**When your ankle becomes a knee - a clinical study on long-term functional outcome and quality of life with a rotationplasty after resection of malignant limb tumors**

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| **Abbreviation** | **Full term** |
| SF-36 | Short-form-health survey 36 |
| MSTS | Musculoskeletal tumor society score |
| ROM | Range of motion |
| HRQL | Health related quality of life |
| COSS | Cooperative Osteosarcoma Study Group |
| VT | Vitality |
| PF | Physical functioning |
| BP | Bodily pain |
| GH | General health |
| PR | Physical role functioning |
| ER | Emotional role functioning |
| SR | Social role functioning |
| MH | Mental health |
| PCS | Physical component summary score |
| MCS | Mental health component summary score |
| EPR | Endoprosthetic replacement |

Abstract

**Background**

Increasing numbers of patients surviving malignant bone tumors around the knee joint have led to an increasing importance to investigate long-term results. This study assessed the long-term results of rotationplasty after resection of malignant bone tumors regarding functional outcome and quality of life to allow better comparison with other treatment options in bone cancer treatment.

**Procedure**

60 participants who underwent rotationplasty due to bone cancer took part in this multicentric questionnaire-based study. The long-term functional outcome was measured by the Musculoskeletal tumor society score (MSTS) and the Tegner activity level scale. The health-related quality of life (HRQL) was assessed by using the Short Form Health Survey (SF-36).

**Results**

Patients treated with rotationplasty (median follow-up of 22 years, range 10-47 years) regained a high level of activity (median MSTS score of 24). Even a return to high level sports was possible (mean Tegner activity level scale of 4). Duration of follow-up did not influence the functional outcome. HRQL scores were comparable to the general German population. Concerns of psychological problems due to the unusual appearance of the rotated foot have not been confirmed.

**Conclusion**

Rotationplasty can be a good alternative to endoprosthetic replacement or amputation, either as primary surgery or as a salvage procedure. Especially for growing children and very active patients rotationplasty should be considered.

**Introduction**

The most common location for bone tumors is the knee region. Especially osteosarcoma is the most common primary malignant bone tumor in children with an incidence of 0.3 per 100,000 per year1 2 3.

Progress in malignant bone tumor treatment including neoadjuvant and adjuvant therapy has entailed in an increasing number of patients surviving malignant bone tumor4.

This increasing number of survivors leads to a rising importance to assess the long-term results of the treatment options regarding functional outcome and quality of life. The minority of patients obtain primary amputation or rotationplasty. Nowadays endoprosthetic knee replacement is the treatment of choice1. It is a difficult decision for doctors, patients and parents to choose the type of surgery that will combine maximum chances for cure with an acceptable cosmetic outcome, high functional outcome and quality of life5.

Limb salvage procedures like endoprosthetic replacement (EPR) are associated with a good cosmetic and functional outcome6 but they are also associated with a high failure rate, so patients face the risk of many future surgeries7 5. In comparison, various studies have also proved high functional outcomes after rotationplasty8 9 10 11 12 but there are often concerns about the psychosocial impact of the unusual limb appearance13 14 15.

Therefore, the aim of this study was to assess the long-term results of rotationplasty after resection of malignant bone tumors regarding quality of life and functional outcome to allow better comparison with other treatment options in bone cancer treatment. These results may help physicians, parents and patients choosing the option of surgery16.

Patients and Methods

Patient recruitment

135 patients who underwent rotationplasty due to a bone sarcoma around the knee were identified from the database of the Cooperative Osteosarcoma Study group of the German Society for Pediatric Oncology and Hematology (COSS). The last known treating physicians of these patients were contacted by COSS and were asked to inform their patients about the study.

A letter that introduced the study, its purpose, a link to the questionnaire and the eligibility to participate were sent to the patient by the treating physician. The questionnaire could be opened and answered via the link in the letter or via QR-code. The questionnaires were completed by participants as self-evaluation.

Informed consent was obtained from each patient. The study was approved by the local Ethical Committee (Universitätsklinikum Frankfurt am Main, Nr. 22/19) and was carried out according to the data protection law of the EU.

