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Outcomes of Oncologic Arthroplasty in Children and Adolescents with Malignant Limb Tumors: A Systematic Review

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Abstract

Background. Amputation was historically the primary surgical intervention for children with limb bone sarcomas. However, the development and refinement of chemotherapy and radiotherapy protocols, along with advances in surgical techniques and implants, have significantly altered the treatment landscape for these patients. Currently, limb-sparing oncologic arthroplasty is the preferred surgical approach for treating limb sarcomas in children.

The aim of the systematic review is to analyse the outcomes of oncologic arthroplasty in children and adolescents.

Methods. A comprehensive literature search was performed in Google Scholar, PubMed, ScienceDirect, and eLIBRARY databases focusing on the keywords "endoprostheses, tumors, children", from 2000 to 2024. Data collection included patient demographics (number of patients, gender, age), follow-up period, disease diagnosis, tumor location, type of endoprosthesis, complications, functional outcomes based on the Musculoskeletal Tumor Society score (MSTS) in percentage, overall survival rates, and prosthesis survival rates.

Results. The review included the data from 30 articles on a total of 792 patients aged 2 to 18 years, with 422 males and 370 females. The average age was 11.4 years, and the average follow-up period was 6.5 years. Osteosarcoma was the most common diagnosis, accounting for 716 (88.8%) cases, followed by Ewing sarcoma with 67 (8.3%) cases. Distal femoral arthroplasties were performed most frequently (573 cases, 71.1%), followed by proximal tibial arthroplasty (148 cases, 18.3%). The most commonly used type of endoprosthesis was the non-invasively extendable type (540 cases, 67%). A total of 756 complications were recorded, resulting in a complication rate of 96%. The complications were predominantly oncologic (188 cases, 25%) and pediatric orthopedic (166 cases, 22%). The 5-year and 10-year overall survival rates were 81.68% and 77.63%, respectively, with an average prosthesis survival rate of 53.93%.

Conclusion. The data obtained indicate an extremely high frequency of complications during oncologic arthroplasty in children, mainly of an orthopedic profile, which requires analysis and development of measures to prevent them, as well as organizational solutions for the correction of these disorders.

Keywords: arthroplasty, oncologic arthroplasty, malignant tumors, children, periprosthetic fractures, periprosthetic infection.

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Исходы онкоэндопротезирования у детей и подростков при злокачественных новообразованиях конечностей: систематический обзор литературы

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Реферат

Актуальность. При саркомах костей конечностей у детей операцией выбора до 1980-х гг. являлась ампутация. Создание и совершенствование протоколов химиотерапии, лучевой терапии, развитие хирургической техники и имплантатов кардинально изменили возможности и результаты оказания помощи данной группе пациентов. В настоящее время органосохраняющая технология онкологического эндопротезирования является методом выбора в хирургическом лечении детей с саркомами конечностей.

Целью систематического обзора является анализ исходов онкологического эндопротезирования у детей и подростков.

Материал и методы. Поиск источников выполнялся в базах данных Google Scholar, PubMed, ScienceDirect, eLIBRARY по ключевым словам «endoprosthesis, tumors, children, эндопротезы, злокачественные опухоли, дети» с глубиной поиска с 2000 по 2024 г. Выполнялся анализ следующих данных: количество пациентов, пол, возраст, период наблюдения, диагноз, локализация опухоли, тип устанавливаемого эндопротеза, наличие осложнений, результаты оценки функции конечности по шкале Musculoskeletal Tumor Society (MSTS) в процентах, общая выживаемость и выживаемость эндопротеза.

Результаты. В анализируемый материал вошли данные из 30 статей, включающих всего 792 пациента в возрасте от 2 до 18 лет, из них 422 мальчика и 370 девочек. Средний возраст — 11,4 года, средний период наблюдения — 6,5 лет. На первом месте по встречаемости расположилась остеосаркома — 716 (88,8%) наблюдений, на втором — саркома Юинга — 67 (8,3%). Чаще всего выполнялось эндопротезирование дистального отдела бедренной кости — 573 (71,1%), а также проксимального отдела большеберцовой кости — 148 (18,3%). Наиболее часто используемый тип эндопротеза — неинвазивно удлиняемый — 540 (67%). Было зафиксировано 756 осложнений, частота встречаемости — 96%. В структуре осложнений преобладали онкологические и ортопедические осложнения на фоне роста ребенка — 188 (25%) и 166 (22%) соответственно. Общая выживаемость пациентов за 5 и 10 лет составила в среднем 81,68% и 77,63% соответственно при среднем значении выживаемости эндопротеза 53,93% за десятилетний период.

Заключение. Полученные данные свидетельствуют о крайне высокой частоте осложнений при онкопротезировании у детей, преимущественно ортопедических, что требует анализа и разработки мер по их предупреждению, а также организационных решений для коррекции данных нарушений.

Ключевые слова: эндопротезирование, онкопротезирование, злокачественные опухоли, дети, периопротезные переломы, периопротезная инфекция.

