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ONCOLOGY

Rotationplasty outcomes assessed by gait analysis following resection of lower extremity bone neoplasms

A SYSTEMATIC REVIEW AND META-ANALYSIS

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Aims

The standard of surgical treatment for lower limb neoplasms had been characterized by highly interventional techniques, leading to severe kinetic impairment of the patients and incidences of phantom pain. Rotationplasty had arisen as a potent limb salvage treatment option for young cancer patients with lower limb bone tumours, but its impact on the gait through comparative studies still remains unclear several years after the introduction of the procedure. The aim of this study is to assess the effect of rotationplasty on gait parameters measured by gait analysis compared to healthy individuals.

Methods

The MEDLINE, Scopus, and Cochrane databases were systematically searched without time restriction until 10 January 2022 for eligible studies. Gait parameters measured by gait analysis were the outcomes of interest.

Results

Three studies were eligible for analyses. Compared to healthy individuals, rotationplasty significantly decreased gait velocity (-1.45 cm/sec; 95% confidence interval (CI) -1.98 to -0.93; p < 0.001), stride length (-1.20 cm; 95% CI -2.31 to -0.09; p < 0.001), cadence (-0.83 stride/min; 95% (CI -1.29 to -0.36; p < 0.001), and non-significantly increased cycle time (0.54 sec; 95% CI -0.42 to 1.51; p = 0.184).

Conclusion

Rotationplasty is a valid option for the management of lower limb bone tumours in young cancer patients. Larger studies, with high patient accrual, refined surgical techniques, and well planned rehabilitation strategies, are required to further improve the reported outcomes of this procedure.

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Keywords: rotationplasty, gait analysis, extremity bone neoplasms, orthopaedic oncology, oncology pediatric

Introduction

The introduction of limb sparing surgery procedures for the treatment of lower limb tumours has expanded the surgical arsenal for these clinical problems, as compared to older highly disabling techniques, such as amputation. This is the corollary of the constant evolution in chemotherapy, radiotherapy, and surgical techniques.[1] The current reconstruction options include: modular endoprosthetic reconstruction; bone graft reconstruction; bone transport; arthrodesis; and rotationplasty. With prosthetic and allograft reconstructions being in the spotlight of orthopaedic oncology,[2,3] rotationplasty is often neglected as a potent and viable treatment modality.

In rotationplasty, the ankle joint takes over the function of the knee joint by 180° rotation after wide-margin tumour resection

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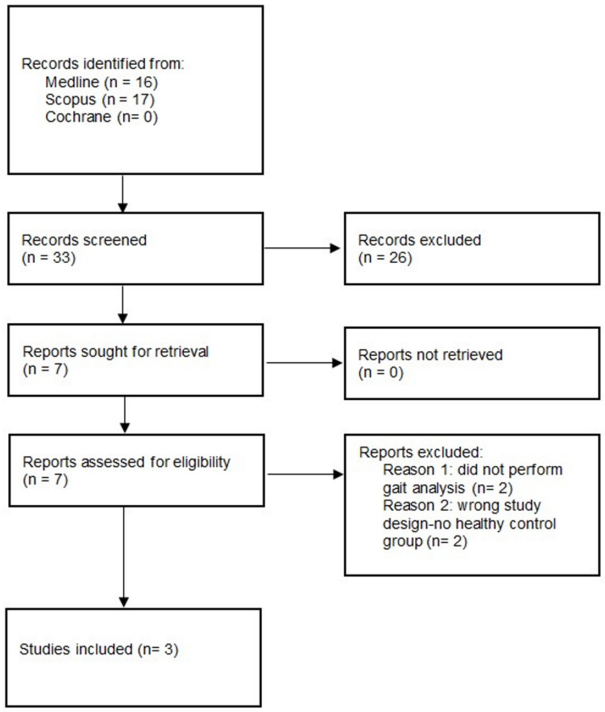

Fig. 1
Review flowchart.

that also includes the knee.[4] It can be considered as a treatment option when there is no tumour infiltration of the sciatic nerve and range of motion (ROM) of the ipsilateral ankle is preserved.[5] Despite being the reconstruction technique closest to amputation, rotationplasty is characterized by durable functional results and a plethora of advantages.[6-10] In a comparative study, patients with rotationplasty demonstrated equal functional performance when compared with prosthetic reconstruction patients, and favourable outcomes when compared with patients that underwent above-the-knee amputation.[11] However, a definite representation of the outcomes of rotationplasty versus the healthy population is missing from the literature.

