQoL
Gottsauner-Wolf et al. [12]	60	5 (3)	Not available	Physical examination§, radiography (qualitative)§, prosthesis fitting§, questionnaire‡
Hanlon and Krajchich [13]	21	8 (4–11)	Not available	MSTS score (previous version)§, activities, radiography (qualitative)§
Hillmann et al. [14]	33	6 (2–16) ‖	24 (80%)	HRQoL
Hillmann et al. [15]	43	7 (1–18)	24 (79%, 3)	Gait analysis
Hillmann et al. [16]	14	10 (3–19)	24 (79%, 4)	Foot condition and loading
Hillmann et al. [17]	61	7 (5)	Not available	Sports participation, aerobic capacity§
Hopyan et al. [18]	5	9 (5–14)	20 (67%, 18–22)	Activities
McClenaghan et al. [22]	8	Not available	Not available	Gait energetics
Rodl et al. [24]	22	12 (10–18)	Not available	HRQoL, life contentment
Steenhoff et al. [26]	8	2 (1–5)	Not available	Gait analysis
Veenstra et al. [28]	33	6 (1–11)	Not available	HRQoL
Forni et al. [7] ‡	20	17 (4)	Not available	HRQoL, interview§
Current study	25	15 (3–22)	25 (82%, 20–26)	Physical examination, gait analysis‡, foot condition, radiography (quantitative)§

* Mean (SD or range); † mean (percentage of maximal score, SD or range); ‡ included the patients in the current study; ‖ median (range), included patients who underwent other type of surgery; § data available for part of total patients; ¶ evaluated but representative scores not available; MSTS = Musculoskeletal Tumor Society; HRQoL = health-related quality of life.

rotated distal tibia, usually the surgically treated leg is made to be longer than the contralateral thigh in skeletally immature patients at surgery. Nevertheless, another factor may influence postoperative thigh length: in case of a very large tumor that requires subtotal resection of the femur, a longer surgically treated leg cannot be achieved. Although the final surgically treated residual thigh-shank length after growth cannot be fully predicted at the time of surgery because of several uncontrolled factors, a great discrepancy between the residual thigh-shank length and the contralateral thigh could be responsible for gait alterations. Exploratory cluster analysis showed that whereas no MSTS, age, and sex-related parameters differed between the two clusters, the patients in cluster A (less successful procedures) had longer contralateral and surgically treated limbs and a higher discrepancy between the residual thigh-shank and the contralateral thigh length. These patients walked with knee kinematics different from those of control subjects. Patients in cluster B (more successful procedures) had shorter contralateral and surgically treated limbs, a smaller discrepancy between the surgically treated

residual thigh-shank and the contralateral thigh length, and knee kinematics closer to those of control subjects. Similarly, the patient with the shortest residual thigh-shank length (10%), in the full gait cycle, had complete loss of the phase of knee loading absorption with a stiff-legged pseudoknee during stance (Fig. 5). From the findings in our study, we speculate that patients with less difference between residual thigh-shank and contralateral thigh length can walk in a more physiologic and efficient way. Additional studies are required to verify these findings. In particular, assessment on standing scanograms of early postoperative residual limb length and the pattern of growth of the surgically treated limb with time would be worth investigating and of interest for clinicians.

In our study the MSTS score generally was not different from the reported values for patients at similar followup, ranging from a mean of 20 to 28 [8, 11, 14–16, 18] (Table 5). In particular, the MSTS score was not different from the only study reporting the score at a short followup [11]. Furthermore, the MSTS score was similar in the two clusters with respect to the residual thigh-shank length,

Fig. 5A–B (A) The graph shows the patient's knee flexion and extension angles for the surgically treated leg. The red lines indicate the values for the surgically treated leg. The green lines indicate the values for the contralateral limb. The gray solid and dotted lines

indicate the mean and SD values for the control subjects. (B) The graph shows the patient's knee flexion and extension angles for the surgically treated leg. The gray solid and dotted lines indicate the values for the control subjects.

possibly because this instrument is a comprehensive evaluation of the entire extremity and patient as a whole [6]. The reduced surgically treated side foot length and dorsiflexion were consistent with previous findings [12], accounting for the altered loading condition inside the prosthesis [10]. Reduced foot length was partly attributable to smaller foot bones; patients who underwent surgery at an older age seemed to have less difference. Narrowed joint spaces and signs of osteosclerosis were present in the foot, with greater prevalence at the subtalar and talonavicular joints, but were not symptomatic. This is in agreement with previous findings of nonsymptomatic degenerative changes in the rotated ankle and foot bones [10, 12] but with time, could account for a reduction of ROM of the entire foot, limiting the pseudoknee flexion during gait, and contributing to possible gait performance deterioration. In this respect, older patients have been shown to have reduced ROM, especially ankle ROM, with consequent restriction of daily activities [10].

Finally, in line with previous studies [5, 8], our patients walked slower than the control subjects mainly owing to reduced cadence, and with the pseudoknee relatively flexed during the gait cycle. The decreased propulsive force of the surgically treated side and loss of original function of the gastrocnemius, which facilitates swing initiation [25], are responsible for the smaller cadence, resulting in slower walking speed [8]. Compared with the only previous short-term study reporting values at a mean of 24 months followup [5], our patients walked with a better vertical force pattern during midstance and with increased knee flexion-extension ROM. However, a difference in ground reaction forces between the surgically treated and contralateral sides was still present. Overloading of the healthy lower limb and offloading of the prosthetic lower limb confirm findings described in patients after amputation [4, 27]. The prosthesis does not

fully replace the function of the gastrocnemius, which normally generates energy to the leg to initiate swing [27]. The power provided by the prosthetic limb during gait has been found to be minimal, this being made possible by the highly efficient energy-saving, pendulum-like oscillation over the prosthetic limb, based on a “learned nonuse” and contralateral compensation pattern [27]. Additional studies are needed to support this biomechanical hypothesis in patients after rotationplasty.

Our study has some limitations. First, a selection bias could have had an effect on findings. Not all the patients who underwent rotationplasty at our institute participated in the study, therefore the findings are representative only of the patients assessed. An additional selection bias is that, of the 25 patients included in the study, only a subgroup of 16 patients had gait analysis and 22 had radiography. The number of patients included in the study and in the subgroups was a function of geography that determined who did or did not have gait analysis or radiographs, and not a patient-selection process. In addition, the wide range of the followup and inclusion of some patients with short-term followups (less than 5 years) could have had some effect on findings. However, the MSTs score, mean followup, and mean length difference between thighs in the three subgroups were not different. Second, performing exploratory multiple comparisons and cluster analyses, we should be aware of an increased false-positive rate and compromised statistical power. We carefully inspected each value when focusing on equality between the groups. Third, alignment of the prosthesis and anatomic differences between the pseudoknee and the contralateral knee might cause offset on knee rotations on the surgically treated side as measured by gait analysis with marker positioning. However, the increased ROM of the surgically treated side knee during walking compared well with that of control subjects despite this possible offset.

Patients with knee rotationplasty in adulthood have, on average, a longer residual thigh-shank which is related to the gait pattern: patients with better matching knee height had a gait pattern closer to that of control subjects. Early functional results, as measured by the MSTS score for function and pain, appear to be maintained in early adulthood, at a mean of 15 years after surgery. Gait analysis, in terms of ground reaction forces and knee ROM, was improved in adulthood although a difference in limb loading was still present. Based on these findings, surgeons should endeavor to have the center axis of rotation of the contralateral knee and the pseudoknee at skeletal maturity. An excessive residual thigh-shank length in adulthood could require contralateral lengthening to improve functional results.

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