Clinical Case Report

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Periprosthetic femoral fractures around tumor endoprostheses treated with limited revision surgery combined with allograft

A case report

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Abstract

Rationale: Old periprosthetic femoral fractures (OPFFs) around a tumor prosthesis of the knee present formidable problems for orthopedic oncologists; large bone defects and inappropriate biomechanics of the revision implant design can impair successful reconstruction. Limited literature is available on the reconstruction of OPFFs using revision combined with massive allograft following resection of a bone tumor around the knee joint. In this study, we present the first reported case in the English literature of a limited revision followed by several segmental allografts for the reconstruction of the knee joint.

Patient concerns: This case involved a 45-year-old female who was treated for a malignant fibrous histiocytoma (MFH) of the knee joint with surgical excision of the lesion and replacement of the defect using endoprosthetic reconstruction when she was 25 years old. Her surgical history was remarkable for a left tumoral knee prosthesis implanted 20 years ago. Nine years before revision, the patient had fall damage; however, she was able to walk independently and with moderate pain. In the 9-year period, prosthesis malfunction caused progressive left lower extremity shortening and a persistent swelling pain in the left thigh.

Diagnoses: According to her clinical history, imaging results and physical examination, we confirmed the diagnosis of OPFFs potentially due to aseptic loosening and trauma injury.

Interventions: In this study, we present the first case of OPFFs around a tumor endoprosthesis that was successfully treated using limited revision combined with a massive allograft.

Outcomes: At 80 months after revision surgery, the patient had made a sufficient recovery from her symptoms. The bone union was complete without tumor recurrence or implant failure.

Lessons: We propose that if prosthesis fracture is detected, revision surgery should be attempted as early as possible, and for patients with OPFFs, the use of limited revision combined with massive allografts may be useful for safely and adequately reconstructing OPFFs around the knee joint. However, patients should be followed-up closely after surgical treatment because of the high risk of revision.

Abbreviations: CT = computed tomography, EPR = endoprosthetic reconstruction, MFH = malignant fibrous histiocytoma, OPFFs = old periprosthetic femoral fractures, ORIF = open reduction internal fixation, PFF = periprosthetic femoral fracture, ROM = range of motion, UCS = unified classification system, VAS = visual analog scale.

Keywords: complications, knee joint, old periprosthetic femoral fractures (OPFFs), revision surgery

1. Introduction

Primary malignant bone tumors account for less than 1% of all cancers, and until the 1970s, the treatment of high-grade extremity sarcoma routinely consisted of amputation of the affected limbs.[1–3] Currently, with the advent of effective chemotherapy, better imaging tools, and improved understanding of tumor behavior, 70% to 85% of all malignant bone tumors are treated by limb salvage with promising oncological and functional results.[3] The knee joint is the most common site for primary malignant bone tumors, and various techniques, such as implantation of osteoarticular allografts, allograft-prosthetic composites, vascularized bone grafts, and custom-made endoprostheses, have been employed for the reconstruction of knee joints affected by malignant bone tumors. With the development of new operative techniques, innovations in materials and improved prosthetic design, it is likely that endoprosthetic reconstruction (EPR) has become the primary treatment and is the most commonly used approach following the excision of knee joint tumors or surgery for other nontumorous conditions.[4–6] With improvements in the cooperating disciplines and molecular targeted antitumor drugs, the long-term survival of patients with primary malignant bone neoplasms has improved. Recently, Mittermayer et al[1] reported that the survival rate and functional capability of patients treated with tumor prostheses were satisfactory after the resection of a malignant tumor around

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the knee joint. Even though the potential advantages of EPR in terms of mobilization with full weight bearing, great concern remains over the long-term reliability of these reconstructions, most notably concerns of infection, breakage, aseptic loosening and periprosthetic femoral fractures (PFF). Additionally, PFF, occurring in 10% to 24% of patients, are the main cause of failure in the later years after surgery. Because most patients with primary bone tumors are young and are expected to lead active lives, revisions of EPR occur frequently and inevitably.[7–10] According to published literature, periprosthetic fractures around standard knee replacements have been widely studied, and their management is now fairly routine. In contrast, the treatment of PFF around tumor endoprostheses has been poorly studied.[10–12]