Gait analysis has become a valuable tool in contemporary orthopaedics, and can serve a key role in the assessment of kinetic and kinematic outcomes of various procedures pertaining to musculoskeletal oncology. In this systematic review and meta-analysis, we aim to evaluate the impact of rotationplasty on gait parameters and compare them with the normal values found in the general population.

Methods

Search and selection. The MEDLINE, Scopus, and Cochrane library databases were systematically searched in 10 January 2022 without time restriction for studies of any duration and design that performed gait analysis in patients who underwent rotationplasty after lower limb tumour resection and compared their gait parameters with those of healthy individuals. The search algorithm contained: "gait", "gait analysis", "neoplasm*", "tumour*", "rotationplasty". This systematic review and meta-analysis was reported in accordance to the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) reporting guideline.

Two independent invsestigators (PF, NF) screened the articles by title and abstract. Reports that were potentially eligible were sought for retrieval and underwent full-text evaluation. If consensus on eligibility was not reached between the two investigators, a third investigator (DV) was involved to evaluate the article. Database searches were supplemented by forward and backward reference list screening. The eligibility was defined by the PICO framework: Population (P), patients undergoing gait analysis; Intervention (I), patients that underwent rotationplasty after lower limb tumour resection; Comparison (C), gait analysis of healthy individuals; and Outcomes (O), differences of the gait parameters between the two groups. Reasons for exclusion were: articles that did not refer to lower limb tumours as the cause of surgery; studies that did not perform gait analysis or did not perform a gait comparison with another group; studies that did not present the gait analysis results; and studies that were not in English. The outcomes of interest for our research were the spatiotemporal gait parameters: gait velocity; stride length; gait cycle duration; swing duration; stance duration; and cadence.

Data extraction. The data extraction was performed by two authors (PF, DD) independently. Any disagreement were resolved by consensus. Records of the same trial reporting at different follow-ups were considered a single trial. In case of double reporting data, data from the most informative publication and highest level of evidence were used. The standardized data exctraction form included: first author; year of publication; study design; number of participants (overall and by group); age at surgery; type of lower limb tumour; tumour location (hip, knee, ankle); type of surgery; gait analysis system used; time of gait analysis from surgery; gait velocity (metres/second (m/s) or centimetres/second (cm/s); stride length (cm or percentage of height), cycle duration (seconds (sec)), swing duration (sec), stance duration (sec), and cadence (steps/minute).

Statistical analysis. We calculated the pooled summary mean differences, and as well as the corresponding 95% confidence interval (CI), pooled the study specific estimates using random-effects models.[12] The pooled summary mean differences were calculated for the patients who went through rotationplasty compared to healthy individuals. The presence of heterogeneity was estimated with the Cochran's Q statistic and was quantified with I^2.[13] Possible small study effects (an indication of publication bias) by visual inspection of funnel plots and Egger's

Table I. Characteristics of the included studies.

Author, yr	Intervention, n	Healthy controls, n	Mean age at surgery, yrs (SD or range)	Mean evaluation after surgery, mnths (SD or range)	Tumour type	Location of tumour	Gait assessment
Benedetti et al, 2016[20]	16	20	11 (7)	200 (59)	Osteosarcoma, Ewing's sarcoma	Distal femur	An eight-camera stereophotogrammetric system (Vicon 612; Oxford Metrics, UK), and two forceplates (Kistler, Switzerland)
Fuchs et al, 2003[21]	7	25	9.4 (5 to 14)	99.6 (48 to 192)	Osteosarcoma	Distal femur and proximal tibia	Eight-camera expert vision real time system (Motion Analysis, USA). 21 retroflective markers. 3D marker data collected at 60 Hz. Bertec and Kistler force plates (Motion Analysis) for kinetic data collection.
Catani et al, 1993[22]	10	10	10.7 (7.5 to 14)	23.9 (10 to 50)	Osteosarcoma, Ewing's sarcoma	Distal Femur	Motion analyzer (Elite; BTS, Italy) for kinematic and temporal-distance measurements, and a force-plate (Kistler, Switzerland) for ground reaction forces.

