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Case Report

Two Cases of Polyethylene Granuloma After Distal Femoral Endoprosthesis With All-Polyethylene Tibia

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ABSTRACT

Limited literature exists on complications specific to the all-polyethylene tibial component in distal femoral replacement (DFR). Unlike in primary arthroplasty with polyethylene components, polyethylene granuloma has not been reported in DFR with an all-polyethylene tibia. Here, we report 2 cases of polyethylene granuloma in patients with primary bone sarcoma who underwent DFR with an all-polyethylene tibia. Radiologically, evidence of intraosseous granuloma formation and periprosthetic osteolysis was observed at the anterior tibial metaphysis. Both patients underwent an operative debridement of polyethylene granuloma and necrotic tissues, followed by a revision to a long-stem, cemented metal-backed tibia with impacted allograft to fill the defect. Polyethylene granuloma should be considered a differential diagnosis in the presence of a periprosthetic lytic lesion after DFR with an all-polyethylene tibial component.
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Introduction

Distal femoral replacement (DFR) is one of the most common limb-sparing orthopedic oncology procedures, with its main indication in primary bone sarcoma [1]. With advances in surgical technique, prosthetic design, and systemic therapies, this limb-preserving procedure has shown to achieve an equal overall survival to amputation, while also providing improved function, cosmesis, and psychological benefits to patients [2]. Implants used in DFR include distal femoral components, a hinge system often with a rotating capacity, and tibial components. Tibial components are available in the form of all-polyethylene or metal-backed tibial baseplates.

The main complications of DFR include deep infection, aseptic loosening, hardware structural failure, and local recurrence of malignancy [1,2]. As a result, up to 33% of patients with DFR require a revision within 10 years [2-4]. However, in contrast to knee arthroplasty with polyethylene components, which may lead to the formation of polyethylene granuloma or pseudotumor [5-8], such complication of polyethylene granuloma after a DFR was not seemingly reported to date. The formation of polyethylene granuloma is driven by mechanical wear of polyethylene and periprosthetic accumulation of wear debris, which trigger macrophage-driven foreign body reaction [5-8]. This results in chronic lymphohistiocytic inflammation, which in turn leads to periprosthetic osteolysis [5,7,8]. Here, we report 2 cases of polyethylene granuloma after DFR with an all-polyethylene tibial component for primary bone sarcoma.

Case histories

Patient consents for the use of demographical and clinical information for publication were obtained.

Case 1

Patient A is a 52-year-old female who had distal femur osteosarcoma and underwent a wide resection and distal femur replacement using MRS implant (Stryker, Kalamazoo, MI) with a cemented all-polyethylene tibial implant. She had an uncomplicated recovery until 9 years postoperatively when a well-

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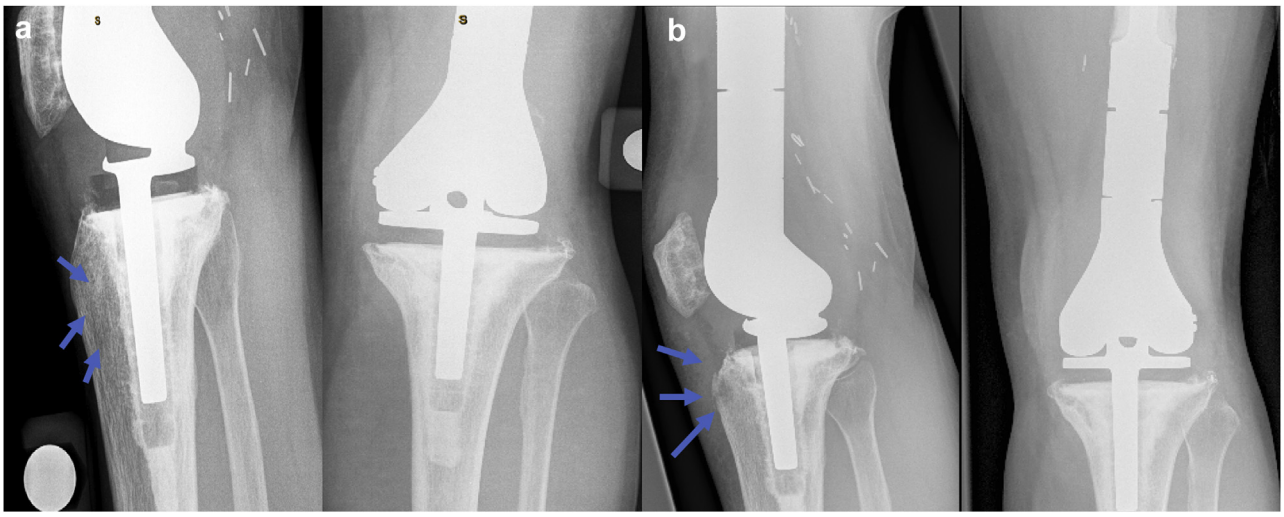

