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Subtotal proximal resection and tibial reconstruction with a modular endoprosthesis complemented with custom-made short distal stem (a case report)

V.Yu. Karpenko, A.L. Karasev✉, A.F. Kolondaev, K.A. Antonov, N.A. Lyubeznov

National Medical Research Center of Traumatology and Orthopedics n.a. N.N. Priorov, Moscow, Russian Federation

Corresponding author: Anatoly L. Karasev, karaseva81@mail.ru

Abstract

Introduction Malignant tumors are frequently localized in the long bones. Radical resection and reconstruction with megaprostheses is the gold standard of surgery in this group of patients. Unfortunately, the use of standard modular components is not possible in subtotal resection or is associated with a high risk of instability. Development of personalized shortened components of endoprostheses based on 3D computer modeling expands the possibilities of limb salvage surgical treatment. **Materials and methods** We describe a case of surgical treatment of a patient with extensive tibial fibrosarcoma. Pre-operative diagnosis based on CT, MRI, PET-CT and biopsy was low-grade fibrosarcoma, post-operative diagnosis was the same. Radical subtotal proximal resection of the tibia was performed, and modular knee megaprosthesis based on 3D-modelling custom-made distal short tibial component of hybrid fixation was used for reconstruction. Rehabilitation after surgery included wearing knee and ankle orthoses. **Results** No tumor recurrence or metastases were revealed one year after surgery, functional and radiological results were excellent. Patient walked without support, her gate was correct, and MSTS score was 83 %. **Discussion** In recent years, custom-made short components of oncological endoprostheses using 3D computer modeling have been developed. The short custom-made tibial component used by us in the report is a combination of a short cemented stem locked with two extraosseous plates with a rough surface. It simultaneously ensures the strength of the implant and increases the contact with the distal tibia. Excellent radiological and functional results obtained one year after the operation allow us to hope for a positive outcome in the medium term and to delay extirpation of the tibia. **Conclusion** Radical bone resections and megaprosthetic reconstruction in malignant tumors provide the best functional results. Implementation of based on 3D-modelling custom-made prosthetic components in extensive resections is a perspective trend in limb-salvage surgery.

Keywords: bone tumor, tibial resection, arthroplasty, megaprosthesis, custom-made prosthesis

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INTRODUCTION

The most common primary malignant skeletal tumors are osteosarcoma, chondrosarcoma, and Ewing's sarcoma (85–95 % of all cases); less common are fibrosarcoma, undifferentiated high-grade pleomorphic sarcoma, plasmacytoma, angiosarcoma, and some others [1].

In the second and third decades of life, bones are more often affected by osteogenic sarcoma. The incidence of chondrosarcomas and fibrosarcomas increases with age, and metastatic lesions become prevalent. The incidence in the population, according to the analysis of the SEER database, is 18.8 cases per 100,000, while the number of new cases in the United States alone can reach one hundred thousand per year [2].

Long bones are one of the most common sites of malignant skeletal tumors. Their lesion by tumors leads to limb dysfunction, and in the case of a pathological fracture, to severe pain, patient's immobility, and the development of severe life-threatening somatic complications.

Timely surgical intervention aimed at removing the tumor saves or prolongs patients' life and improves their quality of life.

In the past, the main surgical method for treating patients with malignant tumors of long bones was mutilation operations, amputations and disarticulations. However, with the development of medical technologies, it became possible to carry out limb-salvage treatment.

To manage extensive defects of long bones after resections, osteoplastic methods can be used with application of massive allogeneic grafts, autografts, transosseous extrafocal osteosynthesis, the Masquelet technique [3, 4, 5, 6, 7].

Nevertheless, the use of oncological endoprostheses in patients with malignant tumors in most skeletal locations provides the most rapid and complete recovery of limb function, and therefore this approach is currently considered the leading method of surgical treatment.

A significant increase in the life expectancy of patients with bone tumors in the recent decades due to the improvement of combined treatment regimens and surgical techniques imposes new requirements for the "survival" of implants and their functionality [8].