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INTRODUCTION

Primary malignant bone tumors account for 0.001% of all newly diagnosed malignant neoplasms. The incidence rate is 1 case per 100,000 population, with the majority occurring in children and adolescents [1]. In the past, the only treatment option was amputation of the affected limb. Even with such a radical intervention, the 5-year survival rate after amputation remained consistently low, especially in children, not exceeding 20% [2].

With the development of chemotherapy and improved survival rates for patients with musculoskeletal tumors, the issue of alternative treatments to amputation arose. Currently, there is no convincing evidence of differences in survival rates and local recurrence frequency between patients who undergo limb amputation and those who receive limb-sparing operations [3]. However, arthroplasty in this patient group is associated with a high rate of complications that often require additional surgical interventions and may hinder the achievement of satisfactory functional outcomes [3].

Specific complications, such as limb length discrepancy (LLD), occur when treating pediatric patients. For example, resection of the distal femur in children under 10 years old can result in limb growth retardation of up to 1.6 cm per year, potentially leading to a limb length discrepancy of 10-20 cm by the end of skeletal growth [3]. Small sample sizes and their heterogeneity in various publications make it difficult to fully analyse the outcomes of oncologic arthroplasty in children and adolescents, complicating the determination of treatment strategies for this patient group [4].

The aim of this review was to analyse the outcomes of oncologic arthroplasty in children and adolescents.

METHODS

A literature search was performed in the Google Scholar, PubMed, ScienceDirect, and eLIBRARY electronic databases using the following keywords: “endoprostheses, tumors, children”, from 2000 to 2024.

Inclusion criteria for publications in the systematic review:

1) articles in Russian or English;
2) full text available;
3) patients’ age under 18 years;
4) publication type: case-control studies, clinical series studies, clinical observations.

Exclusion criteria:

1) duplicates;
2) sources without mentions of tumors or endoprostheses;
3) when examining the text, it was found that oncologic arthroplasty had not been performed or had been performed only on adults;
4) no data on complications, or reports only on infectious complications;
5) mixed results of arthroplasty in adults and children.

The following data were analyzed: number of patients, gender, age, follow-up period, diagnosis, tumor location, type of prosthesis, presence of complications, limb function assessment results using the Musculoskeletal Tumor Society (MSTS) scoring system (in percentage), patient survival rates, duration of revision-free survival of prostheses, frequency, and structure of complications after arthroplasty.

For the description and classification of complications in oncologic arthroplasty, the Henderson classification was used in the analysed literature. According to this classification, five types of complications are defined. In 2014, the International Society of Limb Salvage (ISOLS) supplemented this classification with a sixth type — pediatric complications. This category includes complications related to growth plate blocking and deformity formation, as well as dysplastic joint changes associated with arthroplasty [5].

Statistical analysis

The collection, storage, and analysis of the obtained data were carried out using Microsoft Excel 2019. Absolute and relative values of various characteristics, as well as the median (Me) and interquartile range, were used to describe the data. The Mann-Whitney U test was employed to test null hypotheses. Differences were considered statistically significant at a two-sided significance level of $p < 0.05$.

RESULTS

From all identified publications, 30 articles were selected after applying exclusion criteria, from which the relevant data collection and analysis were carried out [1, 6-34].

The selection scheme of publications with quantitative data representation is shown in Figure 1.

The selected sources included data on 792 patients aged 2 to 18 years, with an average age of 11.4 years. Gender distribution was 422 boys and 370 girls. The average follow-up period for patients mentioned in the publications was 6.5 years.

The studies were conducted by oncologic and orthopedic centers as well as specialized university clinics. Quantitatively, the majority of publications originated from authors in China [10, 25, 29, 32, 33], the United Kingdom [21, 24, 26, 27, 30], the USA [9, 11, 22, 23], Egypt [7, 34], the Russian Federation [1, 8], Poland [15, 20], and Lebanon [12, 18, 28]. More detailed information on the geographic distribution, number, and volume of publications by patients is presented in Figure 2.

The structure of morphological diagnoses reflects known statistics for malignant tumors of the musculoskeletal system in children and adolescents. Osteosarcoma predominated with 716 (88.8%) cases, followed by Ewing sarcoma — 67 (8.3%) cases. Other musculoskeletal tumors were extremely rare: embryonal rhabdomyosarcoma — 1 case, chondrosarcoma — 3 cases, malignant giant cell tumor — 1 case, pleomorphic sarcoma — 5 cases. Additionally, there were 13 cases where patients had bone metastases with a primary diagnosis of osteosarcoma or Ewing sarcoma.

The location of tumors treated with arthroplasty is shown in Figure 3, with distal femoral and proximal tibial regions being predominant.