Figure 1. Plain radiographs showing polyethylene granuloma in patient A, left knee. (a) Initial presentation at 9 y postoperatively with asymptomatic benign appearing lesion posterior to the tibial tubercle (blue arrows). (b) Minimally displaced tibial tubercle fracture at 13 y postoperatively (blue arrows).

circumscribed, benign-appearing lytic lesion measuring 2 × 3 cm, just posterior to the tibial tubercle, was noticed on the follow-up radiographs (Fig. 1a). As there was a low clinical and radiological suspicion of malignant recurrence, infection, or loosening and the patient remained asymptomatic, the decision was made to closely follow this lesion. Three years after, the patient presented to our clinic after a fall that resulted in a nondisplaced tibial tubercle fracture but with an intact extensor mechanism. The fracture did not extend to the cement mantle, and the stem was not loose. She was managed with full weight-bearing and a long knee immobilizer for 6 weeks and recovered uneventfully. One year after, she developed another episode of anterior knee pain after forcefully bending her knee. An ultrasound imaging showed partial tear of the patellar tendon, as well as displaced bony flakes seen on radiographs. As the patient still had an intact extensor mechanism, the decision was made to treat conservatively. Five months later, the patient continued to have pain, and repeat radiographs showed no evidence of healing of the bony avulsion (Fig. 1b). Clinically, a tender soft-tissue mass was palpable anteriorly. Therefore, we proceeded to an open biopsy and curettage of the lesion. Tissue cultures were also taken. The tissue specimen showed reactive fibrosis, histiocytic infiltration, and giant cell reaction to foreign material. An examination of the tibial stem demonstrated a contact between the defect and the tibial stem with evidence of micromotion at the top level of the implant while the lower part of the stem remained well fixed. The bumper was found cracked and thus was removed, followed by removal of the bushings, which were completely worn out. After curettage, the cavity was allografted. The femoral component and bushings were also exchanged. Two months later, a computed tomography (CT) scan showed a progressive lytic lesion that extended to the bone cement interface posteriorly and breaching the cortex anteriorly (Fig. 2). The patient was indicated for a major revision, but as she was reluctant to have the surgery, the decision was made to continue close surveillance. A follow-up CT at 3 months showed a stable lesion, and the patient remained asymptomatic. At subsequent clinical and radiological follow-up, the patient remained asymptomatic, and the lesion did not progress until 6 years after her first revision, at which point she presented to the hospital with 3 recent episodes of acute pain in her proximal tibia each lasting a few days, accompanied by gait difficulties. Imaging demonstrated evidence of lesion progression and break of the cement mantle of the tibial component (Fig. 3a). After ruling out an infection, the patient

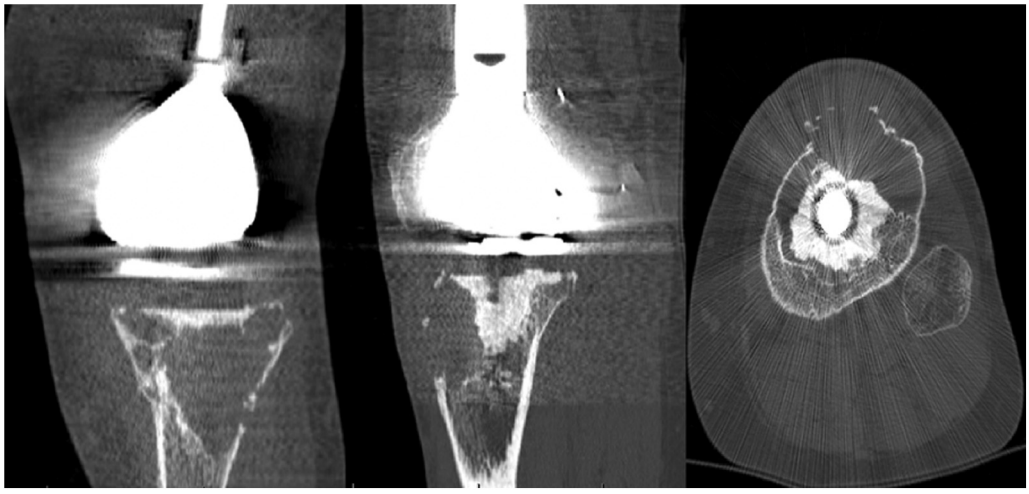

Figure 2. Patient A, left knee. CT images showing a periprosthetic osteolytic lesion breaching the anterior cortex and extending to the bone-cement interface.