Extensive subtotal damage to the long bone poses significant difficulties for endoprosthesis fixation, often making it impossible to save adjacent joints. This can exclude limb-salvage surgery or drastically worsen the functional result, increase the risk of complications in the postoperative period and limit the "survival" of the endoprosthesis.

The use of standard stems in such cases is impossible, and therefore, various authors have proposed short components based on intra- and extramedullary fixation. They provide both fixation of the implant and preservation of the adjacent joint and, to a large extent, its functionality.

The development of 3D computer technologies provided an opportunity to design personalized shortened components of endoprostheses to carry out these surgical interventions. Our case report is a case of surgical treatment of a patient with an extensive lesion of the tibia by fibrosarcoma. To manage the post-resection defect, a modular knee joint endoprosthesis was used, combined with a distal tibial component, that was fabricated custom-made on the basis of 3D computer modeling and hybrid fixation.

Purpose To evaluate the prospects of using a personalized distal tibial component of hybrid fixation, fabricated using 3D computer modeling, in subtotal proximal resection of the tibia with replacement of the defect with a modular knee joint endoprosthesis, we report a clinical case of radical surgical treatment of a patient with an extensive bone tumor lesion due to fibrosarcoma.

MATERIAL AND METHODS

A 45-year-old female patient applied in 2019 with complaints of a painful tumor-like mass in the right lower leg. From the anamnesis, it became known that since 2004 she had been complaining of radiating pain in the right leg. She received treatment from a neurologist with effect for dorsopathy with protrusions of the intervertebral discs of the lumbar spine, lumboischialgia. In December 2018, she noted the appearance of a painful tumor-like formation on the anterior surface of the right lower leg in the middle third, which slowly increased. She was examined at the place of her residence, and a neoplasm of the right tibia was revealed radiographically.

Visual examination revealed a tuberous, sharply painful tumor-like formation that was 3 cm in size and protruded under the skin on the anterior surface of the right lower leg in the middle third.

According to CT data, a significant part of the diaphysis and partially of the proximal metadiaphysis of the right tibia are obturated for 250 mm with pathological tissue with areas of destruction of the cortical layer, its uneven thickening and cancellous reorganization from the side of the endosteum. In the middle third of the tibia, the cortical layer was somewhat swollen, there were zones of lytic destruction with pathological masses entering the surrounding soft tissues and periostitis (Fig. 1).

MRI detected an inhomogeneously hyperintense (hypointense in T1WI) formation that occupied the entire diameter of the bone with uneven involvement of the cortical layer in the diaphysis of the right tibia and extension to the proximal metadiaphysis. In the middle third, destruction of the cortical layer with perifocal edema and periostitis was noted. The pathological substrate extended beyond the bone with the formation of a soft tissue component (Fig. 2).

According to the results of PET/CT with 18F-FDG, a neoplastic formation with a high pathological metabolism was determined in the medullary canal of the diaphysis and proximal metadiaphysis, SUVmax = 11.9; in the middle third of the diaphysis, there was an interruption of the cortical layer with the exit of the neoplasm beyond the bone. There were no signs of regional or distant metastatic lesions.

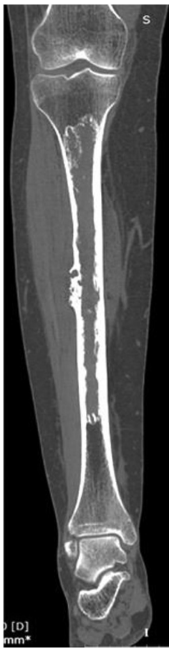

Fig. 1 CT image of the tumour involving a large part of the tibial diaphysis and proximal metadiaphysis (frontal section)

