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CASE REPORT

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Successful limb-salvage procedure using a bioexpandable prosthesis after infected primary reconstruction of the distal femur in a skeletally immature patient: a case report

Jan Lesensky[1*], Ana C. Belzarena[2] and Michal Benes[3]

Abstract

Background Periprosthetic infections pose a devastating complication in skeletally immature patients treated for an orthopaedic oncological condition. Reconstructive approaches to revision procedures are often limited, and many cases still require amputation.

Case Presentation In this report, we present our unique experience with the bio-expandable MUTARS® BioXpand prosthesis, utilized during the second stage of a revision surgery in an adolescent female patient. Initially, the patient underwent reconstruction using a conventional endoprosthesis following the resection of a high-grade distal femur osteosarcoma; however, she developed a deep infection six months later. During a two-stage revision procedure, the infection was successfully eradicated at the cost of loss of growth potential at also the site of proximal tibia. The initial 5 cm limb-length discrepancy was restored through the application of bioexpandable endoprosthesis, which allowed for an 8 cm gain in bone stock. At the last follow-up appointment, the patient was fully weight-bearing and demonstrated excellent clinical outcomes, with no evidence of infection or tumor recurrence.

Conclusion This successful limb-salvage procedure indicates that bioexpandable endoprosthesis may serve as a viable and effective reconstructive option in revision surgery for skeletally immature individuals.

Keywords Bioexpandable prosthesis, MUTARS® BioXpand, Periprosthetic infection, Limb-salvage surgery, Knee revision, Lengthening nail, Precise nail

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Background

Periprosthetic infection is a major complication among oncological patients undergoing endoprosthetic reconstruction following bone tumor resection [1–8]. Immunosuppression resulting from chemotherapy and to some extent radiation therapy, extensive soft tissue excision, and longer operative time make the patients more susceptible to infections [2, 6, 8]. Treatment of an infected tumor megaprosthesis is a lengthy process often necessitating repeated prolonged hospital stays, multiple surgical interventions, and delays in oncological treatment, potentially impacting the overall prognosis [3, 6]. Cases complicated by deep infections have worse functional outcomes, and in many instances culminate in a devastating limb amputation [2, 3, 6, 8].

Limb-salvage surgery in the pediatric age group poses a specific difficulty due to the immature skeletal system represented by small bone dimensions, limited anchoring conditions for prostheses, and, in particular, the presence of open growth plates. Equal limb length at maturity, together with acceptable functional outcomes, are the main objectives in contemporary practice [9]. Introduction of expandable endoprostheses has been pivotal for addressing the concerns of limb length discrepancy resulting from resection of the affected growth plate. The first expandable endoprostheses utilized either repeated open or percutaneous lengthening and were associated with an elevated risk of periprosthetic infection [4, 10]. Technical advancements delivered the creation of non-invasive expandable implants which significantly decreased the rate of infection [1]. Nonetheless, deep infections cannot be fully dissipated, partially so due to the above-mentioned risk factors that remain in place for this patient population. Currently there is a growing need for implant alternatives that can be implemented in revision surgeries that can address the unique physiological requirements of skeletally immature patients suffering from periprosthetic infection. Despite the recent utilization of non-invasive expandable endoprostheses in revision surgery for deep infection [11], data is limited regarding the application of bioexpandable prosthesis to date.

Here we report the complex case of a young female patient who underwent resection of a malignant tumor of the distal femur and developed a periprosthetic infection of the primary reconstruction and was subsequently successfully treated with a two-stage revision using a MUTARS® BioXpand prosthesis (Implantcast, Germany).

Case report

Patient history

An 11-year-old female patient diagnosed with high-grade osteosarcoma of the left distal femur underwent wide resection and subsequent prosthetic reconstruction with an endoprosthesis (Fig. 1). Six months after the index surgery, immediately after completing the last cycle of adjuvant chemotherapy, the patient was admitted to the hospital with septic shock due to an acute deep infection. Extensive pus formation led to skin tearing on the anteromedial aspect of the knee exposing the implant.