It is noteworthy that the management of PFF around removed tumor endoprostheses may be different from that of conventional joint replacements and represent a challenge for the orthopedic surgeon owing to patient characteristics and implant differences.[7,13] Tumor endoprostheses are massive, often with limited bone available for fixation, and patients with old periprosthetic femoral fractures (OPFFs) usually exhibit large-scale bone defects, difficult-to-identify anatomic structures due to soft-tissue pigmentation, and limb shortening with accompanied peripheral soft tissue damage.[8,12] Furthermore, these patients may be reluctant to undergo revision surgery associated with a high rate of risk and complications. To the best of our knowledge, there is very limited evidence regarding the treatment of OPFFs around tumor endoprostheses.

In this study, we present the first case involving an OPFF around a tumor endoprosthesis that was successfully treated using limited revision combined with a massive allograft, and we also review relevant papers and discussions regarding this form of endoprosthetic failure regarding clinical features and treatment. The patient gave her informed consent for the submission of this case study for publication.

2. Case presentation

2.1. Ethical statement

All procedures performed in studies involving human participants were in accordance with the ethical standards of the ethical committee in our hospital and with the *Declaration of Helsinki* its later amendments or comparable ethical standards. The patient gave informed consent for the submission of this case study for publication. This study was performed in accordance with relevant guidelines and regulations.

2.2. Clinical data

In July 1991, a 25-year-old female patient was diagnosed with "left distal femoral malignant fibrous histiocytoma (MFH)" because of pain in the left thigh 5 months ago. She underwent tumor resection followed by EPR of the knee joint. The preoperative and postoperative pathological examination confirmed left femoral malignant fibrous histiocytoma (MFH). After surgery, the patient also underwent adjuvant therapy and recovered well. At the last follow-up, there was no local recurrence of the oncologic disease, and the prosthesis showed no signs of mobilization (Fig. 1A). In August 2002, the patient reported fall damage with direct trauma to the left lower limb. At that time, a mass appeared on the front of the left thigh. However, she was able to walk independently and with moderate pain, which was aggravated while moving and eased after rest. Over the next 9 years, the patient experienced progressive left lower extremity shortening and persistent swelling with pain in the left thigh. In June 2011, the patient was referred to our institution, with increasing pain in her left knee and with lower limb length discrepancy and extorsion deformity (Fig. 1B-C). The physical examination revealed that the left lower limb was 10 cm shorter than the length of the right lower limb, and there was more swelling in the left thigh than in the right thigh. A postoperative scar of 20 cm in length was seen in front of the left knee joint (Fig. 1C). There was no obvious skin damage or subdermal ecchymosis in the incision. The tenderness was obvious in the middle segment of the left thigh, and an irregular, hard mass could be palpated. The range of motion (ROM) was limited. The patient was not cooperative with physical examination due to pain in the left knee. The flexion and extension were significantly restricted, but the ROM was in the normal range in the hip joints. Subsequently, the ROM of the right hip and knee joints was normal. Muscle strength and tension of the right lower limbs were also normal, and the bilateral dorsal pedis arterial pulses were good. The dorsiflexion of the ankle and each toe was good. No other positive findings were found on physical examination. Neurologic examination revealed no specific findings, including that her bilateral lower limb reflexes of the knee and ankle were normal. The pathological reflex was not found. Laboratory investigations revealed routine blood parameters and erythrocyte sedimentation rates all within the normal ranges and no signs of infection. The patient had long-term laminated object manufacturing. Arterial-venous color Doppler ultrasound for bilateral limbs showed that no significant abnormality was found in the arteries or veins of either limb. Her preoperative visual