SD, standard deviation.

test[14] would be used if at least ten studies were included in the meta-analysis, which is not applicable for this article. All analyses were performed using Stata version 14 (StataCorp, USA). Risk of bias was assessed using the Newcastle-Ottawa quality assessment scale.[15]

Results

Study selection and population characteristics. The systematic search of the electronic databases (MEDLINE, Scopus, and Cochrane) identified a total of 33 studies, seven of which were selected for full text screening. After exclusion of four studies due to non-availability of gait analysis parameters and no control group,[16-19] three studies were considered eligible for data extraction and meta-analysis according to our criteria of eligibility.[20-22] Figure 1 shows the flowchart of the study selection process.

Table I presents the characteristics of the included studies. A total of 88 patients were included in the analysis, of whom 33 (37.5%) underwent rotationplasty and 55 (62.5%) served as the healthy control group. Patients underwent surgery for lower limb sarcomas at a mean age of 10.37 years (4 to 18), and were followed-up for 108 months (10 to 259) after the rotationplasty procedure.

Risk of bias of the included studies is presented in Table II. The main factors that induced bias was the selection of the healthy cohorts that were not strictly matched to the patient cohorts, as well as the small follow-up of the Catani et al[22] study.

Study outcomes. The three included studies provided information for a variation of outcomes Adequate data for quantitative synthesis were derived for fait velocity, stride length, cycle time, swing time, stance time, and cadence. Regarding gait velocity, measured as cm/sec, three studies reported results. The quantitative synthesis of the three studies showed no statistically significant results (mean difference -1.45 cm/sec; 95% CI -1.98 to 0.93; I^2 = 7%) for the comparison among patients who went through rotationplasty and healthy individuals (Figure 2).

Table II. Risk of bias assessment.

Author, yr	Selection	Comparability	Outcome
Benedetti et al, 2016[20]	***	*	***
Fuchs et al, 2003[21]	***	*	***
Catani et al, 1993[22]	***	*	**

The three included studies also reported results on stride length, measured in cm. The quantitative synthesis showed that the patients who went through rotationplasty had a statistically significant reduction in stride length by a summary mean difference of -1.20 cm (95% CI -2.31 to –0.09; I^2 = 78.7%) compared to healthy individuals (Figure 3).

Regarding cycle time measured as sec, two studies provided data for quantitative synthesis. The quantitative synthesis of the two studies showed no statistically significant results (mean difference 0.54 sec; 95% CI -0.42 to 1.51; I^2 = 66.6%) for the comparison among patients who went through rotationplasty and healthy individuals (Figure 4).

Two studies reported results on swing time, measured as percentage of stride. A slightly statistically significant

