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Proximal tibial replacement with megaprosthesis in the setting of proximal tibial nonunion: A case report

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ABSTRACT

Background: Fracture nonunion is a major concern among an orthopaedic patient population, especially in those who have sustained traumatic fractures involving the tibia. Strong risk factors for nonunion include age, smoking history, and a poor diet. The incidence of nonunion also increases with each additional failed surgical intervention.

Methods: Our retrospective case study involved 56-year-old woman with a history of chronic low back pain, osteopenia, malnutrition, smoking, marijuana use, and alcohol use, who presented with a proximal tibia fracture after a fall, initial treatment included temporization with multiplanar external fixation and subsequent internal fixation. Five weeks later, she presented with atrophic nonunion. She subsequently underwent multiple unsuccessful surgeries to address her nonunion, including open repair with bone grafting and multiplanar external fixation for bone transport. Ultimately, the nonunion was addressed by proximal tibia replacement with megaprosthesis with excellent clinical results.

Results and conclusion: Replacement of a proximal tibia with megaprosthesis is a viable option for limb salvage, especially when all alternative treatments have been unsuccessful.

Introduction

The lack of functional recovery from a fracture after 3 months is an indicator of a nonunion [1], defined by the lack of healing after 9 months and no radiographic indication of healing at 3 months [2]. The risk of nonunion for traumatic orthopaedic fractures is \sim 5–10 % [3,4] and \sim 7.4 % for fractures of the tibia [5]. However, the risk is influenced by many factors [4,6–9], including age, smoking, and poor nutrition [9,10], which should be considered when treating patients with tibial fractures [8,11,12].

First-line surgical management includes resection and debridement of the nonunion to create healthy bleeding cancellous bony surfaces to stimulate healing and maximize surface contact at the fracture site [13]. However, the choice of surgical approach for nonunion fractures depends on the adequacy of reduction, construct stability, and soft tissue condition [14]. Notably, nonunion of tibial fractures is associated with failures of prior surgical interventions [3]. Less common salvage procedures must be considered after others have failed.

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We present the case of a 56-year-old woman with a proximal tibia fracture who underwent multiple revision surgeries, including open repair with bone grafting and proximal tibia resection and multiplanar external fixation to address atrophic nonunion. In a final salvage attempt, nonunion resection and proximal tibia replacement (PTR) with megaprosthesis were performed. PTR is typically reserved for oncologic nonunion cases [15,16], and this is the first known report of successful PTR after persistent atrophic nonunion.

The patient was informed that this case would be submitted for publication and agreed to the submission of associated data.

Case history

A 56-year-old woman with a history of chronic low back pain, osteopenia, malnutrition (body mass index, 17), smoking, marijuana use, and alcohol use was treated for a left proximal tibia fracture from a fall in November 2018: multiplanar external fixation was initially performed due to swelling followed by internal fixation 3 days later.

Five weeks later she reported to the emergency department with pain and erythema at the incision site; superficial cellulitis with no drainage or dehiscence was noted. Laboratory test results indicated leukocytosis (15×10^9 cell/L) with an erythrocyte sedimentation rate of 37 mm/h. Imaging showed little evidence of bone healing. The patient was discharged after a course of intravenous antibiotics.

Five months after the initial injury, in April 2019, the patient reported persistent pain and difficulty ambulating. A CT scan showed atrophic nonunion of the tibia; a revision open reduction internal fixation was performed with debridement of the nonunion and utilization of proximal femur autograft via a reamer irrigator aspirator. The patient was additionally treated with extracorporeal shockwave therapy and vitamin D supplementation. Intraoperative cultures at this point resulted as negative.

The leg pain persisted at 4 months; a CT scan showed nonunion of the tibia as well as fractures of several screws. A second revision open reduction internal fixation was performed with debridement and grafting with a mixture of iliac crest, ViviGen, and bone morphogenetic protein 2 (BMP-2). Cultures from intraoperative samples tested positive for *Klebsiella pneumoniae*, indicating chronic osteomyelitis. The patient was given ceftriaxone throughout her stay and discharged on a course of ciprofloxacin. She continued extracorporeal shockwave therapy and vitamin D supplementation and reported substantially decreasing her smoking.