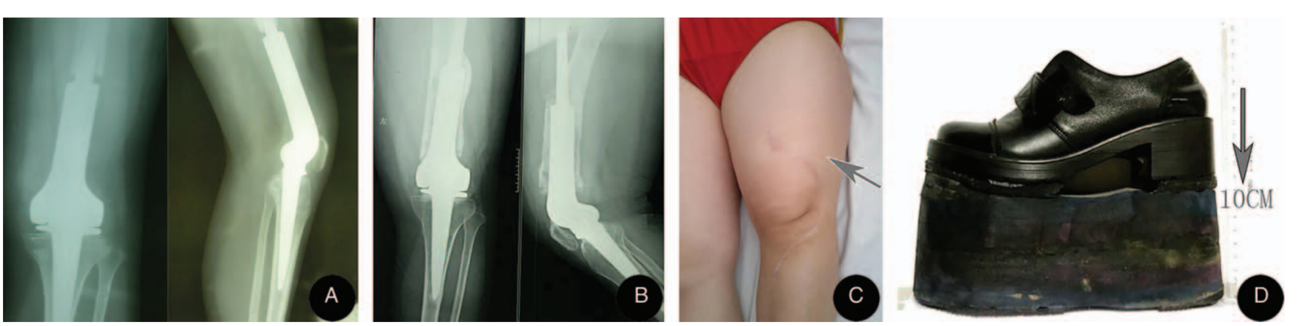

Figure 1. Radiograph (A) at the 2-year follow-up (in 1993) showing a replacement of the custom-made endoprosthesis of the knee joint with no sign of tumor recurrence or periprosthetic fracture. An anteroposterior standing radiograph (B) of the lower left limb showing fracturing of the proximal femoral stem, and the prosthesis stem on the femur side had passed through the femur toward the front. Preoperative photo (C) showing swelling and deformity of the left thigh (gray arrow). The patient's specially designed shoes (D), approximately 10 cm tall.

analog scale (VAS) score was 8. She had no other, related past medical history.

2.3. Imaging examinations

During hospitalization, the patient underwent thorough radiographic examinations. Plain X-rays of the left femur demonstrated that the prosthesis stem on the femur side passed through the femur toward the front, and the prosthesis had broken through the proximal cortex (Fig. 1B). Reconstructed computed tomography (CT) of the left femur showed that the prosthesis had pierced the femur because of loosening and the left lower limb deformity, and the worn-out prosthesis was surrounded by soft tissue with no signs of tumor recurrence (Fig. 2A–C).

2.4. Operative procedures

According to the general symptoms, clinical history, all imaging examinations including plain radiographs and CT, and laboratory findings of this patient in our department, the definitive diagnosis of OPFF around tumor endoprostheses was made.

Considering the risk of revision surgery and the patient refusing revision surgery for personal reasons, we performed limit revision combined with massive allograft around the tumor endoprosthesis in a one-stage operation through an anterior approach. Via an anterior incision, 25 cm of the knee joint (Fig. 2D), including the initial scar, was excised along with the biological pseudomembrane around the surface of the endoprosthesis. Samples were taken for bacterial culture and pathological analysis (Fig. 3A). During the surgical procedure, the lateral femoral muscle fibers were bluntly dissected and pulled open toward the front and back to expose the femur and prosthesis. There was no osteolysis, and both prosthetic stems remained well integrated with the bone (Fig. 2E). Then, the scars and proliferous bony fibrous tissue were cleaned up. We transected the femur and extracted the loosened prosthesis through proximal fenestration and distal reaming (Fig. 2F-G). After disinfection of the original prosthesis, we filled the femoral marrow cavity with bone cement followed by implantation of the distal prosthesis (Fig. 2H-I). To maintain the stability of the prosthesis, 2 large allogeneic cortical bone plates were fastened to the bone defect of the anterior femur by 3 wires (Fig. 2J). Subsequently, a swing saw was used to polish the proximal end of the tibia, and the medullary cavity of the tibia was filled with bone cement followed by implantation of the tibial prosthesis (Fig. 2K-L). Eventually, the wound was washed and sutured after cement solidification.