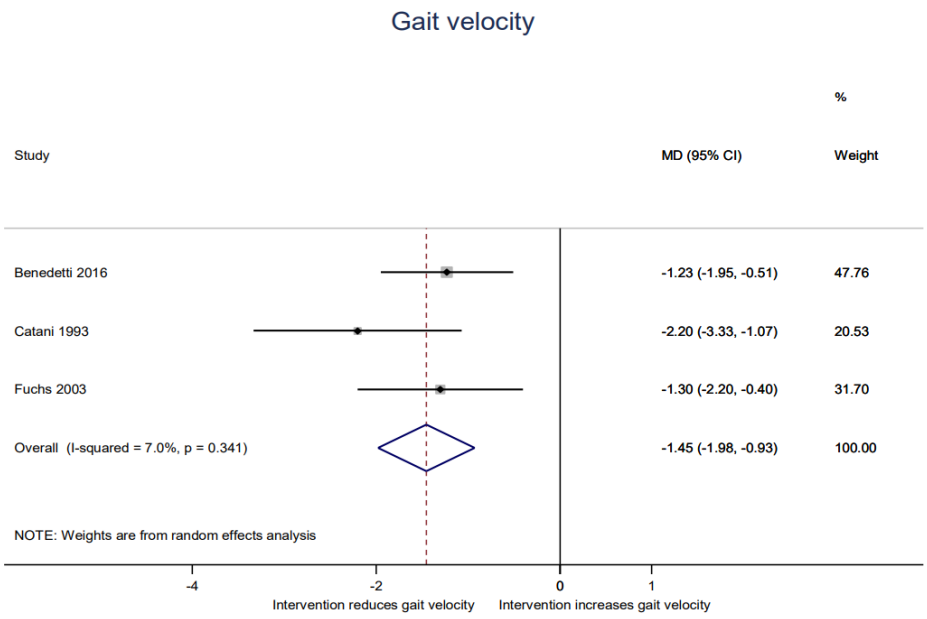

Fig. 2
Forest plot of gait velocity (metre/second).

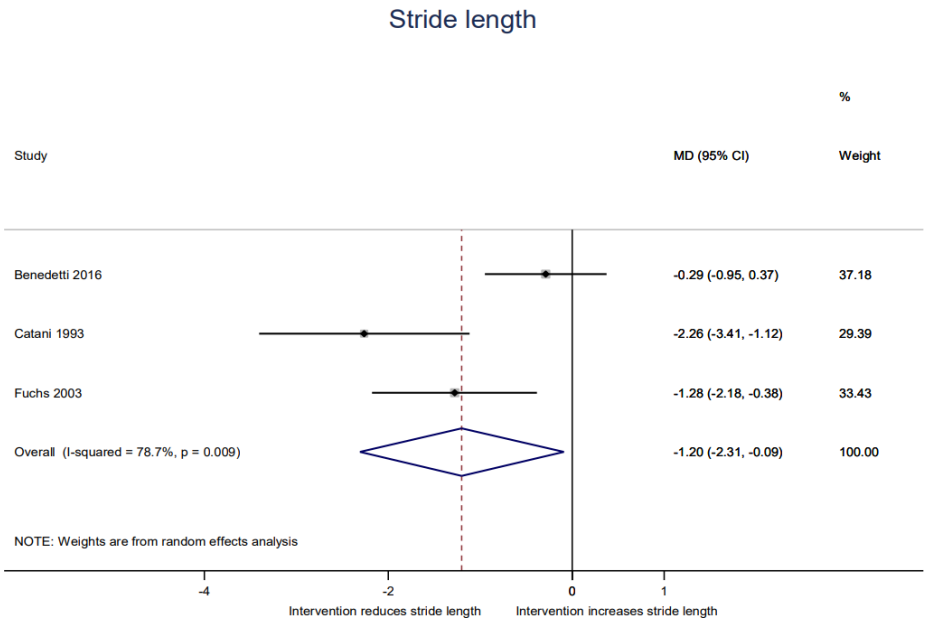

Fig. 3
Forest plot of stride length (% height).

increase in swing time for patients who went through rotationplasty compared to healthy individuals was observed (mean difference 0.57%; 95% CI 0.03% to 1.11%; $I^2 = 0\%$) (Figure 5).

Two studies reported results on stance time, measured as percentage of stride. A slightly statistically significant reduction in stance time for patients who went through rotationplasty compared to healthy individuals was observed (mean difference -0.57%; 95% CI -1.11% to 0.03%; $I^2 = 0\%$) (Figure 6).

Finally, the quantitative synthesis of the three studies showed a statistically significant reduction in cadence, measured as stride/min, by a summary mean difference of -0.83 stride/min (95% CI -1.29 to –0.36; $I^2 = 0\%$) compared to healthy individuals (Figure 7).