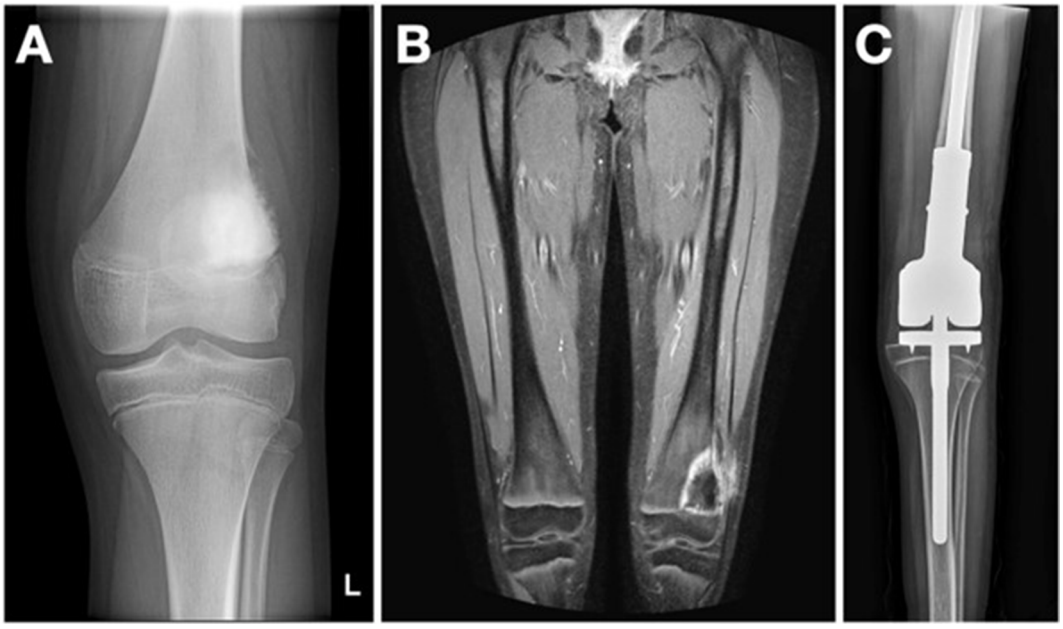

Fig. 1 X-ray (**A**) and magnetic resonance imaging (**B**) showing the tumor of the left distal femur. Resection was followed by reconstruction with growing endoprosthesis (**C**)

First stage - explantation of the infected endoprosthesis

Surgical revision was indicated for infection control. The original implant was removed, and thorough debridement of soft tissues and intramedullary canals was performed, including high-pressure lavage irrigation. A reinforced temporary cement spacer with antibiotics was used to fill the defect after explanting the prosthesis. (Fig. 2). Calcium phosphate beads with Vancomycin (STIMULAN®) were placed inside the medullary canals as well as into the soft tissues. A vacuum-assisted device was used to cover the surgical site. Microbiological and bacterial PCR testing turned positive for polymicrobial flora including *Staphylococcus Epidermidis*, *Neisseria Mucosa*, *Cutibacterium Acnes and Bacillus sp*. Genetic testing turned negative for all tested genotypes of antibiotic resistance. Laboratory tests showed standard antibiotic sensitivity.

Interim period

Empiric antibiotic therapy with Ciprofloxacin and Metronidazole was started. These were switched to Linezolid and Cefixime once cultures were finalized. Clinical improvement with wound healing and normalization of inflammatory markers was achieved. Unfortunately, despite the recommended activity restriction, the patient developed a Salter-Harris type V-like fracture of the proximal tibia due to independent transition to full weight-bearing. This resulted in the formation of a bony bridge across the lateral half of the physis (Fig. 2) eliminating the growth potential of proximal tibia.