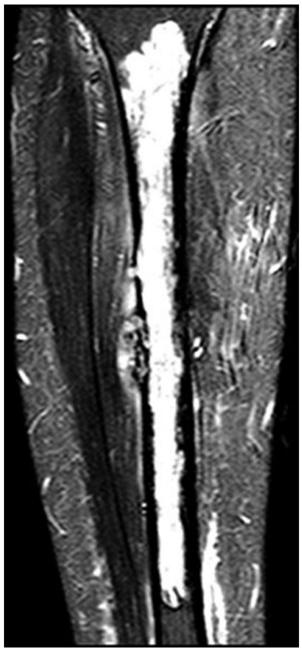

Fig. 2 MRI- image of tibial tumour extension (STIR, frontal section)

A trephine biopsy of the neoplasm of the right tibia was performed, a histological conclusion was confirmed fibrosarcoma G1 and the diagnosis was established: fibrosarcoma of the right tibia cT2N0M0 G1 stage IB.

The clinical case was discussed at the council, and a decision was made on surgical treatment tactics. Based on the prevalence of the tumor process, a distal fragment of about 6 cm long should have been left from the tibia due to the operation of a radical volume. Given the relatively young age of the patient, it was decided to save the ankle joint. Therefore, using 3D computer modeling, a custom-made short distal component was fabricated based on hybrid fixation (including an intramedullary locking cemented 4.5-cm long stem and two bone plates), as well as a set of instruments for its implantation (Fig. 3).

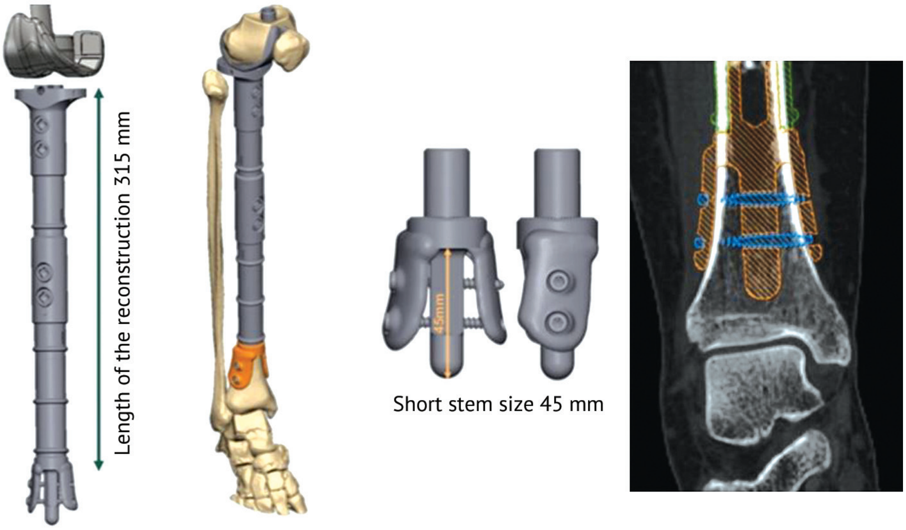

Fig. 3 Diagram of the modular prosthesis of the knee with custom-made distal component based on hybrid fixation

Surgical intervention was performed including subtotal resection of the right tibia and arthroplasty of the right knee joint.

An incision was made in the skin and subcutaneous adipose tissue, bordering the site of the biopsy, from the anteromedial surface in the lower third of the thigh, bypassing the patella from the medial side, then along the anterior surface of the lower leg throughout its entire length. The tibia, together with the tumor, was ablastically separated, the sample was harvested as a single block, and sent for a planned morphological study. Further, the distal tibia was processed with rimmers and with the help of a Volkmann spoon. The custom-made distal component was fixed with bone cement and locking screws.

Using standard instruments, the distal femur was treated under for implanting the proximal component of the endoprosthesis. The medial belly of the gastrocnemius muscle, required for covering the endoprosthesis, was separated. The endoprosthesis was assembled and installed (Fig. 4).

