Clinical case
https://doi.org/10.18019/1028-4427-2024-30-3-438-445

Total ankle arthroplasty in an adolescent who suffered hematogenous osteomyelitis of tibia

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Abstract

Introduction Acute hematogenous osteomyelitis can persist as chronic infection in pediatric patients due to delayed diagnosis and treatment. An extended bone defect of the limb caused by osteomyelitic destruction is a rare clinical scenario in pediatric surgery. Metaepiphyseal and articular involvement suggests specific and long-term treatment with a high risk of disability.

The objective was to report a clinical case of a teenager who suffered chronic hematogenous osteomyelitis of the tibial bones and raise awareness of primary diagnosis and treatment.

Material and methods A clinical case of a teenager treated for extensive destruction of the tibial bones caused by chronic hematogenous osteomyelitis is reported. Clinical and functional evaluation of the effectiveness was produced at the stages of treatment.

Results A positive functional result was achieved due to staged surgical treatment including radical debridement of the chronic infection nidus followed by total ankle arthroplasty.

Discussion Acute hematogenous osteomyelitis persisting as chronic infection could be caused either by a wait-and-see strategy when the patient first sought medical help or a long-term follow-up. There is no consensus on the use of reconstruction or replacement of large long bone defects extended to the joints caused by chronic infection. Staged treatment including total joint replacement with custom-made endoprosthesis can be one of the options.

Conclusion Staged surgical treatment including radical debridement followed by delayed total joint replacement with custom-made endoprosthesis provided satisfactory functionality for the limb without signs of infectious and inflammatory activity at a follow-up period of more than a year.

Keywords: joint replacement, ankle joint, osteomyelitis, cement spacer, surgical site infection, pediatric orthopaedics, clinical case

For citation: Zorin VI, Berdes AI, Glukhov DA, Mushkin AYu. Total ankle arthroplasty in an adolescent who suffered hematogenous osteomyelitis of tibia. *Genij Ortopedii*. 2024;30(3):438-445. doi: 10.18019/1028-4427-2024-30-3-438-445

INTRODUCTION

Acute hematogenous osteomyelitis is a common invasive infection encountered in the pediatric population [1, 2, 3]. Despite the decreased incidence of the disease in recent years and the success of antibacterial therapy, the risk of transition from an acute process to a chronic condition is high and is estimated at 30–40 % [2, 3]. Regardless of the cause, chronic bone infection remains a difficult and challenging problem; relief as a result of treatment can lead to permanent disability of the patient due to severe orthopaedic consequences [4].

The case report demonstrates the importance of timely diagnosis, routing and treatment of an adolescent patient with acute hematogenous osteomyelitis of the tibia and associated orthopaedic complications.

The objective was to report a clinical case of a teenager who was treated for chronic hematogenous osteomyelitis of the tibial bones and raise awareness of primary diagnosis and treatment.

MATERIAL AND METHODS

A 17-year-old patient, a resident of the regional center, was seen by the authors and presented with pain, swelling, a fistula in the right leg and failed weight-bearing on the right lower limb.

The onset of the disease occurred at the age of 15 years (2018) in the form of acute pain, swelling of the lower third of the right tibia, hyperthermia of 38 °C. He was examined by a pediatric surgeon at a regional hospital and received outpatient treatment including antibacterial (amoxicillin + clavulanic acid per os) and anti-inflammatory therapy. He was hospitalized five days later due to persistent symptoms and diagnosed with acute hematogenous osteomyelitis of the right tibia. According to the documentation provided, drilling of the distal metadiaphysis of the right tibia was produced and antibacterial therapy recommended including ceftriaxone, gentamicin, lincomycin, metronidazole. There were no microbiological findings reported. He was followed up by a pediatric surgeon on an outpatient basis after discharge from the hospital. A fistula developed in the lower third of tibia within three months. The patient changed dressings and underwent repeated courses of antibiotics.

The patient was admitted to the pediatric surgical department of a multidisciplinary hospital, Moscow, at four months and was diagnosed with chronic hematogenous osteomyelitis of the lower third of the right tibia. According to the documentation, bone re-drilling was produced, *St. epidermidis* identified with no data on sensitivity, a course of antimicrobial therapy with ceftriaxone, amikacin, ciprofloxacin administered. The patient was discharged from the hospital and an elective surgical treatment offered with no specific description of the procedure. He was followed up by a pediatric surgeon for 18 months, seen by an orthopaedic surgeon and an oncologist who ruled out a tumor process and recommended debridement.