The patients had to fulfil the following inclusion criteria: (1) undergone rotationplasty at least 10 years prior, (2) be at least 18 years at time of study participation, (3) informed consent.

We received 72 answered questionnaires.12 questionnaires were excluded from this study: Six did not fulfill inclusion criteria, one patient received secondary amputation and five duplicates were found. Finally, a total of 60 patients were included in this study (Fig. S1). The other invited patients did not respond. However, there are no references whether they failed to answer due to outdated information of phone numbers, postal address or any other reasons.

**Questionnaires**

The patients clinical and demographic characteristics were assessed by the questionnaire. The functional outcome was ﻿assessed by the ﻿Musculoskeletal tumor society score (MSTS)17 and the Tegner activity level scale 18.

The MSTS-Score consists of three general components (pain, function, emotional acceptance) and three components specific to the affected extremity (use of external supports, walking ability and gait). Each item ranges from 0 to 5 points, so the maximum total score is 30 points which indicates a high level of function17. However, this score does not regard the level of sports activities.

Therefore, the Tegner activity level scale was used as a second device. This score rates activity based on sports activities and work on a scale from zero to ten18. Ten points indicates a maximum activity level while an activity of zero represents invalidity or limited ability19.

﻿﻿Health related quality of life (HRQL) was assessed by using the﻿ Short Form Health Survey (SF-36). The SF-36 quantifies the individual health just as disease related quality of life. It contains 36 items with eight multi-item subscales: physical functioning (PF), physical role functioning (PR), bodily pain (BP), general health (GH), vitality (VT), social role functioning (SR), emotional role functioning (ER), mental health (MH) 20. Each scale ranges from 0-100. These eight subscales can be scored independently or can be transformed into two summary scores: ﻿Physical component summary score (PCS) and Mental health component summary score (MCS). The PCS includes PF, PR, BP, GH and the MCS includes VT, SR, ER, MH 21 22.

**Statistics**

Statistical analyses were performed using R (Version 4.0.2) and SPSS (Version 26.0, SPSS Inc.).

Categorical variables were summarized as the frequency and percentage and numerical variables as median (range), first and third quartiles (Q1, Q3) and mean (SD).

Numeric variables: age at surgery, BMI and time since surgery were categorized in two groups according their median. Furthermore, age was categorized in five groups: 18-29, 30-39, 40-49 and 50-59 years. These variables were analyzed as numeric and category.

The data was analyzed to determine Gaussian distribution using visual methods such as qq-plots, histograms, boxplots and violin plots as well as the Wilk-Shapiro test.

The differences of the MSTS-score, Tegner activity level scale or SF-36-score between the sociodemographic/clinical variables or impairments groups were explored. Due to the non-normal distribution of the scores, the differences were assessed using the Mann-Whitney test or the Kruskal-Wallis test, as appropriate. The Spearman’s rank correlation was employed to analyze the association between the scores and numerical variables.

The differences of SF-36 scores between our patient cohort and the general German population were assessed with the one sample median test. The mean value was used as the median for the reference group, since both are equal in normally distributed data.

All significance tests were two-sided and the 5% significance level was used to denote statistical significance. No statistical adjustment was made for the present analysis.

**Results**

**Participant characteristics**

60 participants who underwent rotationplasty surgery due to a bone sarcoma around the knee fulfilled the inclusion criteria and participated in this study. The median time from surgery to study participation (follow-up) was 22 years (range 10-47 years).

Table 1 presents the socio-demographic and clinical characteristics. 32 participants (53%) were male and 28 (47%) were female. Median age at study participation was 36 years (range 19-56) and median age at time of surgery had been 12 years (range 3-36 years).

Most of the patients (52%) received a type-A1 rotationplasty ﻿according to the classification described by Winkelmann23 24. The most common side was the right leg (60%). 52 participants (86%) reported a range of motion (ROM) >60° of the ankle joint. 95% wore their exoprosthesis the whole day. Impairments due to the exoprosthesis are shown in Table 1.