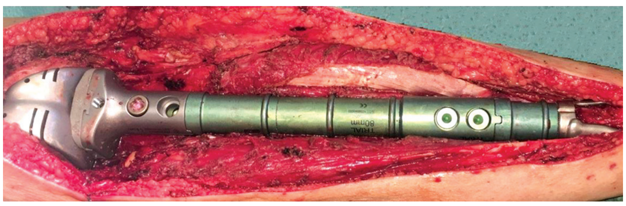

Fig. 4 Photo of the modular prosthesis with fitted components in the wound upon tumor resection

With the help of lavsan ligatures, the reconstructive lavsan sheath was fixed to the proximal part of the endoprosthesis, and was also tightened in the middle. The patellar ligament was fixed to the sheath. A muscular case for the endoprosthesis was thus formed (Fig. 5). The wound was sutured with the installation of drains to the endoprosthesis and gastrocnemius muscle. The operation time was 205 minutes, blood loss was 300 ml.

Fig. 5 Image of the wound after covering the implant with a medial head of the gastrocnemius and formation of the muscle bed

The postoperative pathological study confirmed the initial diagnosis.

In the postoperative period, dressings, antibiotic prophylaxis, and prevention of thromboembolic complications were administered. The wound healed by primary intention, the sutures were removed on the 14th day after surgery.

On the second day after the operation, a rehabilitation program was initiated; the patient started doing physiotherapy exercises. On the 10^{th} day, the patient was verticalized in orthoses for the knee and ankle joints, with fixation of the lower leg at an angle of 180° and feet of 90°. In the following days, measured walking with crutches was prescribed, with a partial load on the operated limb.

RESULTS

One month after the operation, the condition of the patient did not required wearing the ankle joint orthosis, the range of motion in the knee joint increased in a dosed manner due to the adjustable hinges by 20° every 10 days, and the load on the operated leg gradually increased. The patient was allowed to move without additional support and orthosis three months after the operation.

At the control clinical follow-up and ultrasound study three months after the operation, signs of suppuration and tumor growth were not detected. The patient moved independently, limping slightly on her right leg. Range of active movements in the ankle joint was: plantar flexion 30°, dorsal extension 5°; in the knee joint, flexion was 100°, the deficit of active extension of the lower leg in the sitting position was 8°.

The functional result according to the MusculoSkeletal Tumor System (MSTS) was 74 %.

A follow-up examination after a year showed no progression of the tumor. The patient moved independently, without lameness. Movements in the ankle joint were: plantar flexion 40°, dorsiflexion 20°. Tibial flexion was 120°, active extension 5°. The functional outcome for MSTS was 83 %.

Radiography showed that the endoprosthesis was stable, no migration of its components or break of their integrity was noted (Fig. 6, 7).

Fig. 6 X-rays of the distal tibia in two views, one year follow-up

Fig. 7 X-rays of the proximal tibia in two views, one year follow-up

DISCUSSION

Fibrosarcoma is a mesenchymal tumor, the morphological substrate of which is atypical spindle-shaped cells grouped into bundles and fibrous stroma of thin collagen fibers, often forming moiré structures, arranged in a herringbone pattern that do not form osteoid or cartilaginous matrix. Tumor tissue invasively spreads in bone structures, destroying them. The degree of cell atypism and the amount of collagen stroma may vary greatly.

Radiologically, the tumor manifests itself as lytic foci of destruction, located more often in the metaepiphysis or metadiaphysis of long bones, less often in the diaphysis, in most cases intraosseously, although occasionally there is also periosteal localization [9, 10, 11]. Fibrosarcoma was distinguished from the group of sarcomas by Budd and McDonald in 1943 [12]. Unlike osteosarcoma and Ewing's sarcoma, this tumor most frequently affects people aged 20 to 60 years, males are somewhat more often affected. Elderly patients often develop secondary fibrosarcoma on the background of benign diseases (Paget's disease, fibrous dysplasia, etc.) or as a result of previous radiation therapy. Long bone involvement accounts for more than half of fibrosarcomas, with the metaphyseal region being the most frequently involved. Spread of the tumor to the epiphysis or diaphysis is a systematic phenomenon, while isolated diaphyseal lesions are less common (about 10 %). The tibia ranks second after the femur among the most common locations of fibrosarcoma [10, 11, 13].