The patient was seen by the authors of the publication after 22 months from symptom onset. On admission he had evidence of normal health with no clinical signs of a systemic inflammatory reaction; normothermia; used crutches with no support on the right tibia. Measurements of an absolute length of the lower limbs showed D < S by 1 cm due to shortening of the tibia. The patient had neither active nor passive movements in the right ankle joint with painful attempts to move the limb. He was unable to bear weight on the limb due to pain. Soft tissues were swollen in the lower third of the right tibia and the ankle joint. He had hyperesthesia on the medial and the anterior portions of the tibia, a fistula-ulcer with scanty serous-hemorrhagic discharge (Fig. 1, a). The patient could maintain movements and sensitivity in the toes. Imaging examination with radiography and CT scan demonstrated diffuse lesion of the distal epimetadiaphysis of the right tibia and widespread fine-focal destruction, larger cavities, sequestration, damage to the articular surface and narrowing of the joint space (Fig. 1, b).

Fig. 1 Photo and radiographs of the patient produced on admission showing (*a*) appearance of the limb; (*b*) lateral view of tibia and the ankle; (*c*) sagittal CT slice

RESULTS

Based on the nature and the duration of the disease the condition was recognized as chronic osteomyelitis of the tibia resulting from acute hematogenous osteomyelitis. Staged treatment was indicated to the patient due to persistent local signs of an active inflammatory process, severely impaired functionality of the ankle joint and impaired capacity to maintain weight on the lower limb. Radical debridement was produced at the first stage.

The surgical treatment performed for the patient included osteonecrectomy of the lower third of the tibia with extensive resection of the distal epimetadiaphysis of the tibia within healthy tissues, placement of a gentamicin-loaded cement spacer, excision of a fistula-ulcer at the right ankle joint, stabilization of the tibia and the foot with Ilizarov frame using wires and half-pins (Fig. 2). The surgical material (necrotic bone tissue) was sent for bacteriological and histological examination.

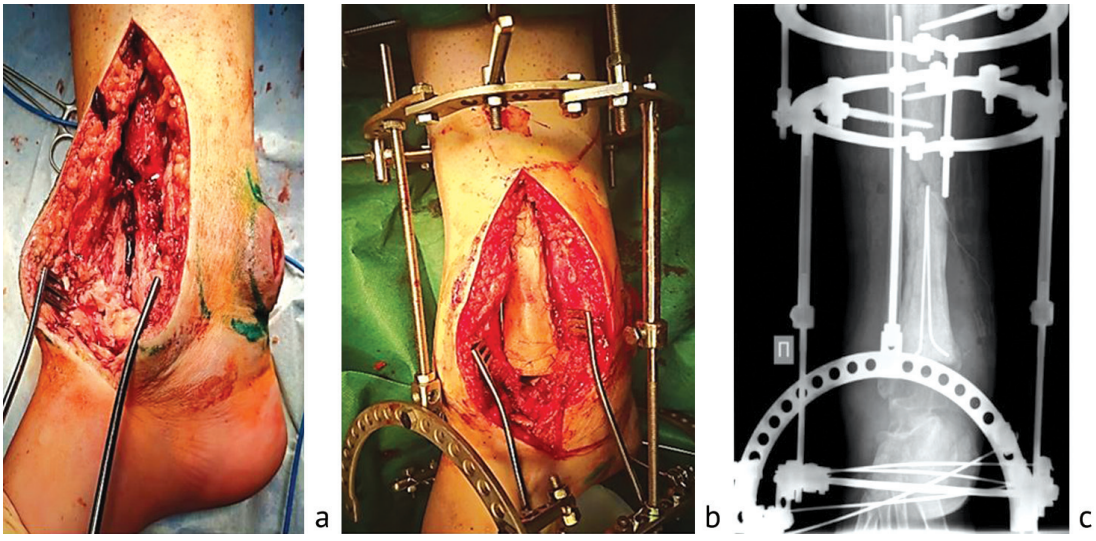

Fig. 2 Photo and radiograph of the patient's distal tibia on the right: (*a*) diffuse necrotic changes in the distal epimetadiaphysis of the tibia; (*b*) cement articulating spacer placed after debridement; (*c*) postoperative radiograph showing tibia and the foot fixed with frame using wires and half-pins

Bacteriological examination detected no microflora growth. Histological examination showed bone and cartilage degeneration, fragments of fibrous and granulation tissue with moderate lymphoplasmacytic infiltration, leukocytes indicating chronic nonspecific osteomyelitis (Fig. 3).