In limb-sparing surgery for malignant limb tumors in children, the most utilized prosthesis

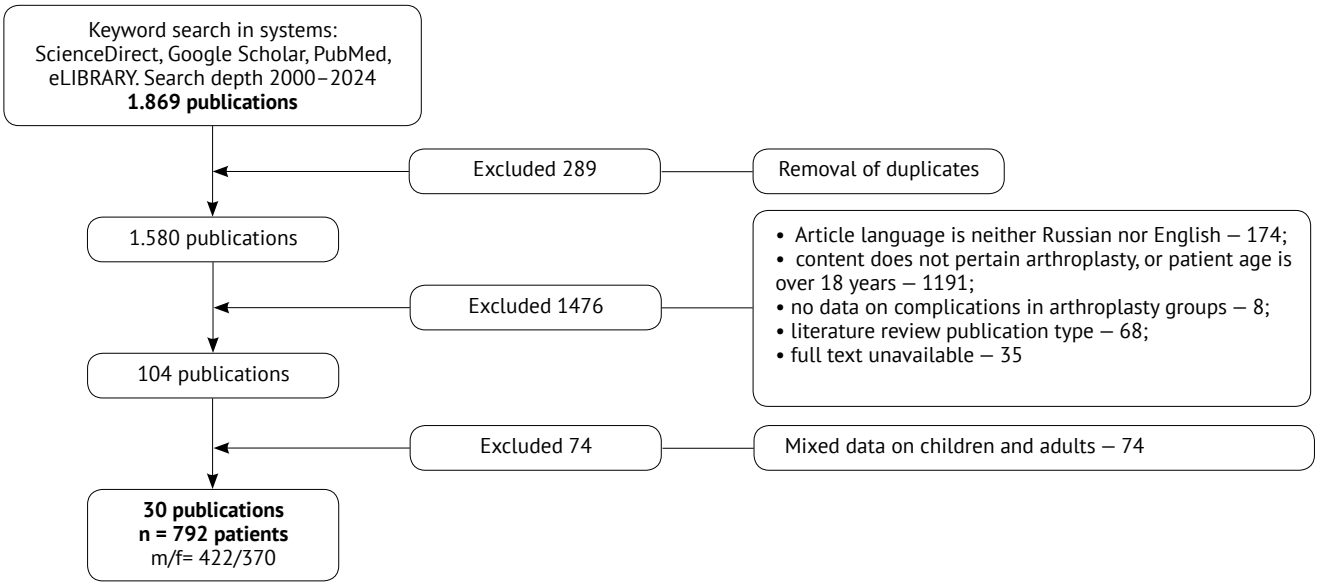

Figure 1. Flowchart of literature screening rocess

Figure 2. Geographic distribution of studies included in the systematic review (indicating the number of studies/number of patients)

type was a non-invasively extendable one with a magnetic extension mechanism. According to the publication analysis, it was used in 540 (67%) patients. Surgically (minimally invasive or via limited approach) extendable prostheses were significantly less common, used in 42 (5.2%) patients. Modular implants were applied in 211 (26.2%) cases. In one study, monolithic endoprostheses manufactured by 3D printing were mentioned — 13 (1.6%) cases.

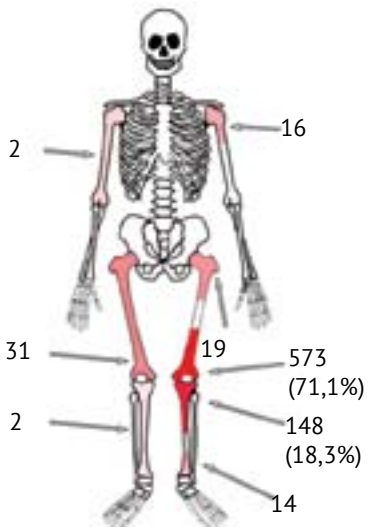

Figure 3. Tumor location

Treatment outcomes are intrinsically linked to complications associated with arthroplasty. The structure and statistics of complications according to the Henderson–ISOLS classification [5] are provided in Table 1. A total of 756 complications were recorded in 792 patients, accounting for 96%. Some patients experienced two or more complications. Oncologic and pediatric complications were the most prevalent. Data on aseptic prosthetic loosening, though less frequently reported, indicated that this complication had occurred in approximately 1 out of every 10 patients. The detailed structure of complications according to the Henderson–ISOLS classification is shown in Figure 4.

Table 1

Structure of complications according to the Henderson–ISOLS classification

Type of complication	Variants and number of complications
I	
Soft tissues Total 82 (11%): • functional — 34 (41%) • defect of closure — 48 (59%)	Neuropathy — 15 (18%) Joint stiffness — 25 (30%) Defects of ligaments and tendons — 8 (10%) Wound dehiscence — 12 (15%) Necrosis — 12 (15%) Delayed wound healing — 10 (12%)
II	
Prosthetic aseptic loosening	Prosthetic loosening without structural or infectious causes in the overall structure of complications — 74 (9,71%)
III	
Structural Total 134 (17%): • endoprosthesis-related (component failure or malfunction of extension mechanism) — 94 (70%) • bone-related (periprosthetic fracture) — 40 (30%)	Failure of the extension mechanism — 41 (31%) Prosthetic component failure — 53 (39%) Periprosthetic fracture — 40 (30%)
IV	
Infectious complications Total 112 (15%)	Superficial soft tissue infection — 14 (12%) Periprosthetic infection — 78 (70%) Infection requiring amputation — 20 (18%)
V	
Oncologic complications Total 188 (25%)	Local recurrence — 160 (85%) Metastatic recurrence — 28 (15%)
VI	
Pediatric complications Total 166 (22%)	LLD > 2 cm — 138 (83%) Limb deformity — 15 (9%) Implant-associated joint dysplasia (endoprosthesis dislocation, subluxation) — 13 (8%)