Despite the rarity of the tumor, prognostic factors for the course of the disease have been studied to date. Thus, in a statistical analysis of the SEER database, which includes 2235 patients with fibrosarcoma, it was found that the most significant independent factors of 3-year and 5-year survival were the grade of malignancy of the tumor, its size, stage of the disease and the possibility of radical surgery, as well as age [14].

The surgical method is the leading one in the treatment of bone fibrosarcoma. There are no recommendations for chemotherapy treatment or radiation therapy. Preoperative and postoperative chemotherapy in stages IIB and III was proposed, but the effect was noted only in a small number of patients [15].

In our case, the clinical and radiological manifestations of the tumor were atypical for high-grade sarcomas: the tumor developed slowly and affected the tibial shaft over a large extent, destroying it completely and forming a soft tissue component only in a limited area. The morphological picture turned out to be characteristic of G1 low-grade fibrosarcoma, which correlated with the clinical course and radiological findings.

Low grade of malignancy, stage IB of the disease, limited destruction of the cortical layer with the possibility of performing a radical operation are predictive factors for a high probability of 5-year or longer survival. The tactic of surgical intervention chosen by us was subtotal resection of the tibia together with the tumor and placement of a modular endoprosthesis with a custom-made distal component was based on the favorable prognosis of the disease course and a relatively young age of the patient. The technique of resections for malignant tumors of the tibia and implantation of modular or individual knee joint endoprostheses for resection bone defects has been much developed in the recent years. The data on objective assessment of long-term results are available in a significant number of patients.

The use of modern surgical approaches and endoprostheses allows achieving positive mid-term and long-term results in most patients [16]. Thus, Myers et al. who performed reconstruction for resection of the tibia and replacement with modern designs of rotating-hinge endoprostheses, the need for repeated operations at 5 and 10 years was 12 and 25 %, respectively, and aseptic instability at 10 years occurred in only 3 % of cases [17].

On the other hand, the problem of implant fixation in extended resections of long bones has not yet been solved. If it is impossible to use standard stems due to a small length of the remaining bone fragment, the risk of endoprosthesis instability increases dramatically.

Streitburger et al. analyzed the experience of using 8 types of implants for extensive resections of the femur and tibia or localization of tumors near the joints in patients with malignant tumors. The authors provide data on a high number of postoperative complications, especially aseptic instability, for implant fixation in the metadiaphyseal or metaepiphyseal region. Direct transfer of the classical stem design of modular endoprostheses to short stem design does not justify itself. According to the researchers, further work to improve the personalized components of endoprostheses will enable their more active implementation in practice [18].

To date, a number of studies have analyzed the positive mid-term and long-term results of the use of Compress endoprostheses with short intramedullary stems of the original design for extended resections of long bones. Their use enables to preserve unchanged a significant part of the diaphyseal part remaining after resection, and to expand the possibilities of subsequent revision operations in case of mechanical complications, and allows postponing a more traumatic bone extirpation [19, 20]. On the other hand, their implantation is impossible in the metadiaphyseal and metaepiphyseal parts of long bones; that fact significantly narrows their application.

It has been shown that additional locking of shortened stems significantly increases the reliability of fixation and reduces the risk of aseptic instability of endoprostheses in extended resections.

Thus, according to the results of Dieckmann et al. in 15 patients with extensive resections of the distal femur, instability of the thickened locking short proximal stem was noted only in one case 58 months after implantation [21]. In the work of Zhao et al., to fix the endoprosthesis in the distal metaepiphyseal part of the tibia in 5 patients, individual components fabricated by 3D modeling were used, which are short wide tibial stems with a highly porous coating and locking, with an average length of 2.6 cm. At 6-month follow-up, complete osseointegration and reliable fixation of the implant were achieved in all cases [22].