2.5. Pathological examination and postoperative course

Pathologic examination (Fig. 3B-C) of the excised tissue showed that massive acute and chronic inflammatory cell infiltration was observed in the peripheral hyperplastic fibrous tissue of the

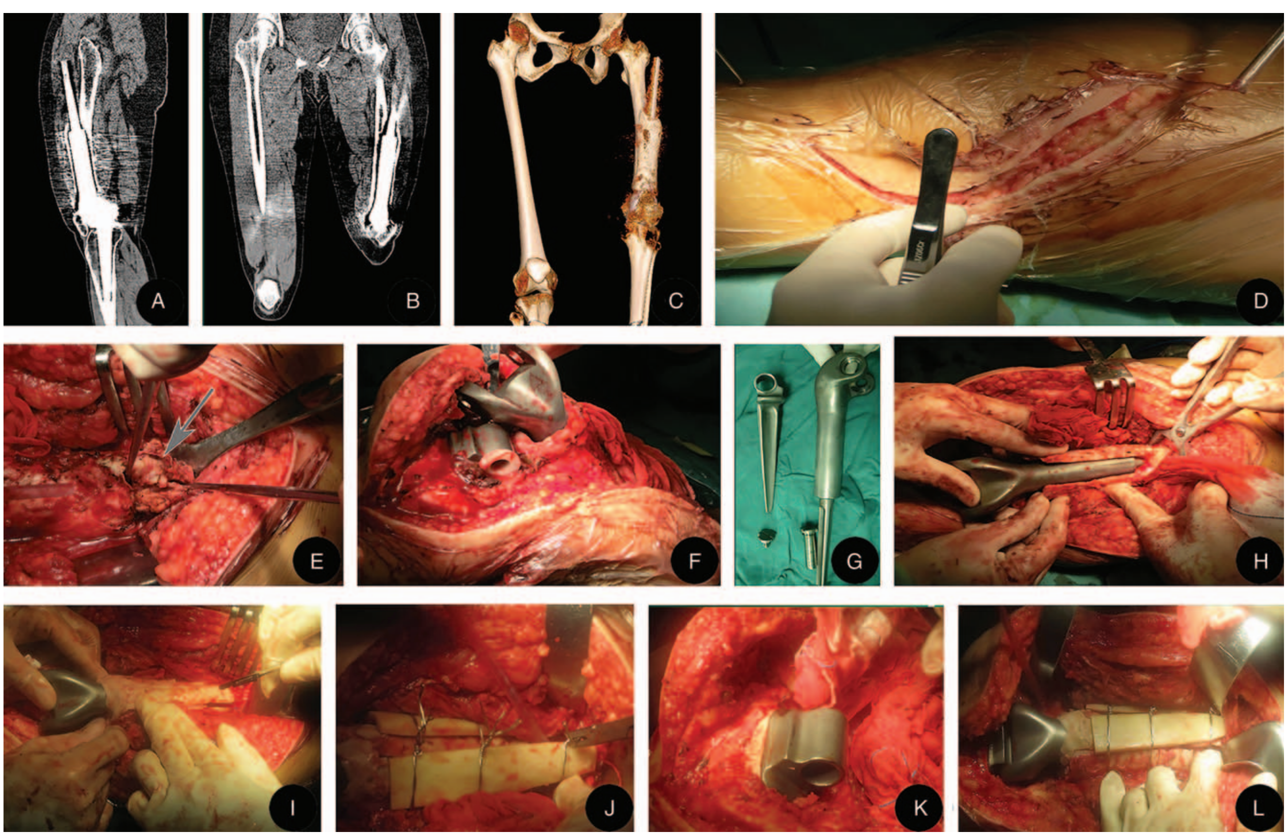

Figure 2. Computed tomography (CT) images of the left femur. Sagittal (A) and coronal (B) images as well as a reconstructed CT image (C) showing that the proximal part of the prosthesis penetrated the upper femur surrounded by a callus. There was no evidence of tumor recurrence. An intraoperative photo showing the incision was taken at the lateral femur starting from the small tuberosities and down to the lateral knee joint (D). The scars and proliferous bony fibrous tissue adjacent to the prosthesis were cleaned up (E). Then, we extracted the loosened prosthesis through proximal fenestration and distal reaming (F, G). Subsequently, the femoral marrow cavity was filled with bone cement and surface of the prosthesis was covered, followed by implantation of the distal prosthesis (H, I). Two allogeneic cortical bone plates were fastened to the bone defect of the anterior femur by 3 wires (J). Eventually, the prosthesis was successfully connected (K, L).

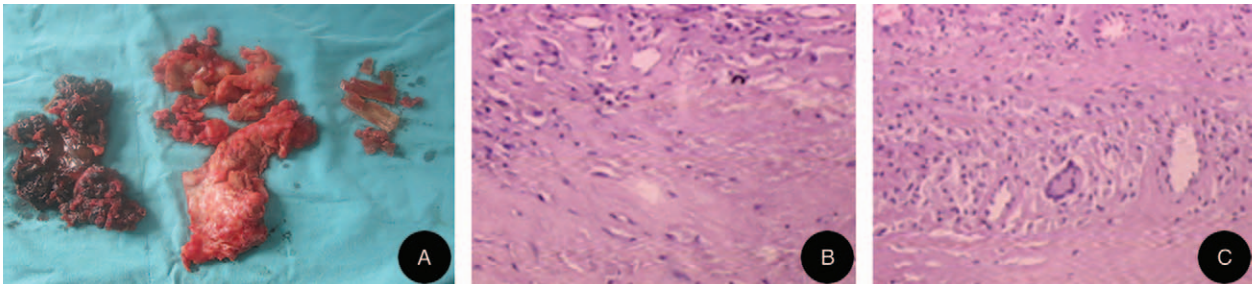

Figure 3. Specimens removed from around the prosthesis (A). Histopathology photomicrographs of the excised specimen. (B) Low-power view shows massive acute and chronic inflammatory cell infiltration (H&E, original magnification ×40). (C) High-power view (H&E, original magnification ×100) shows abundant hyperplastic fiber tissues with some dead bone tissue and calcification.

prosthesis. In addition, there were large areas of bland necrosis surrounded by reactive fibrosis. Microscopy findings and bacteria culture from the specimen were negative.