At the 4-month follow-up, the patient reported pain and difficulty ambulating. Imaging revealed persistent nonunion of the tibia



Fig. 1. Persistent nonunion after several attempts at revision open reduction internal fixation with bone grafting.

fracture as well as failures of several screws and the lateral locking plate from the most recent revision (Fig. 1). After discussing alternative solutions, the hardware was removed, exposing massive bony necrosis. The resection of the nonunion left a 6-cm defect, preserving the tibial tuberosity and infrapatellar tendon attachment. The decision was made to proceed with bone transport using a multiplanar unilateral external fixator and a percutaneous, low-energy osteotomy at the distal tibial metaphysis (Fig. 2). The docking site was debrided and an iliac crest bone graft was placed for distal tibia regeneration. The external fixator was converted to a hexapod frame after 2 months because of a pin site infection with loosening around the distal femoral ring.

A scheduled docking procedure was performed 7 months later (July 2020), with open debridement, grafting of cancellous iliac crest bone to the docking site, and proximal frame screw compression. Six weeks later, an antegrade intramedullary rod was placed and the fixator was removed. Two months later, the patient presented with pain after twisting her left knee while walking. Imaging demonstrated an acute fracture of the left medial femoral condyle; however, the hardware was intact and the injury was treated nonoperatively.

The patient reported continued pain and deformity at the site of nonunion in December of 2020 (2 years after the index procedure), impacting her quality of life (Fig. 3). At this time, below-the-knee amputation and knee arthrodesis were presented as options, but the patient elected to continue with attempts to salvage the limb. Thus, the hardware was removed and we performed a proximal tibia nonunion resection with total knee PTR arthroplasty. The Ilizarov regenerate of the distal tibia was still flexible, protected by a 90–90 double plating construct. The tibial tubercle osteotomy was reduced using cerclage wires (Fig. 4). A medial gastrocnemius pedicle rotational flap was used to cover the anterior medial portion of the endoprosthesis, and the posterior leg skin defect was covered with a synthetic skin graft. Intraoperative vascular consult was needed to assist with hemostasis and dissection of the posterior vasculature structures because of the distortion of the anatomy from multiple surgeries. The anterior tibial artery was protected for possible devascularization of the lower leg given her multiple operations.

The postoperative plan included strict immobilization in extension. One month after the PTR, the patient returned for a splitthickness skin graft and vacuum-assisted wound closure. Two months after the PTR, the cerclage blocking wires were removed from the healed tibial tubercle; passive range of motion at the knee was 0° -30°.

Two months later, the patient presented to the emergency department reporting increased pain in the lower left extremity. Imaging revealed that several screws around the distal tibial osteotomy had failed. Nonunion was determined on the basis of the patient's history of nonunion and hardware failure. After a 3 month delay for extensive patient counselling and surgical planning, she underwent hardware removal of the failed distal plates, left fibular head resection for autograft of the distal third tibial nonunion, and compression plating with fixation through the tibial crest. Segmental osteotomy of the fibula at the level of the nonunion was performed to allow tibial compression (Fig. 5). Cultures from intraoperative samples grew *P. acnes* in broth only, which was felt to be likely a contaminant per the infectious disease team comanaging her care, and the patient was treated with amoxicillin. She developed a transient foot drop, which resolved.

At nearly 2 years after the megaprosthesis reconstruction, radiographs demonstrated bony healing. The patient reported discomfort at the medial aspect of her ankle related to hardware irritation and underwent hardware removal (Fig. 6).

At final follow-up 5.5 years after her initial injury, and 3.5 years after proximal tibial replacement, the patient maintained neutral alignment of her leg, with a 0° -120° range of motion and strength of 5/5; the neurovasculature in her left foot was intact (Fig. 7). She wore corrective shoes to account for a 2-cm difference in leg length; overall, her pain and disability significantly improved, reporting a Lower Extremity Functional Scale score of 46.3 %. [17]



Fig. 2. Bone transport using a multiplanar unilateral external fixator was performed, utilizing a percutaneous, low-energy osteotomy at the distal tibial metaphysis.



Fig. 3. Persistent nonunion after intramedullary nailing with hypoplastic regenerate.



Fig. 4. Proximal tibial replacement arthroplasty was performed, with 90–90 double plating at the Ilizarov regenerate of the distal tibia, as it was still flexible, and reduction of the tibial tubercle osteotomy using cerclage wires.