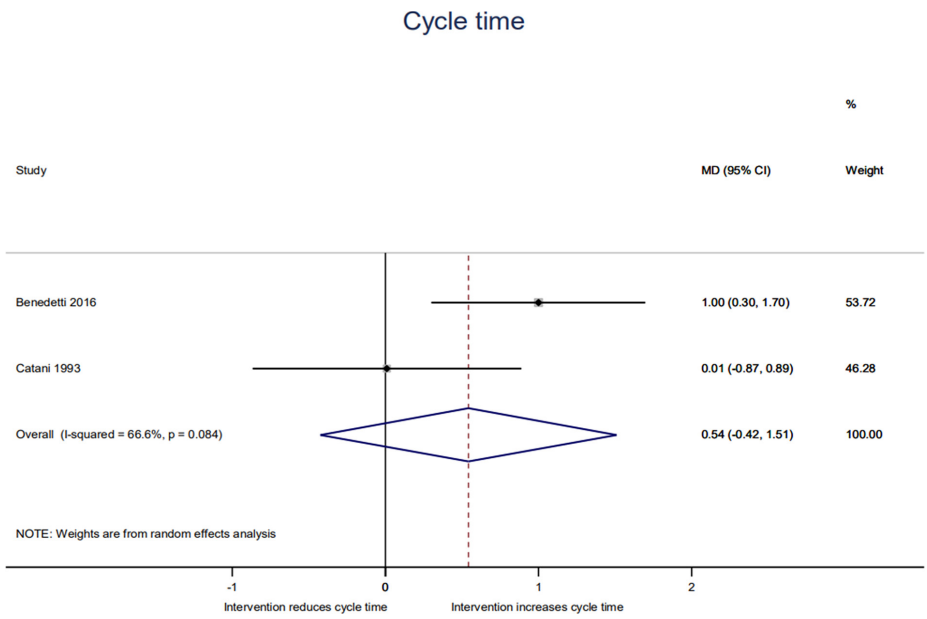

Fig. 4
Forest plot of cycle time (seconds).

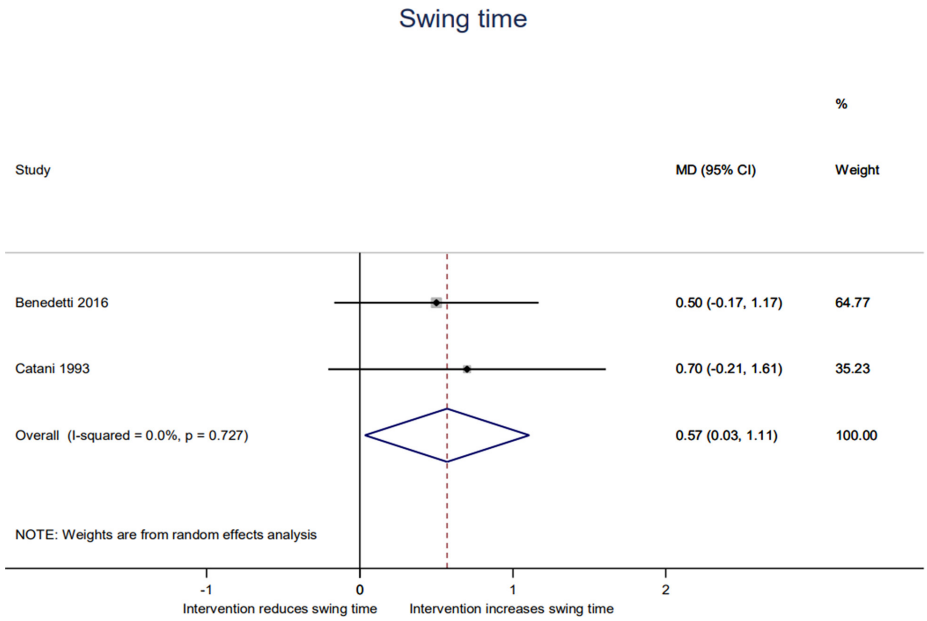

Fig. 5
Forest plot of swing time (% of cycle).