The drainage tube was removed on the sixth day after the operation when the drainage flow was less than 20 mL/24 h. The postoperative course went well. The patient tolerated partial weight bearing ambulation 2 to 3 weeks postoperation and started functional mobilization, including a considerable amount of strengthening work around the quadriceps femoris and tibialis anterior muscle under the guidance of professional rehabilitation orthopedic doctors until bony fusion was achieved at approximately 4 months postoperation. At 74 months after the surgery, the anteroposterior film of the X-ray revealed complete bone union with no implant failures (Fig. 4A-B). Follow-up imaging showed no implant failures at 74 months and good functional outcome (Fig. 4C-D). The VAS score improved to 1.

3. Discussion

For cancers that develop in the bone, such as high-grade extremity sarcoma, local resection or wide excision is associated with low rates of local recurrence compared to intralesional treatment, but the reconstruction of a large bone defect after resection presents a challenging problem to orthopedic surgeons.[14,15] At present, endoprosthetic replacement is widely accepted in the reconstruction of bone defects following resection of a tumor around the knee joint, providing the advantage of rapid recovery, active mobility, and early weight bearing in most cases. Unfortunately, the reported complication rate remains 5 to 10 times higher than rates seen in routine total joint arthroplasties, and subsequent surgical revisions resulting from increased prosthesis-associated complications, such as infection, periprosthetic infections, aseptic loosening, and breakage of the prosthesis, remain an unresolved problem.[9,16–18] Reportedly, mechanical complications accounting for approximately 50% of all failures (e.g., breakage/fracture of the implant, instability due to wear) and biological complications (e.g., infection, aseptic loosening, wound/soft tissue breakdown) are a primary concern following endoprosthetic reconstruction (EPR).[19,20] In 2015, Barut et al[20] reported that revision rates for all patients for any reason after the fracture were 27% and 55% at 5 and 10 years, respectively. In addition, a second revision surgery is usually associated with a comparatively low long-term survival rate; the