The treated lower extremity had now lost the distal femur and the proximal tibia physes, resulting in a 5 cm leg length discrepancy at the time of the planned second stage. The length of the cement spacer was 10 cm, which is too short for an expandable prosthesis as the shortest distal femur implant is 18 cm in length.

The expected length discrepancy at the end of growth was calculated at 10 cm which is unmanageable without the use of a lengthening implant. However, we wanted to avoid resecting additional bone and using a massive implant in the setting of a previous deep infection. A decision was then made to use the bioexpandable MUTARS® BioXpand prosthesis (Implantcast, Germany). The prosthesis is 10 cm long and connected to the FITBONE® extendable motorized nail (Wittenstein, Germany), allowing non-invasive lengthening through callus distraction (Fig. 3). The total lengthening potential of this implant is 8 cm.

Second stage - implantation of the bioexpandable endoprosthesis

The cement spacer was removed and a reconstruction with the BioXpand prosthesis was performed. Since the tibia growth plate was nonfunctional we used a definitive cemented proximal tibial MUTARS component, which is fully compatible with the BioXpand system. A femoral osteotomy site was marked with a Kirschner

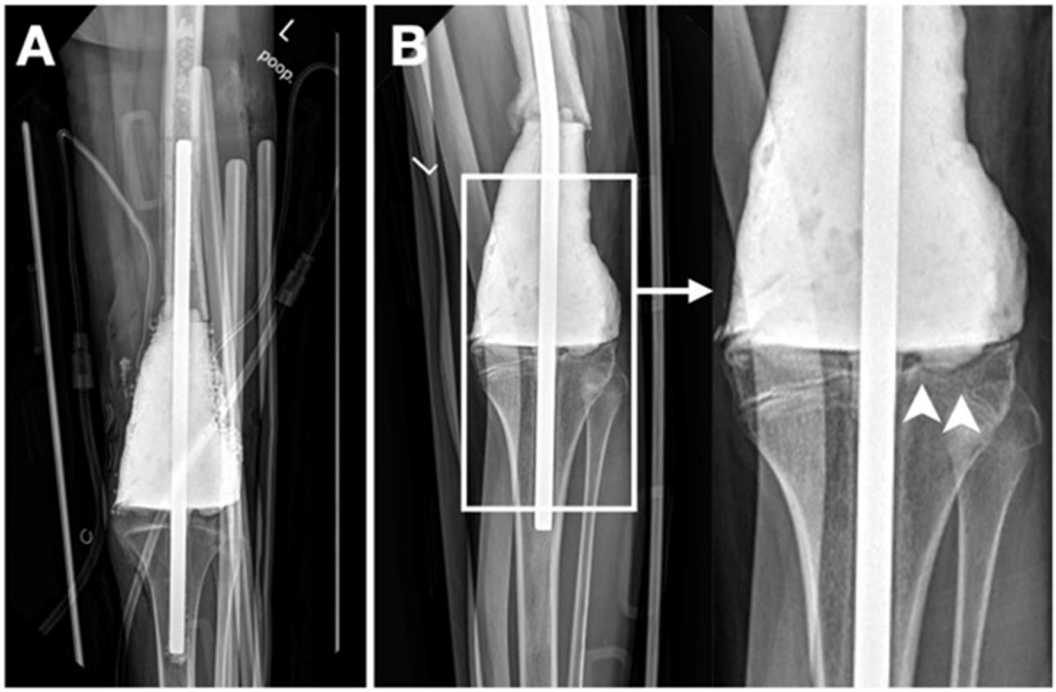

Fig. 2 X-rays images showing the static cement spacer reinforced with a Küntscher nail in situ (**A**), and the Salter-Harris type V-like fracture of the proximal tibia with bone bridging (**B**)