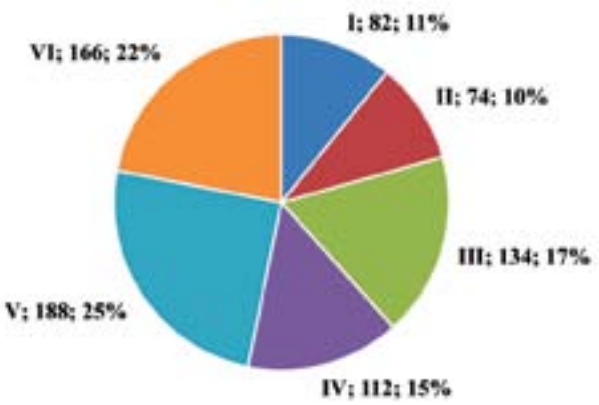

Figure 4. The structure of complications by their types according to the Henderson–ISOLS classification

Based on data analysis, endoprosthesis and patient survival rates were calculated. Prosthesis survival rate was defined by cases necessitating revision and replacement of implant components associated with failure, deep infections, and other causes. Assessment was performed over a 5-year period from the time of prosthesis implantation. During this period, 334 (42%) revisions were required, resulting in a 5-year mean prosthesis survival rate of 58%. Given the high frequency of type V complications, patient survival rate at the five- and ten-year marks was 80.0% and 78.5%, respectively.

The quality of life was assessed in all publications using the Musculoskeletal Tumor Society (MSTS) scale. Some authors used a system with a maximum score of 30 points, while others presented results as a percentage. To facilitate data consolidation, all results were converted to percentages, where the minimum result was 0% and the maximum was 100%. Subsequently, the data were grouped based on patient follow-up duration: less than 6 years and more than 6 years, with the average quality-of-life scores being 85.23% and 80.77%, respectively (Figure 5).

In terms of topography, neoplasms predominantly affected the distal femur (DF) and the proximal tibia (PT). Accordingly, the tumors of the majority of patients experiencing complications following arthroplasty had these localizations. For DF, the total number of complications was 340 (93.15%), while for PT, it was 37 (72.54%). The quality-of-life score according to the MSTS scale was statistically significantly higher (p = 0.0256) in the DF group, with an average score of 86.45% compared to 73.96% in the PT sarcoma group (Figure 6).

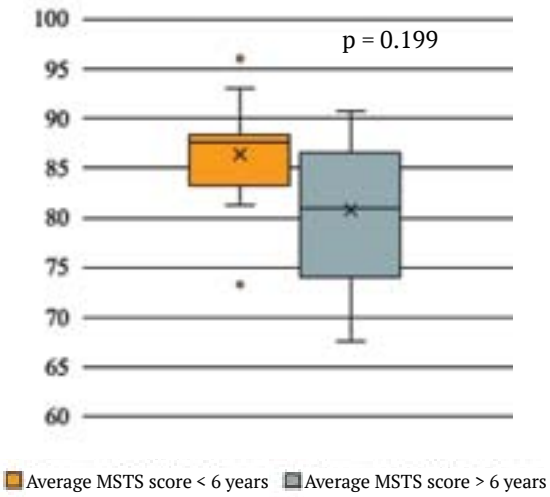

Figure 5. Quality of life measured by the MSTS scale with follow-up periods of less than 6 years and more than 6 years

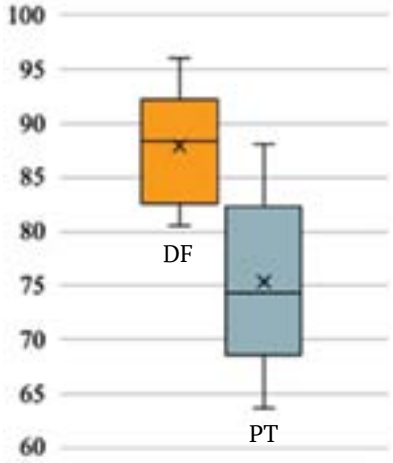

Figure 6. Quality of life after operations on different locations

The analysis of complication rate by endoprosthesis type showed 538 complications in 530 patients managed with non-invasively extendable implants, amounting to 101.51%, indicating that some patients had two or more types of complications. In the group with invasive and modular implants, 161 complications were noted in 197 patients, representing 81.72%. No significant differences in the quality of life and 5-year implant survival rates were observed based on the type of endoprosthesis (Table 2).