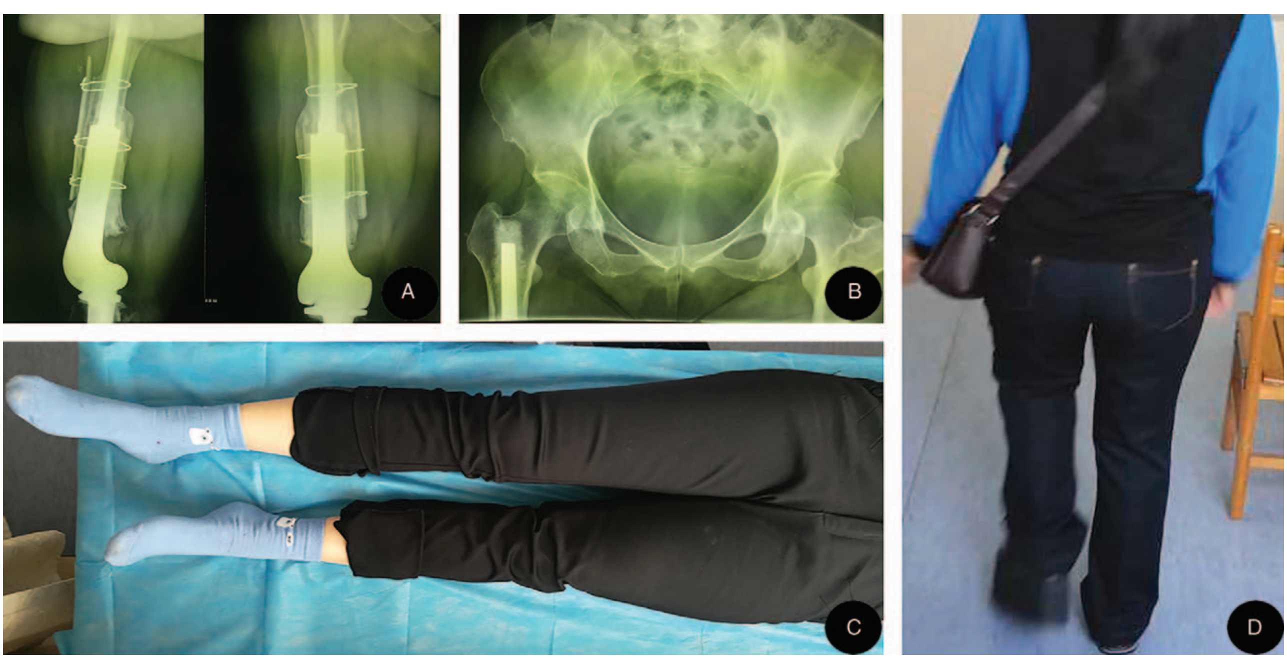

Figure 4. Postoperative X-ray images at the 74-month follow-up revealed that the bone healed well without obvious fracture dislocation or nonunion (A, B). At the last follow-up, we compared the length of both lower limbs of this patient and evaluated the independent capacity of walking (C, D).