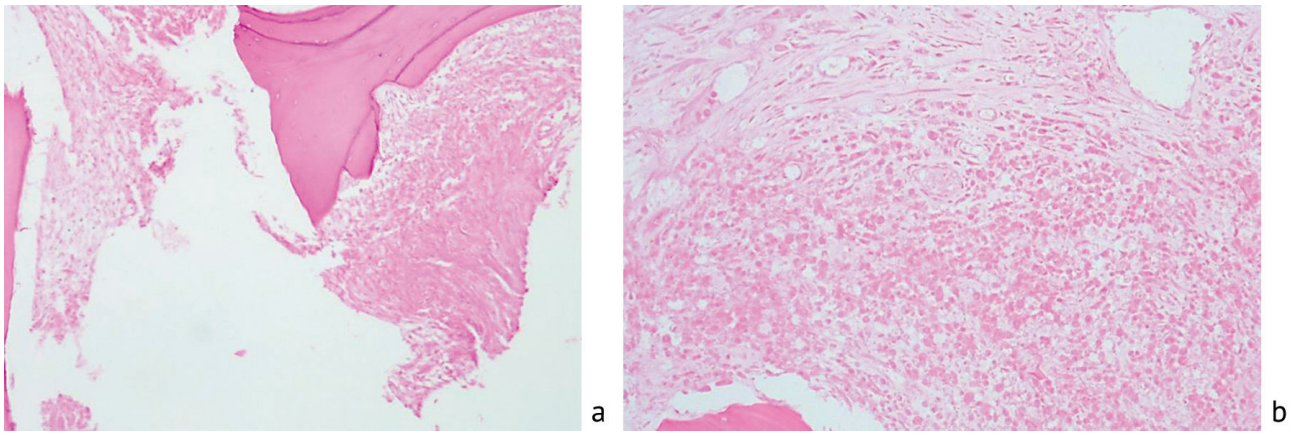

Fig. 3 Histological preparations of surgical material showing (*a*) dystrophic changes in bone tissue, foci of coagulation necrosis without a cellular reaction, stained with hematoxylin and eosin, ×100; (*b*) inflammatory infiltrate in the interosseous cells, stained with hematoxylin and eosin, ×200

Hyperthermic syndrome persisted for seven postoperative days because of antibacterial therapy using cefazolin, rifampicin, metronidazole. With the assistance of a clinical pharmacologist, the therapy was replaced with amoxicillin with clavulanic acid and vancomycin. The hyperthermic syndrome relieved after days. The antibacterial therapy lasted for 4 weeks with regression of tissue edema and exudative process in the fistulectomy area and healing of postoperative wounds. The patient received medication therapy to address local trophic processes (pentoxifylline, aprotinin). The sutures were removed after 20 days. No clinical and laboratory signs of exacerbation of the inflammatory process were noted at 9 months, and the next stage of surgical reconstruction was scheduled.

Two surgical options offered to repair 12 cm defect of the distal epimetadiaphysis of the tibia included bifocal compression-distraction osteosynthesis and ankle fusion and total replacement of the distal tibia and the ankle joint to regain weight-bearing capacity. Strategy options, potential complications, risks and outcomes were discussed with the patient and legal representatives, who refused bifocal osteosynthesis and arthrodesis.

A tailored modular constrained implant of the ankle joint and distal metadiaphysis of the tibia was manufactured to repair the extensive post-resection defect (Fig. 4).

Fig. 4 Design of total ankle replacement

The external fixation device was removed due to long-term clinical and laboratory remission and a trephine biopsy of the tissues of the lower third of the tibia, talus and tibia was performed. No growth of microorganisms was detected with bacteriological examination of biopsy specimens. After 14 days. After dismantling, the cement spacer was removed at 14 days with the external fixation off and the ankle joint was totally replaced. No intraoperative macroscopic signs of infection and inflammation were detected. The spacer bed was lined with a hypervascularized connective tissue membrane (Fig. 5, a).