Discussion

The patient in this case was at risk for fracture nonunion based on multiple factors including her age, smoking, and poor nutrition [9,10] and had undergone multiple surgeries that failed to heal the fracture or relieve her pain and disability. Quality of life with tibial nonunion is reportedly poorer than after myocardial infarction or stroke, [18] with significant risk for increased opioid use. [19]

Although nonsurgical treatments such as extracorporeal shockwave therapy can be successful [20], tibial nonunion is typically treated with revision open reduction internal fixation with bone grafting [21] or external fixation [22]. However, atrophic nonunion is notoriously difficult to treat, possibly because of a lack of vascular support [23] and stem cell activation [24]. External fixation is used to restore function with minimal deformity [25] and is typically successful in patients with infected tibial nonunion. [22] Deformities in patients with proximal tibial nonunion can be corrected with total knee arthroplasty [26], whereas PTR is typically performed in oncology patients [27]. The patient in this case had indications of chronic osteomyelitis, and the treatment failures prompted discussions of all options. In an effort to avoid amputation, the decision was made to perform PTR.

PTR is a surgically challenging technique, with considerations to be made firstly towards surgical dissection and exposure. Even in the setting of PTR for limb salvage outside of an oncologic indication, the complexity of the vascular anatomy in the popliteal fossa must be fully appreciated and protected. [28] Typically, the anterior tibial vessels need to be ligated as they pass from the posterior compartment anteriorly; however, in the setting of our patient with a multiply operated limb, special consideration was made towards protecting these vessels to ensure the limb remained perfused. There is risk of infection with PTR, [29] but this has been reduced with new soft tissue coverage techniques such as the medial gastrocnemius rotation flap, which is frequently electively performed, preferably dissected after proximal tibial resection but before final prosthesis implantation. [30,31]

Attention must also be paid towards maintenance of the extensor mechanism, which is mandatory to allow for active knee extension and full functional recovery. The patellar tendon is typically resected proximal to its attachment on the tibia during resection, and anchored into hooks or holes on the anterior surface of the implant with heavy suture, into which a bone plug is wedged, with the goal of eventual permanent biologic attachment. [28] In our case, the patellar tendon was able to be resected with the attached tibial tubercle, and cerclage wires utilized to provide stability. After these wires were removed, flexion of the patient's knee



Fig. 6. Removal of the symptomatic medial plate after complete bone healing.

improved to 120°. While it is common for patients to require assistive devices after PTR [15], our patient has demonstrated excellent functional outcome with independent ambulation.

PTR can be complicated by aseptic loosening and extensor mechanism dysfunction [15]⁷ [27,32], which interferes with functional recovery, but has been described as a salvage method in patients with substantial bone loss [16]. Specific to our patient, the modular stem of the PTR located just proximal to the site of the nonunion in our patient likely contributed to nonunion at the distal tibial osteotomy, which was later addressed with autograft and compression plating, going onto uneventful healing.

PTR has been demonstrated to be a viable option for limb salvage in the setting of consideration otherwise for amputation or knee fusion; however, the longevity of PTR implant does limit its long-term utility. Five-year survivorship of the PTR implant in oncologic literature has been estimated between 69 and 72 %, and 43 % at 10 years. [33,34] A cohort described by Meyer et al. reported even lower rates of revision in their comparison of fixed vs. rotating hinge implants, estimating 12–32 % at 5 years, 25–61 % at 10 years, and 30–75 % at 15 years favoring a cemented, rotating hinge design after tumor resection. However, it is hypothesized that without concern for oncologic recurrence and radiation-induced complications, the survivorship of PTR prostheses for non-oncologic indications may prove to be superior to the survivorship of the implant reported as related to oncologic literature. [15]

Summary

We suggest that PTR with megaprosthesis can be successful for managing massive bone loss at the proximal tibia. This option offers the possibility of saving the affected limb, restoring much of its function, and improving the patient's quality of life, but patients should be counseled on associated risks and expectations after surgery, including the expected possibility of future revision given the survival rate of the implant.



Fig. 7. Clinical pictures at last follow up demonstrating excellent range of motion, and resolution of foot drop.