Table 2

Comparison of different types of endoprostheses

Comparison parameter	Non-invasively extendable endoprosthesis	Others	p-value between comparison parameters
Median MSTS	85%	74.29%	0.1421
5-year endoprosthesis survival rate	54.3%	60.3%	0.3884

DISCUSSION

The number of publications dedicated to the treatment of osteosarcomata in children has shown a progressive increase over the past decade, highlighting both the relevance of the topic and the accumulation and critical assessment of data on this disease and modern treatment options. Currently, limb-sparing surgery can be applied in 80-85% of cases in children with osteosarcomata [35]. This emphasizes the importance of analysing contemporary data on treatment outcomes. Unfortunately, out of 1.869 publications, only 30 high-quality studies suitable for analysis were found.

Until the 1970s, the predominant method of surgical treatment for osteosarcomata achieved 5-year survival rates in no more than 25% of cases [36]. Advances in modern chemotherapy protocols and surgical technologies have led to a 5-year survival rate of 70% for children with this pathology [35, 37]. According to our analysis, the 5-year survival rate for children reaches 80%. These results allow us to focus not only on life preservation but also on the quality of life comparable to that of children in the general population. The quality of life depends on the treatment outcomes and the frequency and nature of potential complications from aggressive surgery and chemotherapy aimed at achieving sustained remission.

According to our analysis, the primary type of endoprosthesis used in treating sarcomata in children is non-invasively extendable implants, which allow for the lengthening of the operated limb in line with the child's growth and development. Despite a high incidence of structural complications, the quality-of-life scores for this group were higher, confirming the importance of minimally invasive approaches in pediatric surgery and orthopedics. Unfortunately, non-invasively extendable devices are currently not manufactured in the Russian Federation.

The performed analysis revealed a very high complication rate, consistent with the data from certain clinical series presented in various studies [7, 18]. Approximately 75% of all complications were non-oncologic in nature, primarily related to infections and, even more frequently, various orthopedic complications.

According to the data, tumors most often developed during growth spurts (average patient age — 11.4 years), with primary tumor sites typically located in growth zones characterized by high activity and contributing significantly to overall growth (distal femur and proximal tibia). Type VI complications were the second most frequent, raising the need for active involvement of pediatric orthopedic specialists in post-oncologic arthroplasty care. A.V. Petrichenko et al. also emphasize this problem [38].

The high complication rate highlights the quality-of-life scores measured by the MSTS scale, averaging 86.3% for the first 6 years of observation and 80.7% in subsequent years, despite nearly half of the endoprostheses failing within 5 years post-implantation. Possible reasons for this discrepancy include the subjectivity of the scale and the tendency of children to overlook existing problems due to high adaptability. Our study did not find any publications addressing this issue. However, M.G. Vitale et al. have raised questions about the insufficient effectiveness of quality-of-life scales for pediatric orthopedic patients compared to adult patients, particularly the SF-36 scale [39]. Similar traits may apply to quality-of-life assessments in this group. Methodological limitations tied to patient

categories should not be excluded. For instance, patients undergoing arthroplasty for the tumor of the distal femur retain a higher quality of life despite frequent oncologic complications. Given that metastatic recurrence can result in a patient's death, further quality-of-life assessments become impossible, leaving outcomes fixed at a high level. Conversely, aseptic loosening is a prolonged process, often spanning years (usually 2-3 years) [40, 41], masking clinical symptoms and causing minimal concern for the child.

A key aspect of the study is the near-total lack of research on patterns of orthopedic complications, their prevention, and treatment in pediatric patients. Despite the distraction capabilities of modern devices, the lengthening mechanism remains technically complex. The analysis revealed frequent malfunctions, preventing compensation for length discrepancies in limbs. The engineering solutions enabling lengthening require sufficient space, ultimately influencing the extent of bone resection. One potential approach to mitigating this limitation involves seeking for more advanced technical solutions and, where oncologically feasible, opting for surgical procedures that preserve the growth zone and joint. Studies focused on limited resections and biological reconstruction surgical techniques for pediatric musculoskeletal malignant tumors remain highly relevant [42, 43, 44, 45].

CONCLUSIONS

The analysis of publications revealed a high level of interest in pediatric arthroplasty for malignant musculoskeletal tumors. However, the number of comprehensive studies that allow for an in-depth analysis of the accumulated global medical practice is relatively limited. The conducted analysis showed an extremely high frequency of complications in oncologic arthroplasty in growing children, primarily of a non-oncologic nature. This highlights the need to improve oncologic arthroplasty technologies, seek and develop alternative solutions that preserve the natural growth potential, and underscores the importance of involving pediatric trauma and orthopedic specialists in the treatment and follow-up care of this patient category.

DISCLAIMERS

Author contribution

Zorin V.I. — study concept and design, data analysis and interpretation, drafting and editing the manuscript.

Vissarionov S.V. — study concept and design.