The tibial shaft and trochlea tali were resected as estimated. The proximal and distal modules were placed and fixed by press-fit, connected to each other after positioning the fitting components (Fig. 5, b, c). The postoperative period was uneventful, the surgical wound healed by primary intention. Microbiological analysis of the surgical material showed no growth of microorganisms. Gradual weight-bearing on the limb was encouraged after two weeks to be followed by full weight-bearing at one month.

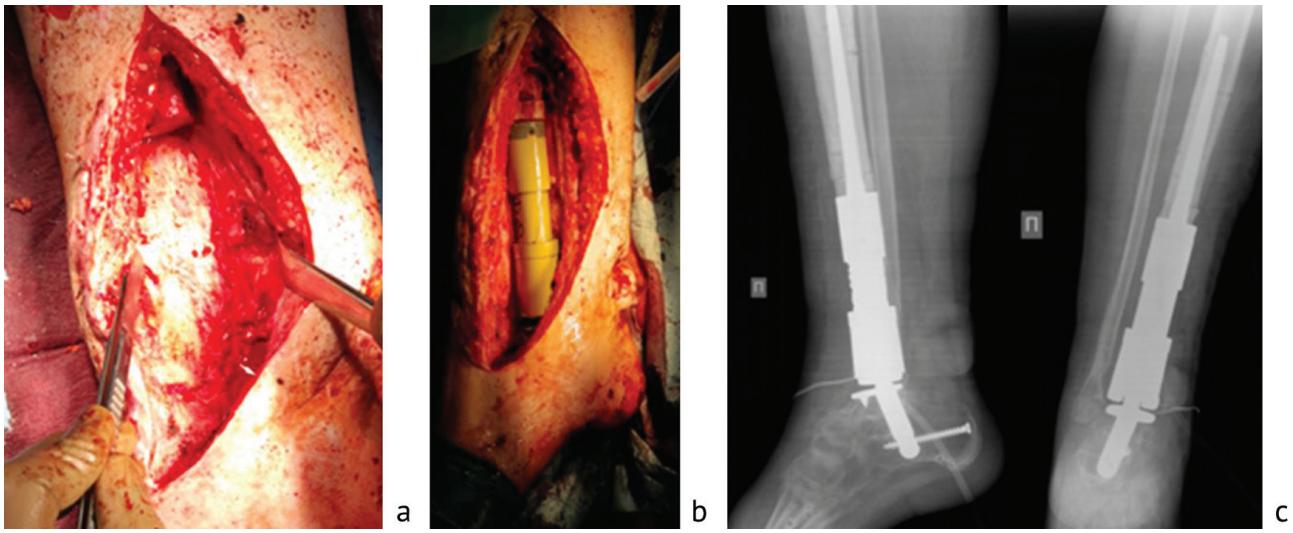

Fig. 5 Intraoperative photographs and radiographs showing: (*a*) the spacer bed being represented by a hypervascularized connective tissue membrane, (*b*) endoprosthesis implanted, (*c*) AP and lateral views of the tibia

The patient could walk without means of support at 14 months and experienced a slight feeling of discomfort walking over a distance of more than 2 km. There were no local signs of inflammation, the axis and length of the limb were restored. The implant was adequately positioned radiologically with no signs of peri-implant bone resorption (Fig. 6).

Fig. 6 Photographs and radiographs of the patient's tibiae and the ankle at 14 months of total ankle replacement (3 years from the onset of the disease)

The quality of life was evaluated preoperatively and at 14 months using the SF-36 questionnaire survey [5] at the time of the first admission to the clinic with physical functioning scoring 25 and 49, respectively, and the mental health scoring 37 and 57. The foot function scored 50 and 77 preoperatively and postoperatively on the AOFAS scale [6], respectively.

DISCUSSION

Diagnosis and treatment of acute hematogenous osteomyelitis in children are presented in modern educational and scientific publications [2, 3], while chronic nonspecific osteomyelitis, recurrent osteomyelitis are not common in pediatric practice. The development and progression of the condition are often accompanied by delayed diagnosis and difficulties in selection of treatment options [4].