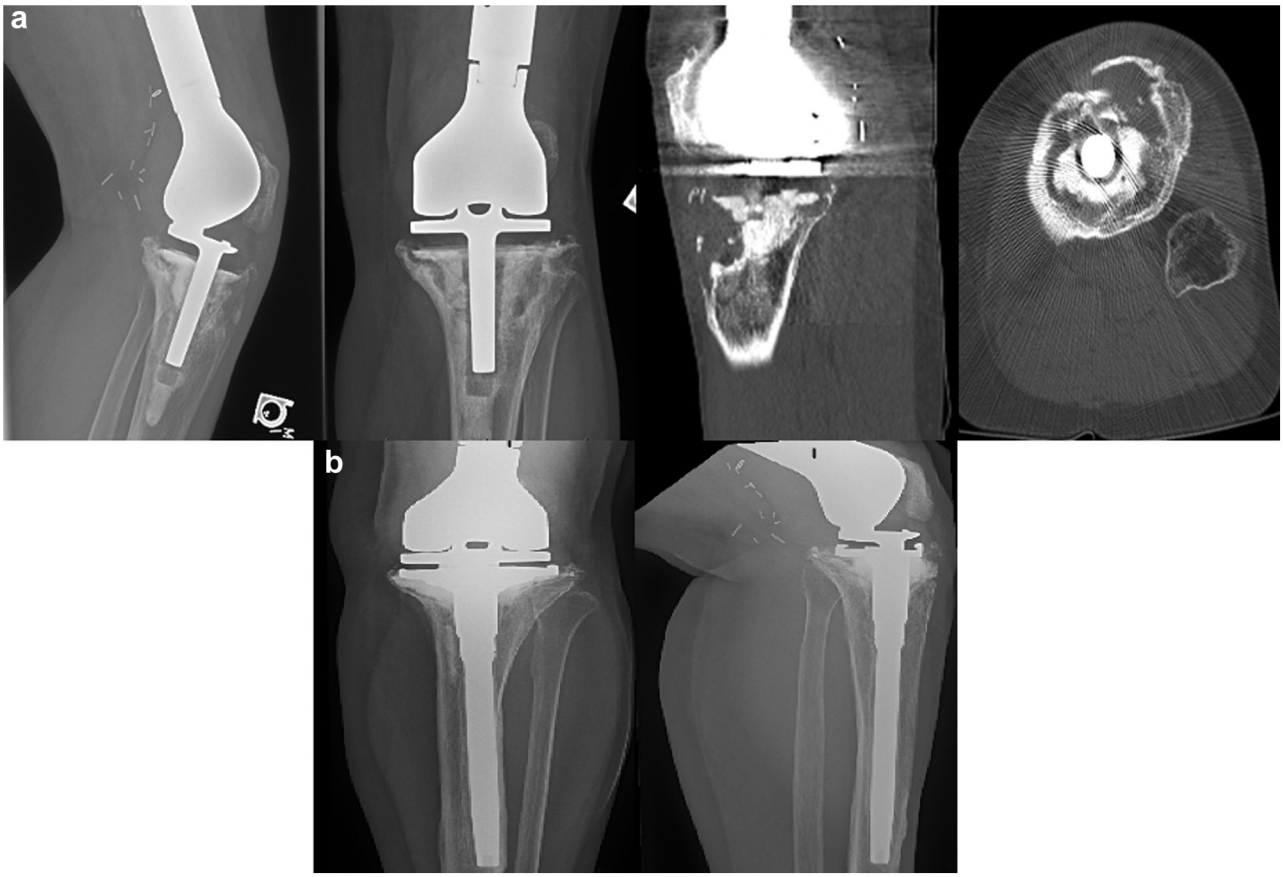

Figure 3. Patient A, left knee. (a) Repeat plain radiograph and CT images demonstrating progression of the osteolytic lesion and break of the cement mantle of the tibial component at 6 y after the initial revision. (b) Plain radiographs 6 y after curettage of the granuloma, revision of the all-polyethylene tibial component to a long press-fit stem with a cemented base plate, and impact grafting of the proximal tibia. Although radiographic findings suggestive of loosening of the tibial stem were observed, these radiographic changes have remained stable since the all-polyethylene tibia revision, and the patient remained asymptomatic.