Discussion

Gait analysis as a reliable tool to assess the effectiveness of lower limb surgery for bone neoplasms have been proposed in several studies, especially in populations who underwent prosthetic reconstructions.[23,24] This is the first meta-analysis that attempts to depict the effectiveness of rotationplasty for lower limb tumours regarding gait parameters measured by gait analysis. The results of this article suggest that rotationplasty affects all the gait parameters, and summarizes the mean differences compared to the gait of healthy individuals. Taken into account the seemingly unfair comparison, this knowledge is not meant to downgrade the importance of the technique, but to enable more informative pre-surgical patient briefing, as well as to potentially guide future trials on lower limb neoplasms.

Three studies were included in the analysis.[20-22] Benedetti et al[20] reported the study with the highest patient

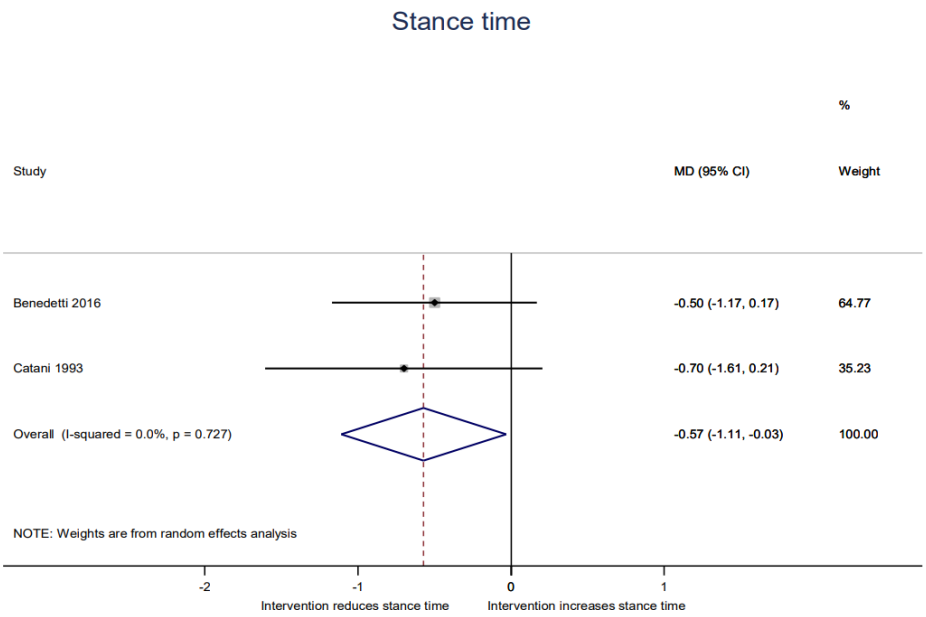

Fig. 6

Forest plost of stance time (% of cycle).

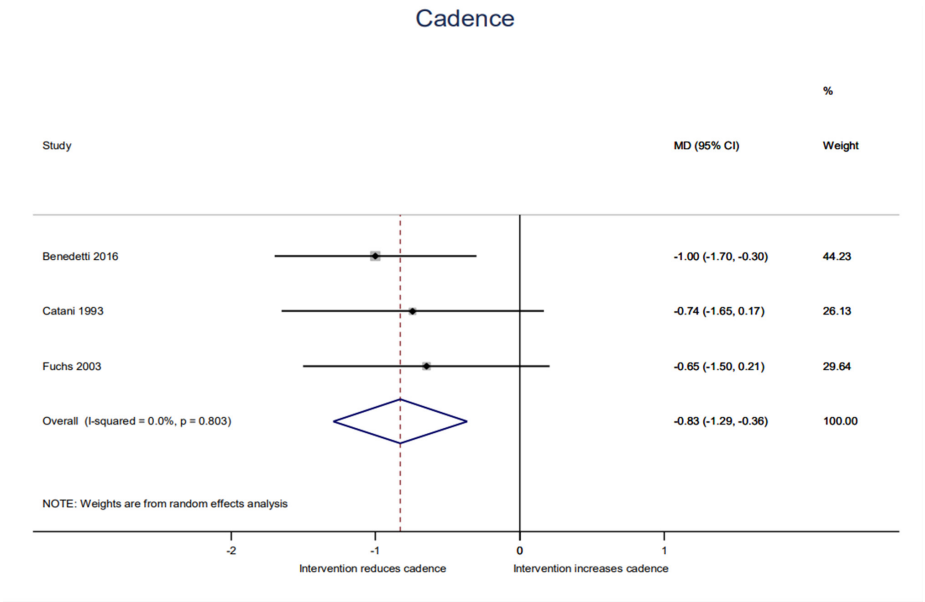

Fig. 7

Forest plot of cadence (stride/min).