5-year survival rate is 57% to 93%, whereas the 10-year survival rate is 60% to 88%.[14]

In our case, the patient's surgical history was remarkable for a left custom-made knee prosthesis implanted 20 years ago. According to her clinical history, imaging results and physical examination, we confirmed the diagnosis of an OPFF with the possible causes of aseptic loosening and trauma injury. Importantly, in this case, no signs of infection or tumor recurrence were found during the preoperative or intraoperative examinations. Approximately 10% to 46% of femur fractures have recurred at the femoral anterior arch due to the anatomy of the anterior arch of the femur, which is also consistent with our case.[9,12,21] For our patient, the possible causes of OPFF may also be mechanical causes, such as a mismatch of the femur arc. Due to the mismatch between the angle at the femoral junction of the tumor prosthesis and the angle of the femoral arch, the indirect stress was concentrated on a certain point of the bone; with the addition of a trauma injury, slow damage occurred in the femoral cortex. According to a multicenter study and comprehensive literature review, Henderson et al[17] demonstrated that there are 5 primary modes of endoprosthetic failure, and structural failure (type 3), such as periprosthetic or prosthetic fracture or a deficient osseous supporting structure, accounts for 17% of all failures, the most common being distal humeral and distal femoral replacements. Moreover, in 2014, Duncan et al[19] presented an overview of the Unified Classification System (UCS) and proposed a rational approach for the treatment of PFF. As OPFFs around tumor endoprostheses are different from those of standard implants, treatment management should focus on the quality and quantity of bone stock, as well as the quality and quantity of soft tissue. As a consequence, the UCS may not be appropriate for every patient. Furthermore, large-scale bone defects present difficulty in identifying anatomic structures due to soft-tissue pigmentation, limb shortening and accompanied peripheral soft tissue damage and also serve as obstacles for treatment. In regard to the review literature, little has been described regarding the management and the results of OPFFs around the knee joint.[14,16,22] Traditional treatment options are controversial, such as bisphosphonate, open or closed reduction and internal fixation, and revision surgery. In a large series of retrospective analyses, Lunebourg et al[23] indicated that PFF around standard implants treated by open reduction internal fixation (ORIF) had a revision rate of 17% at 5 years. In contrast, Macdonald et al,[7] in a series of 14 fractures treated by revision of the implant, showed a more favorable outcome with no revision at 8 years. Recently, Han et al[12] applied 3D printing technology to design prosthetic components and navigators for the treatment of PFF around the knee joint. According to the detailed and comprehensive information of this case, first, we suggested a complete revision surgery with a high rate of risk and complication, but the patient and her family refused for personal concern.

As a consequence, to relieve the pain and achieve a normal functional outcome, we met with senior surgeons to discuss experiences in tumor surgery and knee reconstruction. It is unfortunate that we could not correct the shortening deformity of this patient's leg; however, she still achieved a significant recovery from pain and immobility after treatment with a limit revision surgery combined with massive allografts. In addition, we performed bone cement filling and massive allografts to enhance joint stability and promote bone fusion. Recently, there have been a few papers reporting good results from cement filling of the gaps between the prosthetic stem and the endosteum to improve implant stability.[1,4,8,16,18] Menendez et al[8] and Morgan et al[4] found no aseptic loosening using the cementing technique in a large series of proximal femoral replacements. Despite the advantage of minimizing the risk of stem fracture, the bone stock of this patient is limited, and in order to accept an original-sized stem, massive allografts were used to preserve the bone stock. Van Isacker et al,[24] reported that allografts used for arthrodesis had a greater prevalence of fracture and nonunion, but in our case, these complications did not occur due to the preservation of the soft tissue around the prosthesis and avoidance of vascular injury.

4. Conclusion

To the best of our knowledge, this is the first reported case of an OPFF of the knee joint that was successfully treated by limited revision combined with massive allografts. For patients with OPFFs of tumor knee prostheses, revision surgery should be attempted as early as possible. However, patients should be followed-up closely after surgical treatment because of the high risk of revision.

Author contributions

Conceptualization: Jianhua Wu.
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Investigation: Yongguo Liu, Jun Lei.
Methodology: Jun Lei.
Project administration: Jianhua Wu.
Supervision: Jianjun Wang.
Writing – Original Draft: Jianhua Wu, Jun Lei.
Writing – Review & Editing: Jianhua Wu, Jun Lei.

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