Makarov A.Yu. — data acquisition, analysis and interpretation, drafting the manuscript.

Rybinskikh T.S. — data acquisition, analysis and interpretation.

Rodionova K.N. — data analysis and interpretation.

All authors have read and approved the final version of the manuscript of the article. All authors agree to bear responsibility for all aspects of the study to ensure proper consideration and resolution of all possible issues related to the correctness and reliability of any part of the work.

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Consent for publication. Not required.

REFERENCES

1. Bolshakov N.A., Artemov A.Yu., Vorochai A.M., Khamin I.G., Tikhonova M.V., Slinin A.S. et al. Endoprosthesis reconstruction, as a surgical treatment of children and adolescence with bone tumors around the knee. *Pediatric Hematology/Oncology and Immunopathology.* 2019;18(1):55-61. (In Russian). doi: 10.24287/1726-1708-2019-18-1-55-61.
2. Hagleitner M.M., de Bont E.S., Te Loo D.M. Survival trends and long-term toxicity in pediatric patients with osteosarcoma. *Sarcoma*. 2012;2012:636405. doi: 10.1155/2012/636405.
3. Dotan A., Dadia S., Bickels J., Nirkin A., Flusser G., Issakov J. et al. Expandable endoprosthesis for limb-sparing surgery in children: long-term results. *J Child Orthop.* 2010;4(5):391-400. doi: 10.1007/s11832-010-0270-x.
4. Groundland J.S., Ambler S.B., Houskamp L.D.J., Orriola J.J., Binitie O.T., Letson G.D. Surgical and Functional Outcomes After Limb-Preservation Surgery for Tumor in Pediatric Patients: A Systematic Review. *JBJS Rev.* 2016;4(2):e2. doi: 10.2106/JBJS.RVW.O.00013.
5. Henderson E.R., O'Connor M.I., Ruggieri P., Windhager R., Funovics P.T., Gibbons C.L. et al. Classification of failure of limb salvage after reconstructive surgery for bone tumours : a modified system Including biological and expandable reconstructions. *Bone Joint J.* 2014;96-B(11):1436-1440. doi: 10.1302/0301-620X.96B11.34747.