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CRediT authorship contribution statement

Ellen Lutnick: Writing – review & editing, Writing – original draft, Supervision, Methodology, Data curation, Conceptualization. **Noah M. Braun:** Writing – original draft, Investigation, Data curation. **Evgeny Dyskin:** Writing – review & editing, Supervision, Conceptualization. **Mary Bayers-Thering:** Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

- [1] R. Mundi, D. Axelrod, D. Heels-Ansdell, H. Chaudhry, O.R. Ayeni, B. Petrisor, et al., Nonunion in patients with Tibial shaft fractures: is early physical status associated with fracture healing? Cureus 12 (4) (2020) e7649.
- [2] J.A. Bishop, A.A. Palanca, M.J. Bellino, D.W. Lowenberg, Assessment of compromised fracture healing, J. Am. Acad. Orthop. Surg. 20 (5) (2012) 273–282.
 [3] D.P. Taormina, B.S. Shulman, R. Karia, A.B. Spitzer, S.R. Konda, K.A. Egol, Older age does not affect healing time and functional outcomes after fracture
- [5] D.F. Faorinina, B.S. Shuman, K. Kara, A.B. Spitzer, S.K. Konda, K.A. Egol, Older age does not affect freaming time and functional outcomes after fracture nonunion surgery, Geriatric Orthopaedic Surgery & Rehabilitation. 5 (3) (2014) 116–121.
 [4] M.B. Picklez, D.R. Oldersen, The bicker base for accurate for accur
- [4] M.R. Brinker, D.P. O'Connor, The biological basis for nonunions, JBJS Rev. 4 (6) (2016).
- [5] R. Zura, J.T. Watson, T. Einhorn, S. Mehta, G.J. Della Rocca, Z. Xiong, et al., An inception cohort analysis to predict nonunion in tibia and 17 other fracture locations, Injury 48 (6) (2017) 1194–1203.
- [6] L. Mills, J. Tsang, G. Hopper, G. Keenan, A.H. Simpson, The multifactorial aetiology of fracture nonunion and the importance of searching for latent infection, Bone Joint Res. 5 (10) (2016) 512–519.
- [7] A. Nauth, M. Lee, M.J. Gardner, M.R. Brinker, S.J. Warner, P. Tornetta 3rd, P. Leucht, Principles of nonunion management: state of the art, J. Orthop. Trauma 32 (Suppl. 1) (2018) 852–87.
- [8] R. Tian, F. Zheng, W. Zhao, Y. Zhao, J. Yuan, B. Zhang, L. Li, Prevalence and influencing factors of nonunion in patients with tibial fracture: systematic review and meta-analysis, J. Orthop. Surg. Res. 15 (1) (2020) 377.
- [9] R. Zura, Z. Xiong, T. Einhorn, J.T. Watson, R.F. Ostrum, M.J. Prayson, et al., Epidemiology of fracture nonunion in 18 human bones, JAMA Surg. 151 (11) (2016) e162775.
- [10] J.A. Nicholson, N. Makaram, A. Simpson, J.F. Keating, Fracture nonunion in long bones: a literature review of risk factors and surgical management, Injury 52 (Suppl. 2) (2021) S3–S11.
- [11] M.M. Chaudhary, Infected nonunion of tibia, Indian J. Orthop. 51 (3) (2017) 256-268.
- [12] J. Wilson, S. Boden, K. Cintron, M. Schenker, Optimizing Metabolism to Treat Fractures and Prevent Nonunion, 1 ed, CRC Press, 2019, pp. 389-402.