**Functional outcome**

The average MSTS score was 23 points (median = 24). Figure 1 shows the results of the MSTS score and components. 48 patients (80%) reported no or intermediate pain. Two (3%) patients had modest, not restrictive pain without using opioids (Fig. S2).

While 12 patients (20%) were not restricted during daily activities, 25 patients (42%) reported limitations during recreational activities or negligible disability. Limitations in activities related to work were recorded in four cases (7%).

Regarding emotional acceptance, an overall satisfaction with rotationplasty was found. 88% would choose rotationplasty as treatment option again.

92% didn´t require any assistive devices. 7% used occasionally an assistive device in addition to the exoprosthesis.The reason for and the frequency of using this assistive device is unknown.

While 12 patients (20%) were not limited in the distance they could walk, 20 (33%) had the impression that their walking ability was less than prior to surgery.

Most of the patients reported no (8%), intermediate (22%) or minimal alterations (33%) in their gait pattern. An obvious alteration in gait pattern with an impact on the function was reported by 9 patients (15%).

**MSTS by socio-demographic and clinical characteristics**

A significant relationship between the functional outcome and the patient´s age was found (Table 2). Older participants reported suffering significantly more pain than younger participants (p=0.035) and they also reported less ability to walk (p=0.006) (Fig. S3). Emotional acceptance was significant associated with sex, age at surgery and ROM of the ankle joint. Females reported lower emotional acceptance than males (p=0.014) (Fig. S4). Participants of older age at time of surgery declared worse emotional acceptance than younger participants at time of surgery (p=0.027). Finally, high ROM of the ankle joint (ROM) was reflected in a high emotional acceptance (p=0.011) and better gait scores (p=0.011) (Fig S4).

**MSTS by impairments**

Associations between MSTS and impairments were found (Table 2). Participants with limited mobility of the ankle joint had significant lower scores in all MSTS components except pain and walking ability.

Furthermore, problems with the contralateral leg were associated with lower total MSTS and components (except emotional acceptance and walking support).

Participants who reported swelling had also significant lower MSTS components (function, gait, total MSTS).

**Tegner activity level scale**

With a mean Tegner activity level scale of 4, the patients were regularly engaged in sports activities. 41 (69%) reported that they were active in sports. Eleven of the 41 took part in competitive sports (19%), three of them (5%) at an international level. 19 patients (32%) were not engaged in sports activities at all (Fig. 2).

Younger patients had a significant higher Tegner activity level scale (p=0.099) (Fig. S5). Furthermore, age at time of surgery was associated with the Tegner activity level scale, where younger age at time of surgery showed higher scores (p=0.099).

**Quality of life**

The medians for the summary scores were 52 (range 20-60) for the physical component summary score (PCS) and 52 (range 29-61) for the mental health component summary score (MCS). Figure 3 presents the results of the subscales.

Analysis of the HRQL showed that participants in the age group 18-29 had significant higher scores regarding the subscales PR, BP, VT, MCS but significant lower scores in the subscale PF than the German general population 25 26 (Table S1). In the older age groups (30-39, 40-49) a significant lower PF subscale as well as a significant lower PCS subscale in the age group 40-49 was found.

**Quality of life by participant characteristics**

Age as well as time since surgery shown affected the HRQL components (Table 3). Older patients and patients with a longer time since surgery had lower scores for the subscales: PR, BP, GH, SR, PCS (Fig. S6). A high ROM of the ankle was associated with a high score for the subscale PF. Additionally, it has been shown that there is no difference regarding the HRQL-scores between genders.

**Quality of life by impairments**

Participants with limited mobility of the ankle had significant lower scores for the subscales: VT, ER, MH, MCS and participants who reported arthrosis of the ankle showed significant lower scores for the subscales: GH, VT, SR and PCS (Table 3).

Moreover, participants suffering from problems with the contralateral leg had significant lower scores for the subscales: PR, BP, SR, ER and PCS.

**Discussion**

This multicentric study provides that patients treated with rotationplasty can regain a good functional outcome with a quality of life which is comparable to the German population.

The study of 60 such patients gives an overview about the long-term results of patients treated with rotationplasty for malignant bone tumors.