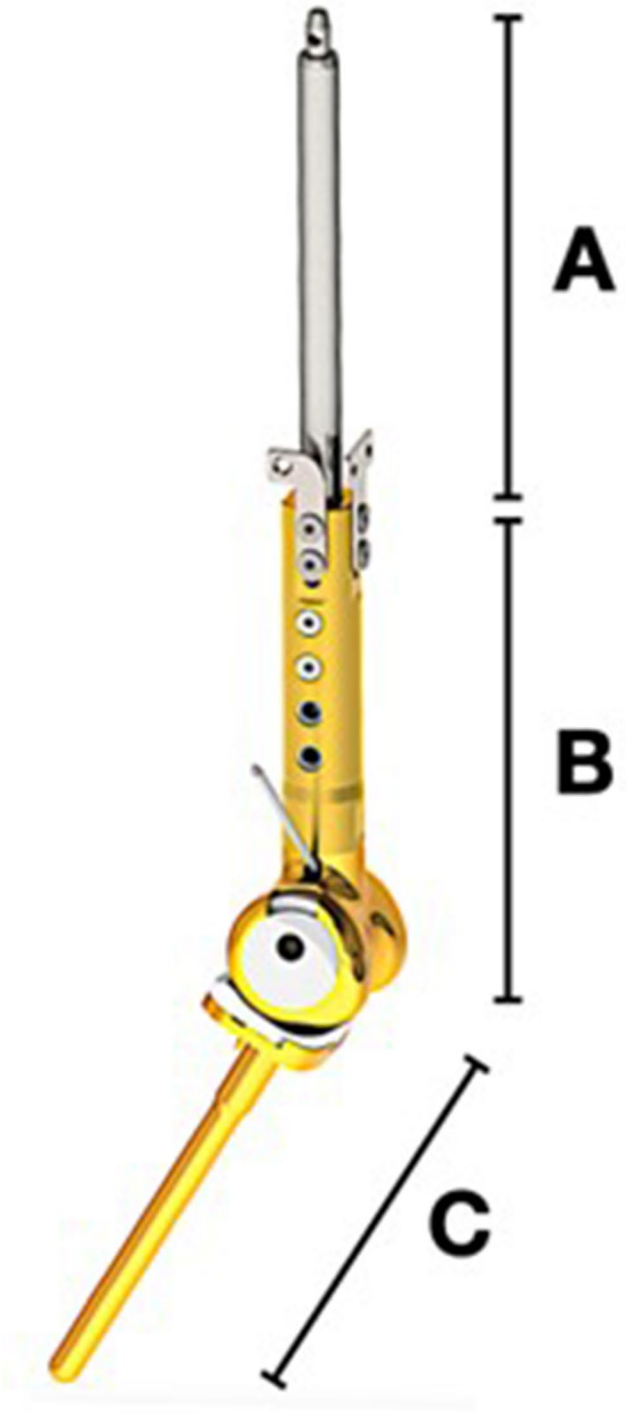

Fig. 3 The MUTARS® BioXpand distal femur prosthesis consisting of FITBONE® motorized nail (**A**), distal femur component (**B**), and tibial plateau component (**C**). Reprinted from Implantcast (n.d.)

wire according to the manufacturer's manual. The osteotomy was performed through a separate anterior thigh incision. The FITBONE® nail was then inserted into the femoral canal and connected to the BioXpand femoral component. The nail was proximally fixed with one