Another direction in the development of implants for extensive resections of long bones is the combination of a short intramedullary component with sufficiently massive bone plates to increase the contact of the structure with the remains of the bone and achieve greater primary stability.

In the work of Hanna et al., extensive defects of the femur were replaced with implants with shortened cemented stems, supplemented with one or two bone plates fixed with screws. After an average follow-up period of 120 months, only one case of aseptic instability was observed among 18 patients, which required a revision after 89 months [23].

The results of the use of endoprostheses for extensive resections of the humerus, femur and tibia in patients with malignant tumors were studied by Stevenson et al. The use of short cemented stems in combination with bone plates with a mean follow-up of 8.8 years was accompanied by the development of aseptic instability in 3 out of 37 cases. The Kaplan-Meier implant survival assessment showed no significant differences compared to serial modular endoprostheses used for smaller resections of the humerus, femur and tibia [24].

The results of resections of the femur and tibia for malignant tumors located close to the knee joint were studied in the work Liu et al. Custom-made, 3D-modelled, cementless fixation components were used, including an ultrashort stem and juxta-articular plates. Despite the small length of the preserved epiphyses (the average length was 2.6 cm), an excellent functional result was achieved in 10 out of 12 cases after an average follow-up period of 22.5 months. There were no cases of aseptic instability or periprosthetic fractures [25].

Extended resections with preservation of the growth zone were performed by Y.Tsuda in 18 children. Endoprostheses were combined with custom-made components, including a short stem (no more than 5 cm long) and bone plates. The mean follow-up period was 67 months. Despite the significant growth of the skeleton during this period, only one case of in each of the complications of aseptic instability, periprosthetic fracture, and disintegrity of the implant structure was noted [26].

Our design combines some of the techniques described above. The cemented stem provides high primary stability and enables early rehabilitation. The stem, which is locked together with the plates, ensures the stability of the structure to rotational loads and the integrity in the proximal part during long-term operation. The rough coating of fairly massive stems and their close contact with the cortical layer reduces the load on the bone bed and cement mantle of the intramedullary stem.

Clinical and radiographic results one year after surgery show the absence of early aseptic instability and allow us to hope for a positive mid-term result.

If it is impossible to save the distal tibia and ankle joint, extirpation of the tibia with endoprosthesis and replacement of adjacent joints becomes an alternative to mutilation.

Extirpation of the tibia with a tumor lesion is the subject of a limited number of works, mainly relating to case reports.

Despite the authors' positive results of replacement of the entire tibia with endoprostheses, numerous questions remain regarding the surgical technique, a reliable assessment of possible complications, and analysis of mid-term and long-term results [27, 28, 29].

However, in mechanical complications after extirpation of long bones with endoprosthesis replacement, in contrast to their resection, limb-salvage revision surgery is associated with great difficulties, and in relation to the tibia, it is very problematic.

Therefore, the development of design and fabrication of custom-madel short components of endoprostheses for subtotal defects of long bones, in particular the tibia, using 3D computer modeling in malignant tumors is becoming an increasingly urgent task, the first results of which are encouraging.

CONCLUSION

Radical limb-salvage surgeries for primary malignant bone tumors are the "gold standard" of modern oncology orthopedics. The leading place among them is occupied by surgical interventions with the use of endoprostheses, which provide the most complete restoration of limb function.

The use of custom-made implant components fabricated using 3D computer modeling in patients with an extended tumor lesion of long bones and / or a tumor close to the joint is a promising method to delay or avoid more extensive radical limb-salvage operations or amputations and achieve positive functional results.

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Information about the authors:

1. Vadim Yu. Karpenko – doctor-kv@yandex.ru;
2. Anatoly L. Karasev – karaseva81@mail.ru;
3. Alexander F. Kolondaev – klndff@inbox.ru, https://orcid.org/0000-0002-4216-8800, SPIN code 5388-2606
4. Kirill A. Antonov – osteopathology6@mail.ru;
5. Nikita A. Lyubeznov – osteopathology6@mail.ru.