Even if malignant bone tumors around the knee are usually treated with EPR, other treatment options need to be considered. While amputation results in loss of the entire limb, EPR is associated with high failure rates and postoperative complications like aseptic loosening or ﻿periprosthetic infections27 28. El Dalil et al. reported that 42% of the patients who were treated with EPR suffered from post-operative complications compared to 25% of the patients who were treated with rotationplasty27. The most frequent reason for complications were infection and mechanical complications 7. They also noted that the risk of complications increased appreciably with revision surgeries7. Besides there is yet an uncertain prognosis for the afflicted limb and patients face to risk many future surgeries which perhaps result in an amputation29 7 30 31. Therefore, further long-term observations are necessary.

In comparison to EPR, rotationplasty is associated with a ﻿relatively low incidence of late complications11 29. Sawamura et al. reported that in 88% of the patients in this sample, rotationplasty was successfully performed. Early post-operative complications were vascular compromise and transient nerve palsy. Late complications were wound complications and fracture32. The present study has shown the following long-term complications: skin alterations, problems with the contralateral leg, lymphoedema and arthrosis of the ankle. Skin alterations caused by mechanical irritation were especially observed in patients who were physically active and participated in extended sports activities (Table 1). Nearly all of the patients reported that these skin alterations were related to a not appropriately fitting exoprosthesis and could be reduced by improving the exoprosthetic fit33.

Arthrosis of the ankle joint was reported in older participants with a mean follow up of 31 years (range 19-38 years). Even in previous studies, morphologic changes of the rotated ankle were noted33 34. Gebert et al. reported non-symptomatic radiologic changes as a result of an adaption process of the rotated ankle which does not compulsorily lead to arthrosis of the ankle joint33 but could lead to a limited neo-knee flexion due to a reduction of the ROM of the ankle joint. These morphologic changes and decrease of ROM were especially found in older patients34.

Two patients reported lymphoedema of the ankle which is treated with compression stockings and 23% of the patients reported problems with their contralateral leg. Referring to this, hip pain and back pain were mentioned. Some patients assumed that these problems were related to an overload of the contralateral leg.

Regarding these findings, it has been shown that differences in the length of the rotated distal tibia in relation to the contralateral femur resulted in altered gait pattern. This length difference was associated with differences in limb loading and knee kinematic, so patients with less length discrepancy had a more physiologic gait pattern34. The present study examines that the participants who reported problems with their contralateral leg also reported changes in their gait pattern. Therefore, we suppose that these patients may have a length difference which seems to affect the walking performance and causes problems with their non-affected limb. In our cohort, problems with the contralateral leg were predominantly found in women and patients with rotationplasty on the left side. Patients with these impairments had significant lower HRQL-scores.

Nevertheless, these participants were satisfied with their walking ability with a mean MSTS score of four points. Catani et al. reported that patients with rotationplasty walked faster and more efficiently than patients with above- knee amputation35. No differences between patients with an EPR or a rotationplasty were found9. Moreover, the maintenance of the proprioceptive feedback of the foot results in a better active (neo-)knee control compared to an above-knee amputation. This allows a smooth and coordinated gait pattern which is similar to the gait of a non-affected person36 37 10. Thus, complex movements like walking stairs were easier to perform37 and even car driving with the prosthetic leg was possible. 65% of the responders could drive a normal car, while 27% needed a converted car (Table 1).

Overall, the participants of this study had a good restitution of function after rotationplasty with a mean total MSTS of 23 points (Figure 1). These results were comparable to patients treated with EPR 5 38 39 40. Hillmann et al. compared patients who were treated with rotationplasty and EPR due to malignant bone cancer. Patients treated with rotationplasty had a mean MSTS score of 24 points and patients treated with EPR had a mean MSTS score of 25 points8. Stevens et al. and Akahane et al. reported superior functional results for rotationplasty compared to EPR and amputation5 38. This indicates that rotationplasty could achieve equal or superior functional results8 38 5.