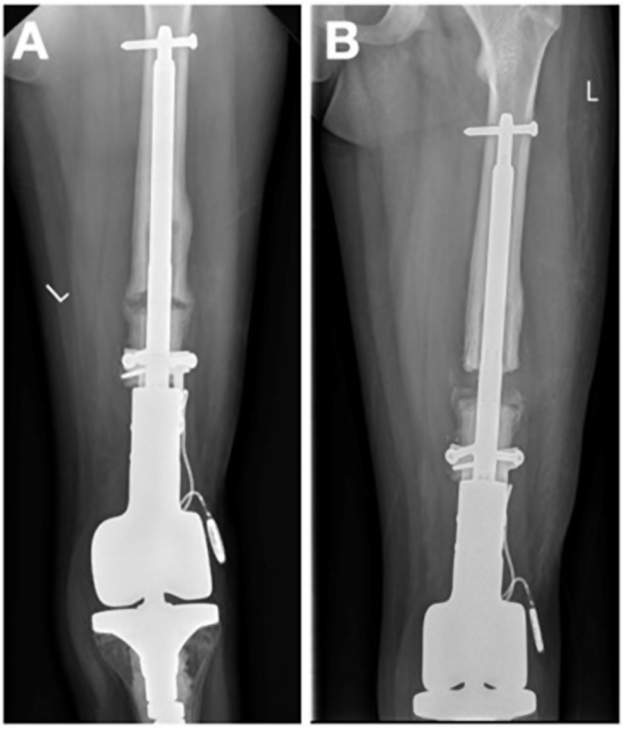

Fig. 4 Anteroposterior (**A**) x-ray images demonstrating the MUTARS® BioXpand implant in situ with bony fusion of the callus encountered during follow-up visit and status after re- osteotomy and elongation by 1 cm (**B**)

bicortical screw, and the distal femoral fragment was fixed to the prosthesis using screws and plates. A receiver for transcutaneous energy transmission was placed subcutaneously on the lateral side of the knee. The implant was lengthened intraoperatively 1 cm. A medial gastrocnemius muscle flap was advanced over the femoral component. All intraoperative cultures were negative.

Five days after the surgery, the nail elongation at 1 mm per day started. The distraction consisted of external transmission of electromagnetic pulses to the subcutaneous receiver. Unfortunately, a bony callus was observed at the 3-month follow-up at the osteotomy site (Fig. 4). Following a discussion with the family, this was attributed to poor compliance of the family with the recommended protocol. The lengthening intervals were inconsistent, and they avoided using an stethoscope for auditory control.

A re-osteotomy was performed from a lateral incision to protect the medial gastrocnemius flap. The functionality of the lengthening mechanism was verified intraoperatively and the nail elongated by 1 cm (Fig. 4). Postoperatively all subsequent elongations were carried out by the medical team without complications.

Conversion to definitive endoprosthesis

Once the maximum lengthening of 8 cm was reached, an exchange of the bioexpandable prosthesis for the definitive modular MUTARS® Distal Femur MK implant (Implantcast, Germany) was performed. An extended lateral approach including the previous approach used for the re-osteotomy was utilized. The newly formed bone was noted to be firm. Additionally, we enhanced the regenerated bone with iliac crest cancellous graft and bone from the reamers. The final uncemented stem was inserted with good primary stability and secured proximally with two bicortical screws. The final adult-size femoral component was connected to the previously implanted tibial part (Fig. 5). Primary closure of the wound was achieved. All intraoperative cultures were negative.

Outcomes

At the 12-month follow-up after implantation of the definitive endoprosthesis the patient was doing well with no evidence of infection or tumor recurrence. Despite the initial arthrofibrosis resulting from the infection and multiple surgical procedures, the patient progressively regained knee range of motion (0-100°) with full active extension allowing her to independently climb stairs. The affected left leg was 1.5 cm longer in the immediate postoperative period (Fig. 6), however, this discrepancy decreased over time with a subsequent discrepancy of less than 1 cm at the last follow-up. Nevertheless, the proximal tibia growth arrest led to an unequal knee joint level of approximately 3 cm (Fig. 6). At the last follow-up the patient was painless, full weight-bearing and had resumed all daily routine activities including recreational sports. Final Musculoskeletal Tumor Society (MSTS) and Toronto Extremity Salvage Scores (TESS) were 27/30 and 87%, respectively.