recruitment period of 23 years. The authors highlighted the importance of small discrepancies in residual thigh-shank length between the treated and contralateral legs, in order to achieve the best walking performance. Furthermore, Fuchs et al[21] compared knee rotationplasty, not only to healthy controls, but also to distal above-knee amputees, and reported that the procedure offers beneficial outcomes that surpass amputation. Finally, Catani et al[22] concluded that rotationplasty is a valid procedure that enables the active control of the pseudo-knee, and allows a smooth and coordinate gait pattern, slightly inferior to normal. Interestingly, Catani et al[22] was the only study that clearly correlated rotationplasty outcomes with rehabilitation, underscoring that functionality is more successful when subjects are submitted to intensive rehabilitative training for equinism of the ankle and for strengthening of the calf muscle. As a point of reference for non-oncological studies, another study of long-term

follow-up and gait analysis also reported sufficient functional and quality of life outcomes for rotationplasty in patients with congenital proximal focal femoral deficiency compared to a control group.[25] The finding the there is a statistical reduction of stride length in roationplasty patients is justified by the decreased ROM in the sagittal plane of the ankle joint (substitute) relative to the knee joint (amputated). In this meta-analysis, it is interesting to highlight that while stride length is reduced, gait velocity appears comparable to normal controls.

Rotationplasty is a procedure that is reported to offer durable results with low complication rates and to evade the phenomenon of phantom pain after amputation.[22,26,27] The most notable complications are vascular and infectious complications, as well as superficial skin necrosis, delayed union at the knee level, and gradual de-rotation of the foot.[26,27] Lastly, the psychological repercussions are of paramount importance and should not be neglected by any means, requiring careful pre- and post-surgical management, as well as substantial long-term follow-up.[27]

The quality of life of cancer patients following rotationplasty has been a topic of great research interest.[16,17,28] It is suggested that the patients report high satisfaction rates regarding functionality, emotional and psychological state, daily activities, as well as sports. Especially for exercise, it has been shown that cancer patients who underwent rotationplasty can and should return to high-level physical activity and sports, which will also play an important role in their functional advancement.[29,30]

Our study also has limitations. Only three studies were eligible for analyses and data regarding all the gait parameters were not available in every study. However, we attempted to comprehensively present the data from the existing scarce sources. Moreover, there was no sufficient evidence to allow a meta-analysis of rotationplasty versus other techniques, such as amputation, which would be of great research interest. However, we elected to include only clinical trials (with control group), and therefore the results of our meta-analysis have higher importance and validity. While constructing future studies, it is important to note that the currently available articles did not offer information regarding prosthetic fitting and precise rehabilitation protocols, which are both critical determinants of the procedure. Finally, this article is directed specifically to cancer populations and future studies could potentially address the effectiveness of rotationplasty in other settings.

In conclusion, rotationplasty for lower limb bone neoplasms is a valid option that offers satisfactory functional outcomes in terms of gait parameters, and should be considered especially for very young cancer patients accompanied by psychological support. The evident negative impact of the procedure in the gait parameters compared to healthy controls is indicative of the essential improvements that are required to be made towards refinement of the rehabilitation strategies. Larger studies with evolved techniques and multiple comparator arms are awaited to offer novel insights on this research topic.

Take home message

- Rotationplasty is a valid and reliable procedure for the management of lower limb bone tumours in young cancer patients.
- Moving forward, larger studies, with high patient accrual and well-planned rehabilitation strategies, are required to further improve the reported outcomes.

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- G. Ntritsos: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Software, Validation, Visualization, Writing – review & editing.
- D. Dimopoulos: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Resources, Validation, Visualization, Writing – review & editing.
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