Besides, it has been shown that the age at time of surgery influences the functional outcome positively. Hillmann et al. reported that younger patients had better scores for walking ability, gait and total MSTS12 which indicates that younger patients were able to adapt to their new situation in a better way12. This is consistent with our results. In the present study, younger patients had higher scores regarding the functional outcome and the HQRL (Table 2 and 3). Moreover, we noted that patients with a younger age at time of surgery had higher scores for the Tegner activity level scale. In our cohort, 69% were engaged in sports activities. 19% participated in competitive sports and 5% did so even at an international level (Fig. 2). Several participants were engaged in more than one sports activity and a variety of different sports were reported. This seems to be consistent compared with the work of Hillmann et al. who reported that patients with rotationplasty can re-engage in a high level of sports41. We also noted that it is possible to return to high-impact activities like skiing, running and handball. Most of the patients (73%) used their normal prosthesis for sports. 18% used an individual sport prosthesis and 8% did sports without their prosthesis. Sports activities that are practiced without prosthesis were skiing, sitting volleyball and swimming. In comparison, Hobusch et al. reported that patients with EPR mostly returned to low impact sports activities only 42 because they try to protect their EPR from a revision operation or an activity associated injury43.

Nevertheless, rotationplasty is currently performed rarely. There are often concerns that the appearance of the rotated foot may cause psychological problems14 15 8 44,but various published reports noted good psychologic adjustment after rotationplasty8 45 14 46: Veenstra et al. reported no significant differences in psychosocial functioning, quality of life and social support compared to healthy peers45. Also, the study of Rödl et al. noted no psychosocial problems, especially for the items partnership and sexuality 14. This is consistent with our findings. Most of the participants were not disturbed by the appearance of the limb. Except three, all participants would choose a rotationplasty as treatment option again. In our experience, patients benefited from visiting ex-patients so that they become aware of the initially unusual appearance of the rotated foot.

With respect to HRQL,we ascertained that patients in the age group 18-29 had higher scores in all subscales except physical functioning (PF) and physical component summary score (PCS). The older age groups (age group 40-49 and 50-59) had higher scores with respect to all subscales except PF, PR, GH, SR and PCS. The differences were particularly significant in the age group 18-29. The median scores of the SF-36 compared to the reference scores are presented in Table S1 26 25. Our finding concerning HRQL seems to be in line with previous studies among patients with bone cancer. Stevens et al. also reported significantly lower scores concerning the physical functioning scores compared to the healthy peers47 and various earlier studies consequently reported lower scores regarding physical functioning and similar or higher scores regarding mental functioning compared to the normal population47 45 48 14.

Akahane et al. and Stevens et al. compared the HRQL of patients treated with EPR, amputation and rotationplasty due to bone sarcoma. While Akahane et al. referred that all the scores of patients with rotationplasty were superior compared to EPR and amputation38, Stevens et al. reported no differences between rotationplasty, EPR and amputation5.

The similar or higher HRQL- scores except physical functioning of cancer patients compared to healthy peers has been described in previous studies49 50. This phenomenon which is called “response shift”50 reveals that life-threatening diseases can generate positive effects.

However, we recognize the following limitations in this study. First, this is a survey study. The questionnaires were completed by participants as self-evaluation.

Secondly, malignant bone tumors are very rare which limited the overall number of patients. In addition, we had difficulties to contact patients treated ages ago. The patients were lost to follow up or died from their underlying malignant tumor. Thirdly, there was no suitable control group for direct comparison.

In conclusion, the role of rotationplasty in the treatment of malignant bone cancer has to be reassessed. In our cohort, rotationplasty was not associated with any long-term disadvantages regarding functional outcome or HRQL. It can be a good alternative to endoprosthetic replacement or amputation, either as primary surgery or as a salvage procedure after failure of endoprosthetic replacement23 8 29 51. Especially in growing children and in very active patients, rotationplasty should be considered when choosing the optimal treatment option. Facing the surgical decision, patient education which may include meeting ex-patients may be useful to choose their individual treatment plan.

**Conflict of Interest**

The authors declare that there is no conflict of interest.

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**Data Availability Statement**

Data available on request due to privacy/ethical restrictions.