6. Penna V., Toller E.A., Pinheiro C., Becker R.G. A new approach to partialknee endoprosthesis in primary bone sarcomas. *Rev Bras Ortop.* 2015;44(1):46-51. doi: 10.1016/S2255-4971(15)30048-3.
7. Badr I., Eid T.A., Salem D., Shaheen Ahmed el Dadawy, Habeeb Mohammed el Sawy, Donati Davide Marie et al. Functional outcome and Limb length discrepancy after endoprosthetic reconstruction in skeletally immature patients with bone tumor. *Egyptian Orthop J.* 2019;54(Suppl 1):25-30.
8. Savlaev K.F., Petrichenko A.V., Ivanova N.M., Sharoev T.A. Complications of endoproshetics for bone tumors in children. *Quantum Satis.* 2021;4(1-4):19-22. (In Russian).
9. Henderson E.R., Pepper A.M., Letson G.D. What are estimated reimbursements for lower extremity prostheses capable of surgical and nonsurgical lengthening? *Clin Orthop Relat Res.* 2012;470(4):1194-1203. doi: 10.1007/s11999-011-2186-2.
10. Zan P., Shen J., Liu K., Wang H., Cai Z., Ma X. et al. Custom-made semi-joint prosthesis replacement combined ligament advanced reinforcement system (LARS) ligament reconstruction for the limb salvage surgery of malignant tumors in the distal femur in skeletal immature children. *Front Pediatr.* 2023;11:1168637. doi: 10.3389/fped.2023.1168637.
11. Benevenia J., Patterson F., Beebe K., Tucker K., Moore J., Ippolito J. et al. Results of 20 consecutive patients treated with the Repiphysis expandable prosthesis for primary malignant bone. *Springerplus*. 2015;4:793. doi: 10.1186/s40064-015-1582-6.
12. Haidar R., Sagghieh S., Muwakitt S., Mumtaz G., Akel S., Kamzoul R. et al. Limb salvage surgery for children and adolescents with malignant bone tumors in a developing country. *Pediatr Blood Cance*r. 2008; 51(6):787-791. doi: 10.1002/pbc.21696.
13. Futani H., Minamizaki T., Nishimoto Y., Abe S., Yabe H., Ueda T. Long-term follow-up after limb salvage in skeletally immature children with a primary malignant tumor of the distal end of the femur. *J Bone Joint Surg Am*. 2006;88(3):595-603. doi: 10.2106/JBJS.C.01686.
14. Gundavda M.K., Agarwal M.G. Growing Without Pain: The Noninvasive Expandable Prosthesis is Boon for Children with Bone Cancer, as well as Their Surgeons! *Indian J Orthop.* 2019;53(1):174-182. doi: 10.4103/ortho.IJOrtho_53_17.
15. Raciborska A., Malesza I., Bilska K., Koziński T., Pachuta B. Double Endoprosthesis in the Management of Refractory Metastatic Primary Bone Tumors in Children and Young Adults. *Adv Orthop.* 2021;2021:9944702. doi: 10.1155/2021/9944702.
16. Shehadeh A., Al-Qawasmi M., Al Btoush O., Obeid Z. Tibia Multiplanar Deformities and Growth Disturbance Following Expandable Endoprosthetic Distal Femur Replacement. *J Clin Med.* 2022;11(22):6734. doi: 10.3390/jcm11226734.
17. Staals E.L., Colangeli M., Ali N., Casanova J.M., Donati D.M., Manfrini M. Are Complications Associated With the Repiphysis(®) Expandable Distal Femoral Prosthesis Acceptable for Its Continued Use? *Clin Orthop Relat Res.* 2015;473(9):3003-3013. doi: 10.1007/s11999-015-4355-1.
18. Masrouha K., Abboud M., Saab R., Muwakkit S.A., Khoury N., Haidar R. et al. Long-term follow-up of children treated with the Repiphysis expandable prosthesis for lower extremity bone sarcoma. *J Pediatr Orthop B.* 2022;31(2):e258-e263. doi: 10.1097/BPB.0000000000000891.
19. Torner F., Segur J.M., Ullot R., Soldado F., Domenech P., DeSena L. et al. Non-invasive expandable prosthesis in musculoskeletal oncology paediatric patients for the distal and proximal femur. First results. *Int Orthop.* 2016;40(8):1683-1688. doi: 10.1007/s00264-016-3163-x.
20. Raciborska A., Bilska K., Malesza I., Rodriguez-Galindo C., Pachuta B. Distal Tibial Reconstruction in the Management of Primary Bone Tumors in Children and Adolescents. *Foot Ankle Int*. 2021;42(11): 1447-1453. doi: 10.1177/10711007211012690.
21. Sewell M.D., Spiegelberg B.G., Hanna S.A., Aston W.J., Bartlett W., Blunn G.W. et al. Total femoral endoprosthetic replacement following excision of bone tumours. *J Bone Joint Surg Br.* 2009;91(11):1513-1520. doi: 10.1302/0301-620X.91B11.21996.
22. Ahlmann E.R., Menendez L.R., Kermani C., Gotha H. Survivorship and clinical outcome of modular endoprosthetic reconstruction for neoplastic disease of the lower limb. *J Bone Joint Surg Br.* 2006;88(6): 790-795. doi: 10.1302/0301-620X.88B6.17519.
23. Cipriano C.A., Gruzinova I.S., Frank R.M., Gitelis S., Virkus W.W. Frequent complications and severe bone loss associated with the repiphysis expandable distal femoral prosthesis. *Clin Orthop Relat Res*. 2015;473(3): 831-838. doi: 10.1007/s11999-014-3564-3.
24. Sambri A., Staals E., Medellin M.R., Abudu A., Gikas P., Pollock R. et al. Stanmore noninvasive extendible endoprosthesis in the treatment of bone sarcoma in the preadolescent. *J Surg Oncol.* 2019;120(2):176-182. doi: 10.1002/jso.25501.
25. Gong T., Lu M., Min L., Luo Y., Tu C. Reconstruction of a 3D-printed endoprosthesis after joint-preserving surgery with intraoperative physeal distraction for childhood malignancies of the distal femur. *J Orthop Surg Res*. 2023;18(1):534. doi: 10.1186/s13018-023-04037-4.
26. Gilg M.M., Gaston C.L., Jeys L., Abudu A., Tillman R.M., Stevenson J.D. et al. The use of a non-invasive extendable prosthesis at the time of revision arthroplasty. *Bone Joint J.* 2018;100-B(3):370-377. doi: 10.1302/0301-620X.100B3.BJJ-2017-0651.R1.
27. Tsuda Y., Fujiwara T., Sree D., Stevenson J.D., Evans S., Abudu A. Physeal-preserving endoprosthetic replacement with short segment fixation in children with bone sarcomas. *Bone Joint J.* 2019;101-B(9):1144-1150. doi: 10.1302/0301-620X.101B9.BJJ-2018-1333.R1.