Discussion

Implantation of megaprostheses after bone tumor resection, together with the pitfalls of effective oncological treatment, carries a high risk of development of periprosthetic infection [2, 6, 8]. Primary malignant bone tumors are frequent in the pediatric population, with the majority of those arising in proximity to the most active growth plates, especially around the knee [9, 12]. Thus, reconstructive options and complication management must take into account the possible leg-length discrepancy caused by the continued growth of the contralateral limb. The use of non-invasive expandable endoprosthesis in revision surgery for skeletally immature patients has been recommended [11]. In our patient, the presented case describes Henderson type 4 A implant failure with subsequent resolution of the deep infection and successful limb-salvage using a bioexpandable endoprosthesis [13].

Multiple approaches to treating periprosthetic infections exist, such as single-stage or two-stage revision, debridement accompanied by antibiotic therapy while

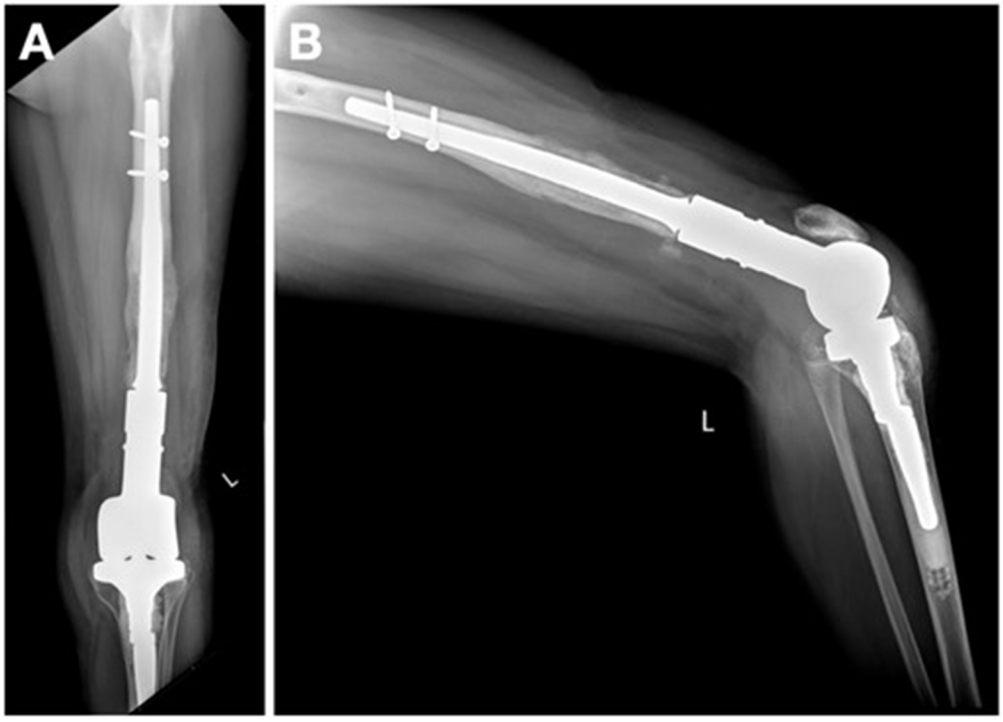

Fig. 5 Anteroposterior (**A**) and lateral (**B**) x-ray images demonstrating the implanted definitive modular MUTARS® endoprosthesis