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**Legends**

**Table 1.** Participant and surgery related characteristics (n=60)

Abbreviations: ROM, range of motion; BMI, body mass index; Q1,Q3, first and third quartiles; SD, standard deviation.

**Table 2.** Significant associations between the MSTS score and participant’s characteristics or impairments.

Abbreviations: ROM, range of motion.

For numeric values, ↓ indicates high value of characteristics or impairments associated with low score; ↑ indicates high value of characteristics or impairments associated with high score.

For categorized variables, ↓ indicates reference level associated with low score.

\* p ≤0.05

\*\* p≤0.01

\*\*\* p≤0.001

**Table 3.** Significant associations between the SF-36 score and participant’s characteristics or impairments

Abbreviations: ROM, range of motion; BMI, body mass index; PF, physical functioning; PR, physical role functioning; BP, bodily pain; GH, general health; VT, vitality; SR, social role functioning; ER, emotional role functioning; MH, mental health; PCS, Physical component summary score; MCS, Mental health component summary score

↓ indicates high value of characteristics or impairments associated with low score.

↑ indicates high value of characteristics or impairments associated high score.

\* p ≤0.05

\*\* p≤0.01

\*\*\* p≤0.001

**Figure 1.** Results of the total MSTS score and components of the overall cohort. A higher score represents a higher functional outcome.

Abbreviations: Q1, Q3, first and third quartiles; SD, standard deviation.

**Figure 2.** Distribution of the Tegner activity level scale (Tegner Score). A higher score represents a better functional outcome and a higher activity level.

Abbreviations: Q1, Q3, first and third quartiles; SD, standard deviation.

**Figure 3.** Results of the SF-36 questionnaire of the overall cohort.

Abbreviations: PF, physical functioning; PR, physical role functioning; BP, bodily pain; GH, general health; VT, vitality; SR, social role functioning; ER, emotional role functioning; MH, mental health; PCS, Physical component summary score; MCS, Mental health component summary score; Q1,Q3, first and third quartile; IQR, interquartile range; SD, standard deviation.

**Supplemental Figure S1.** Flow chart of study participants.

One of the 60 participants had not completed the SF-36 questionnaire.

**Supplemental Figure S2.** Frequency distribution of the six components of MSTS. A higher score represents a better functional outcome.

**Supplemental Figure S3.** Distribution of the Musculoskeletal Tumor Society Score (MSTS) and components categorized by age groups. The scores were transformed in a scale from 0-100. ﻿A higher score represents a better functional outcome. P-values were performed with the Kruskal-Wallis test.

**Supplemental Figure S4.** Boxplots show the significant differences in the emotional acceptance scores by gender (A), by age at surgery (B) and by range of motion (C). The boxplots below exhibit the distribution of walking ability scores by age groups (D) and gait scores by ROM categories (E).

P-values were performed with the Wilcoxon-Mann-Whitney-U-Tess or Kruskal-Wallis test as appropriate.

Abbreviations:ROM,range of motion.

**Supplemental Figure S5.** Distribution of the Tegner activity level scale (Tegner score) by age groups(A) and by age at surgery groups (B). P-values were performed with Wilcoxon-Mann-Whityney-U-test or Kruskal-Wallis test.

**Supplemental Figure S6.** Distribution of the SF-36 and subscales categorized by age group

P-values were performed with the Kruskal-Wallis test.

Abbreviations: PF, physical functioning; PR, physical role functioning; BP, bodily pain; GH, general health; VT, vitality; SR, social role functioning; ER, emotional role functioning; MH, mental health; PCS, Physical component summary score; MCS, Mental health component summary score.

**Supplemental Table S1.** SF-36 scores for patients treated with rotationplasty compared to the German health general population25 based on the mean and median being equal in normally distributed data in the reference sample.

\*p < 0.05

\*\*p < 0.01

bold font indicates significant results.

Abbreviations: PF, physical functioning; PR, physical role functioning; BP, bodily pain; GH, general health; VT, vitality; SR, social role functioning; ER, emotional role functioning; MH, mental health; PCS, Physical component summary score; MCS, Mental health component summary score.