Most authors suggest that chronic osteomyelitis should be treated with radical resection of the osteonecrotic bone to be followed by grafting the residual bone cavity with antibiotic-loaded osteoplastic material [7–9] to prevent hematoma, an ideal breeding ground for microorganisms,

and create a local antimicrobial depot in a therapeutic concentration and promote bone restoration. However, with extensive destruction requiring extensive resections, the resulting bone defects cannot be fully repaired using bone graft. Bone reconstruction can be produced using bifocal osteosynthesis with an external fixation device [10] that can be optionally combined with intramedullary nailing [11, 12], Masquelet technique [13] and microsurgical technique can be employed for soft tissue defects [14]. The techniques are practical for impaired diaphysis and/or metaphysis of long bones to restore weight-bearing capacity and the mechanical axis of the limb to help patients return to a normal lifestyle. With the involved epiphysis, dysfunction of the joint becomes an additional problem leading to disability.

Hikichi et al. reported a clinical case of successful arthroplasty used to treat local infection of the articular surface of the pylon [15], while extensive ankle injuries can be repaired with arthrodesis [16]. Total ankle arthroplasty (TAA) has become an alternative to arthrodesis and has been associated with the current technological advancements, new materials and design options with the outcomes being comparable with the two techniques [17–19]. The survival rate of implants is 81–97.7 % at 5 years and 69–86 % at 10 years [20, 21]. Based on their own experience Mikhailov et al. reported an algorithm for the choice of arthrodesis and total ankle replacement [22]. A study of static and dynamic parameters of supportability and the gait showed that the latter was approaching a physiological level at 24 months of ankle replacement, that suggested maintaining ankle mobility [23] and arthrodesis could be offered for complications of ankle replacement [24].

The problem is that the experience presented relates to classical arthroplasty for primary and secondary crusarthrosis without significant bone destruction. Replacement of a bone segment and simultaneous joint replacement is performed for oncological cases.

Sokolovsky et al. reported the experience of using modular ankle implants in 20 patients over 11 years with mechanical and non-mechanical complications noted in 39.4 % of cases, requiring repeated interventions for complete replacement of the implant [25]. Over 10 years, Karpenko et al. reported the use of staged operations in 3 (33.3 %) of 9 cases of modular ankle replacement due to instability and infectious complications [26]. Zhao et al. performed a systematic literature review and reported better functional results with biological reconstruction of the distal tibia and ankle arthrodesis and a comparable rate of complications compared to modular arthroplasty for tumors [27].

No data on the ankle joint replacement resulting in osteomyelitis could be found in the available literature. In our case, bifocal osteosynthesis with the external fixation device and ankle arthrodesis was the preferred option to restore the limb support. However, the patient and his parents chose the alternative option after discussion of the possible complications, and the treatment allowed the patient to maintain mobility in the joint, at least for the follow-up period. Now the patient is under dynamic observation and remains at risk of developing late complications. However, the positive outcome allows us to expand indications for total ankle replacement towards etiological group of post-infectious lesions and reduce age restrictions on the use of the method.

CONCLUSION

The clinical case demonstrated the relevance of the problem of organizing care for children and adolescents with bone infection and destructive lesions. With outpatient and inpatient care provided by multidisciplinary medical institutions (regional and federal centers), there was a difficulty for specialists to identify an adequate treatment option and that resulted in a long idle period of diagnosis and treatment. This led to a complicated clinical status and chronification of primary acute hematogenous osteomyelitis. The result reported demonstrated the possibility of preserving the support and motor function of the ankle joint with total ankle replacement using a tailored modular implantation after the arrest of the infection. Sufficient experience

and long-term results of ankle replacement are needed for unbiased judgments about the optimal strategy. The design (clinical observation) and the limited of follow-up period of 14 months should be considered as limitations on the reliability of the publication.

Funding *the authors received no specific funding for this work.*

Conflict of interest *None of the authors has any potential conflict of interest.*

Ethical Approval *the authors obtained the informed consent of the patient for examination, treatment, collection, storage and analysis of medical documentation data for scientific and educational purposes, and their publication. the authors confirm that there is no data in the article that is not subject to publication.*

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The article was submitted 16.01.2024; approved after reviewing 05.02.2024; accepted for publication 08.04.2024.

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