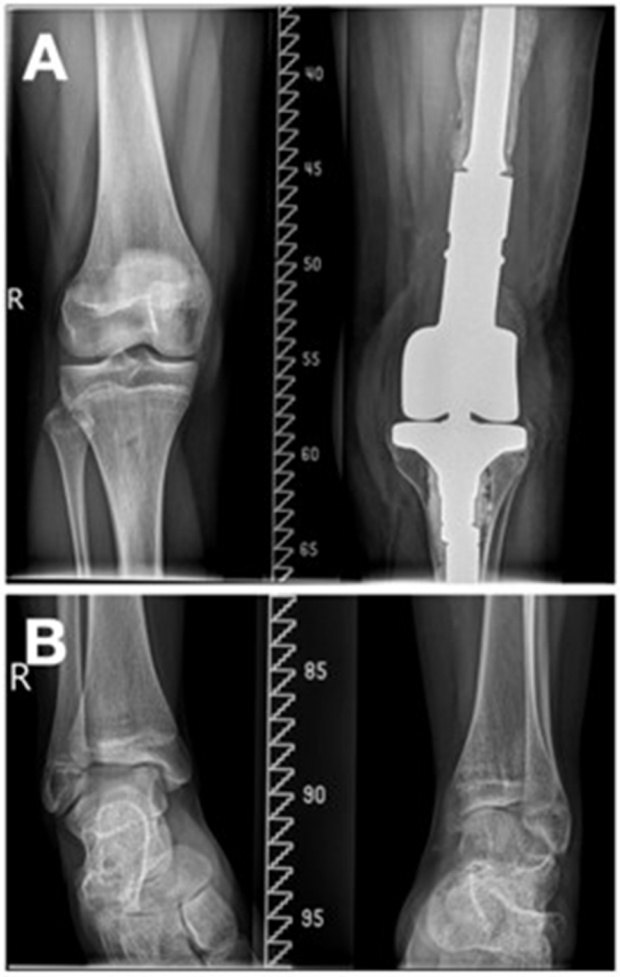

Fig. 6 Final comparative images showing the levels of knee (**A**) and ankle (**B**) joints

retaining the implant (Debridement, Antibiotics, and Implant Retention; DAIR) or its modification with exchange of modular components (referred to as DAIR plus), and amputation [2]. Two-stage revision has been termed the gold-standard for eradicating deep infection in patients treated with megaprostheses for orthopaedic oncological conditions [2, 3, 8]. Furthermore, the early removal of orthopaedic hardware is highly recommended to help achieve adequate infection control [5]. The success rate of the two-stage revision procedures can be as high as 75% [2]. Unfortunately, secondary complications frequently arise following the revision surgery [14–16]. Therefore, patients who undergo a first revision for a deep infection must be counseled regarding the inherent risk for future complications and further need for repeat procedures [7].

Treatment and reconstructive alternatives in children with bone tumors include growing endoprosthesis, osteoarticular allograft, contralateral limb growth arrest or shortening osteotomy, extra-focal distraction osteogenesis, rotationplasty, or amputation [12, 17, 18]. The MUTARS® BioXpand endoprosthesis (Implantcast, Germany) was introduced in 2005 as an alternative for the treatment of bone defects after resection of malignant bone tumors [19]. For this implant, the non-invasive lengthening is performed via a FITBONE® motorized nail (Wittenstein, Germany), which operates as a callus distraction device between the osteotomized bone segments allowing the formation of new bone within the distraction gap. The definitive reconstruction requires multiple surgical steps, including implantation of a temporary prosthesis with a provisional stem and subsequent exchange of the stem for the lengthening nail, or a direct primary implantation of the prosthesis already with the lengthening nail. Replacement with a definitive implant is indicated once the desired leg-length is achieved and the bony callus has maturated enough to hold a definitive stem. The key advantage of this prosthesis, as opposed to traditional expandable implants, is the ability to stimulate the formation of new bone. However, aside from the preliminary report of two patients receiving this novel implant [19], there is no additional data in the available scientific literature. Therefore, to the best of our knowledge, this appears to be the first reported case utilizing the bioexpandable prosthesis for the management of periprosthetic infection in a skeletally immature patient, and possibly also the first case reporting clinical and radiographic outcomes using this reconstructive method.