- [13] S.N. Pierrie, J.R. Hsu, Shortening and angulation strategies to address composite bone and soft tissue defects, J. Orthop. Trauma 31 (Suppl. 5) (2017) S32–S35.
- [14] I.S. Tarkin, P.A. Siska, B.A. Zelle, Soft tissue and biomechanical challenges encountered with the management of distal tibia nonunions, Orthop. Clin. North Am. 41 (1) (2010) 119–126 (table of contents).
- [15] B. Fram, E.B. Smith, G.K. Deirmengian, J.A. Abraham, J. Strony, M.B. Cross, D.Y. Ponzio, Proximal tibial replacement in revision knee arthroplasty for nononcologic indications, Arthroplast Today. 6 (1) (2020) 23–35.
- [16] S. Höll, A. Schlomberg, G. Gosheger, R. Dieckmann, A. Streitbuerger, D. Schulz, J. Hardes, Distal femur and proximal tibia replacement with megaprosthesis in revision knee arthroplasty: a limb-saving procedure, Knee Surg. Sports Traumatol. Arthrosc. 20 (12) (2012) 2513–2518.
- [17] J.M. Binkley, P.W. Stratford, S.A. Lott, D.L. Riddle, The lower extremity functional scale (LEFS): scale development, measurement properties, and clinical application. North American Orthopaedic rehabilitation research network, Phys. Ther. 79 (4) (1999) 371–383.
- [18] M.R. Brinker, B.D. Hanus, M. Sen, D.P. O'Connor, The devastating effects of tibial nonunion on health-related quality of life, J. Bone Joint Surg. Am. 95 (24) (2013) 2170–2176.
- [19] E. Antonova, T.K. Le, R. Burge, J. Mershon, Tibia shaft fractures: costly burden of nonunions, BMC Musculoskelet. Disord. 14 (2013) 42.
- [20] V. Sansone, D. Ravier, V. Pascale, R. Applefield, M. Del Fabbro, N. Martinelli, Extracorporeal shockwave therapy in the treatment of nonunion in long bones: a systematic review and Meta-analysis, J. Clin. Med. 11 (7) (2022).
- [21] E. Gálvez-Sirvent, A. Ibarzábal-Gil, E.C. Rodríguez-Merchán, Treatment options for aseptic tibial diaphyseal nonunion: a review of selected studies, EFORT Open Rev. 5 (11) (2020) 835–844.
- [22] P. Yin, Q. Zhang, Z. Mao, T. Li, L. Zhang, P. Tang, The treatment of infected tibial nonunion by bone transport using the Ilizarov external fixator and a systematic review of infected tibial nonunion treated by Ilizarov methods, Acta Orthop. Belg. 80 (3) (2014) 426–435.
- [23] A.A. Reed, C.J. Jovner, H.C. Brownlow, A.H. Simpson, Human atrophic fracture non-unions are not avascular, J. Orthop. Res. 20 (3) (2002) 593-599.
- [24] T. Tawonsawatruk, M. Kelly, H. Simpson, Evaluation of native mesenchymal stem cells from bone marrow and local tissue in an atrophic nonunion model, Tissue Eng. Part C Methods 20 (6) (2014) 524–532.
- [25] D.A. Wiss, D.L. Johnson, M. Miao, Compression plating for non-union after failed external fixation of open tibial fractures, J. Bone Joint Surg. Am. 74 (9) (1992) 1279–1285.
- [26] A. Kappel, P.T. Nielsen, S. Kold, 1-stage total knee arthroplasty and proximal tibial non-union correction using 3-D planning and custom-made cutting guide, Acta Orthop. 92 (4) (2021) 452–454.
- [27] M.V. Natarajan, A. Sivaseelam, G. Rajkumar, S.H. Hussain, Custom megaprosthetic replacement for proximal tibial tumours, Int. Orthop. 27 (6) (2003) 334–337.
- [28] A. Puri, Tips and tricks of limb salvage: proximal tibia, Indian J Orthop. 48 (3) (2014) 296-300.
- [29] P. Tsagozis, M. Parry, R. Grimer, High complication rate after extendible endoprosthetic replacement of the proximal tibia: a retrospective study of 42 consecutive children, Acta Orthop. 89 (6) (2018) 678–682.
- [30] A.F. Mavrogenis, E. Pala, A. Angelini, A. Ferraro, P. Ruggieri, Proximal tibial resections and reconstructions: clinical outcome of 225 patients, J. Surg. Oncol. 107 (4) (2013) 335–342.
- [31] G.J. Myers, A.T. Abudu, S.R. Carter, R.M. Tillman, R.J. Grimer, The long-term results of endoprosthetic replacement of the proximal tibia for bone tumours, J. Bone Joint Surg. Br. 89 (12) (2007) 1632–1637.
- [32] H. Pilge, B.M. Holzapfel, H. Rechl, P.M. Prodinger, R. Lampe, U. Saur, et al., Function of the extensor mechanism of the knee after using the 'patellar-loop technique' to reconstruct the patellar tendon when replacing the proximal tibia for tumour, Bone Joint J. 97-B(8):1063-9 (2015).
- [33] D. Biau, F. Faure, S. Katsahian, C. Jeanrot, B. Tomeno, P. Anract, Survival of total knee replacement with a megaprosthesis after bone tumor resection, J. Bone Joint Surg. Am. 88 (6) (2006) 1285–1293.
- [34] J.S. Wunder, K. Leitch, A.M. Griffin, A.M. Davis, R.S. Bell, Comparison of two methods of reconstruction for primary malignant tumors at the knee: a sequential cohort study, J. Surg. Oncol. 77 (2) (2001) 89–99 (discussion 100).