28. Saghieh S., Abboud M.R., Muwakkit S.A., Saab R., Rao B., Haidar R. Seven-year experience of using Repiphysis expandable prosthesis in children with bone tumors. *Pediatr Blood Cancer*. 2010;55(3):457-463. doi: 10.1002/pbc.22598.
29. Zou C., Zhao Z., Lin T., Huang Y., Xie X., Yin J. et al. Long-term outcomes of limb salvage treatment with custom-made extendible endoprosthesis for bone sarcoma around the knee in children. *J Orthop Surg Res*. 2020;15(1):14. doi: 10.1186/s13018-019-1534-x.
30. Gilg M.M., Gaston C.L., Parry M.C., Jeys L., Abudu A., Tillman R.M. et al. What is the morbidity of a non-invasive growing prosthesis? *Bone Joint J*. 2016;98-B(12): 1697-1703. doi: 10.1302/0301-620X.98B12.BJJ-2016-0467.
31. Ajit Singh V., Earnest Kunasingh D., Haseeb A., Yasin N.F. Outcome of expandable endoprosthesis: A single centre retrospective review. *J Orthop Surg (Hong Kong)*. 2019;27(2):2309499019850313. doi: 10.1177/2309499019850313.
32. Huang J., Cheng J., Bi W., Xu M., Jia J., Han G. et al. Neoadjuvant Chemotherapy and Expandable Prosthesis Reconstruction to Treat Osteosarcoma around the Knee in Children. *Orthop Surg*. 2023;15(1):162-168. doi: 10.1111/os.13563.
33. Ji T., Yang Y., Li D.S., Tang X.D., Guo W. Limb Salvage Using Non-hinged Endoprosthesis and Staged Correction of Leg-length Discrepancy for Children with Distal Femoral Malignant Tumors. *Orthop Surg*. 2019;11(5):819-825. doi: 10.1111/os.12525.
34. El Ghoneimy A.M., Shehab A.M., Farid N. What is the Cumulative Incidence of Revision Surgery and What Are the Complications Associated With Stemmed Cementless Nonextendable Endoprostheses in Patients 18 Years or Younger With Primary Bone Sarcomas About the Knee. *Clin Orthop Relat Res*. 2022;480(7):1329-1338. doi: 10.1097/CORR.0000000000002150.
35. Eaton B.R., Schwarz R., Vatner R., Yeh B., Claude L., Indelicato D.J. et al. Osteosarcoma. *Pediatr Blood Cancer*. 2021;68 Suppl 2:e28352. doi: 10.1002/pbc.28352.
36. Jafari F., Javdansirat S., Sanaie S., Naseri A., Shamekh A., Rostamzadeh D. et al. Osteosarcoma: A comprehensive review of management and treatment strategies. *Ann Diagn Pathol*. 2020;49:151654. doi: 10.1016/j.anndiagpath.2020.151654.
37. Panez-Toro I., Muñoz-García J., Vargas-Franco J.W., Renodon-Cornière A., Heymann M.F., Lézot F. et al. Advances in Osteosarcoma. *Curr Osteoporos Rep*. 2023;21(4):330-343. doi: 10.1007/s11914-023-00803-9.
38. Petrichenko A.V., Bukreeva E.A., Romanov P.A., Tiganova O.A., Ivanova N.M., Ochkurenko A.A. et al. Musculoskeletal sequelae of childhood bone sarcoma survivors. *Pediatric Traumatology, Orthopaedics and Reconstructive Surgery*. 2019;7(1):57-70. (In Russian). doi: 10.17816/PTORS7157-70.
39. Vitale M.G., Levy D.E., Johnson M.G., Gelijns A.C., Moskowitz A.J., Roye B.P. et al. Assessment of quality of life in adolescent patients with orthopaedic problems: are adult measures appropriate? *J Pediatr Orthop*. 2001;21(5):622-628.
40. Bergin P.F., Noveau J.B., Jelinek J.S., Henshaw R.M. Aseptic loosening rates in distal femoral endoprostheses: does stem size matter? *Clin Orthop Relat Res*. 2012;470(3):743-750. doi: 10.1007/s11999-011-2081-x.
41. Zhang H.R., Zhang J.Y., Yang X.G., Qiao R.Q., Li J.K., Ren Z.P. et al. The effects of length of femoral stem on aseptic loosening following cemented distal femoral endoprosthetic replacement in tumour surgery. *Int Orthop*. 2020;44(7):1427-1433. doi: 10.1007/s00264-020-04629-3.
42. Aponte-Tinao L., Ayerza M.A., Muscolo D.L., Farfalli G.L. Survival, recurrence, and function after epiphyseal preservation and allograft reconstruction in osteosarcoma of the knee. *Clin Orthop Relat Res*. 2015;473(5):1789-1796. doi: 10.1007/s11999-014-4028-5.
43. Scanferla R., Scolari F., Muratori F., Tamburini A., Delcroix L., Scoccianti G. et al. Joint-Sparing Resection around the Knee for Osteosarcoma: Long-Term Outcomes of Biologic Reconstruction with Vascularized Fibula Graft Combined with Massive Allograft. *Cancers (Basel)*. 2024;16(9):1672. doi: 10.3390/cancers16091672.
44. Takeuchi A., Yamamoto N., Hayashi K., Matsubara H., Miwa S., Igarashi K. et al. Joint-preservation surgery for pediatric osteosarcoma of the knee joint. *Cancer Metastasis Rev*. 2019;38(4):709-722. doi: 10.1007/s10555-019-09835-z.
45. Liu W., He H.B., Zhang C., Liu Y.P., Wan J. Distraction-suppression effect on osteosarcoma. *Med Hypotheses*. 2018;121:4-5. doi: 10.1016/j.mehy.2018.09.004.

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