Although each case has its own peculiarities, according to manufacturer's specifications this implant is suitable for distal femoral bone defects measuring a minimum of 10 cm and allows for a maximum elongation of 8 cm (Implantcast, n.d.). Partial weight bearing restrictions with a limit of 20 kg are required to avoid failure of the lengthening mechanism (Implantcast, n.d.). Other routinely used growing endoprostheses, which harbor the lengthening device within, require larger bone defects to be implanted, usually at least 18 cm. Our patient however had only a 10 cm long defect. In order to fit in an 18 cm long femoral component plus the tibial component, this would have required the sacrifice of an additional 10 cm of a healthy bone. Instead, we managed to not only preserve the bone stock, but even gained an additional 8 cm of newly formed bone. The patient benefitted from this implant since it is shorter, less bulky (which facilitates closure in a small limb), and has reduced metal volume, thereby potentially mitigating the risk of deep infection. Furthermore, the new bone growth assured a stronger implant fixation by the increased endoprosthesis-host bone length ratio, which also offers additional host bone for any potential future revisions [19].

Aside from the deep infection, two additional complications, including one pediatric and one mechanical failure, were encountered during the treatment of our

patient [13]. While weight-bearing with the static spacer, the patient sustained Salter-Harris type V-like fracture of the proximal tibia, which ultimately led to the formation of a bony bridge and subsequent growth arrest. Due to this consequence a conventional cemented tibial stem was implanted during the second stage of revision as a definitive reconstruction of the proximal tibia, as there was no need for preserving the physis. This resulted in a lower tibial plateau and longer femur, which contributed to the unequal joint lines and patella alta. Notably, a study conducted by Sambri et al. on patients undergoing distal femoral reconstructions suggested that an increase in patellar height may enhance knee flexion and alleviate anterior knee pain [20]. Despite a slight discrepancy between knee joint levels, equal limb-lengths were achieved, enabling the patient to ambulate painlessly. Furthermore, the lengthening process was complicated by early bony fusion of the callus, attributed to poor adherence to the self-lengthening protocol, requiring an additional surgical intervention. These two complications underscore the critical importance of strict patient and family compliance while managing such complex cases. Consequently, frequent follow-up appointments and close surveillance are essential to promptly identify any disturbances and to initiate appropriate treatment.

Although, to our knowledge, this is the only case exclusively reporting the utility of bioexpandable endoprosthesis in an infection-related revision procedure, we must acknowledge several limitations. As this report focuses on a single patient, the findings may not be extrapolatable to other individuals with similar conditions. Larger studies are needed to validate the observations and conclusions drawn from this case, thereby comparisons with similar cases or randomized controlled trials are necessary for a more robust analysis. Furthermore, the follow-up period for this case may be insufficient to evaluate long-term outcomes or complications related to this reconstructive option. Although there is scant evidence for the bioexpandable endoprosthesis, the current case supports its use in pediatric revision surgery, making such a report valuable to expand the portfolio of reliable reconstructive options.

Conclusion

In selected cases fulfilling the indication criteria, the implantation of a bioexpandable prosthesis during the second stage of revision surgery for periprosthetic infection may represent a viable and effective option for skeletally immature patients. Unlike the standard growing implants, the bioexpandable prosthesis provides distraction osteogenesis with formation of good-quality bone, which ultimately leaves the patient with a larger bone stock for strong definitive implant fixation and any potential revisions. This case further emphasizes the importance of strict patient and family compliance with frequent follow-ups while using the bioexpandable endoprosthesis.

Author contributions

Conceptualization, Methodology, Formal Analysis, Investigation, Resources, Data Curation, Writing– Original Draft Preparation, Writing– Review & Editing, Supervision, Project Administration: J.L., M.B. Conceptualization, Resources, Data Curation, Writing– Original Draft Preparation, Writing– Review & Editing, Supervision: A.C.B.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

This retrospective observational study of a human participant for a case report was reviewed and approved by Faculty and Hospital ethical committee. Consent for participation was waived given the retrospective nature of this study.

Consent for publication

Consent was obtained for publication.

Competing interests

The authors declare no competing interests.

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