underwent curettage of the granuloma, a revision of the all-polyethylene tibial component, which was found to be loose, to a long press-fit stem with cemented base plate, and impaction allografting of the proximal tibia. The femoral component and the bushing were revised (Fig. 3b). The histological assessment of the curetted tissues from the defect showed evidence of reactive fibrosis with diffuse histiocytic infiltration and intracytoplasmic fine polyethylene particulates, consistent with polyethylene granuloma (Fig. 4). The patient recovered completely and remained asymptomatic until 4 years after, 17 years after the index procedure, when she presented to the emergency department after an episode of knee instability. At the time, a left thigh deformity was observed. Further investigations revealed a fatigue failure and fracture of left distal femur MRS stem implant at the level of the male morse taper. Therefore, she underwent a removal of left distal femur cemented MRS stem, followed by a revision with a standard Compress stem (Zimmer Biomet, Warsaw, IN) and a custom-made Compress to MRS taper adaptor component, maintaining distal femur MRS component and prosthetic knee in situ. At the recent 2.5-year clinical follow-up visit, the patient was doing very well with a normal extensor mechanism, and the range of motion of her knee was 0 to 90°. She has returned to her preoperative activity level. Although radiographic findings suggestive of loosening of the tibial stem were observed (Fig. 3b), these radiographic changes have remained stable, and the patient remained asymptomatic.

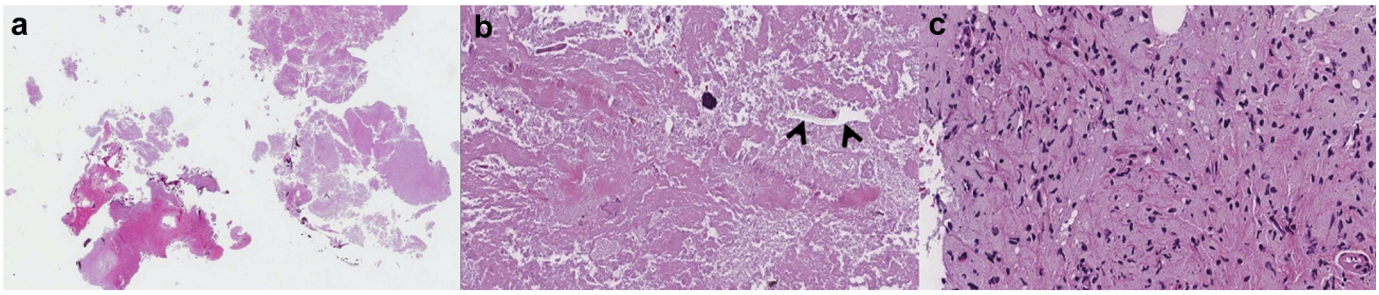

Figure 4. Surgical specimen of patient A. (a) Periprosthetic tissue with osteolysis and fibrinous debris. (b) Refractile thread-like polyethylene macroparticle (black arrowheads) free in fibrinous debris. (c) Sheets of macrophages containing fine polyethylene microparticles in the cytoplasm of macrophage.

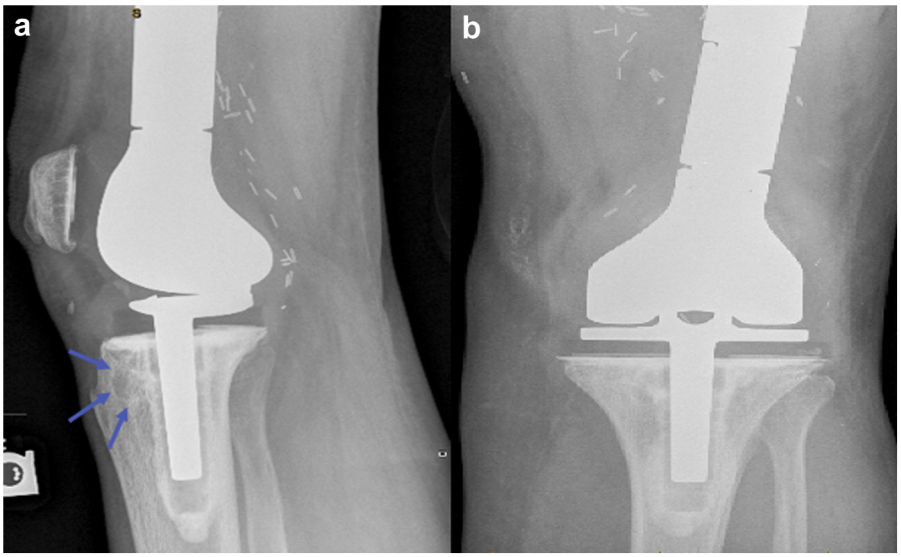

Figure 5. Plain radiographs showing polyethylene granuloma (blue arrows) in patient B, left knee. Initial presentation at 3 y of follow-up with asymptomatic benign looking lesion posterior to the tibial tubercle. (a) Lateral view. (b) Anterior-posterior view.

Case 2

Patient B is a 69-year-old woman who was diagnosed with distal femur Ewing's sarcoma. The patient was successfully treated with left distal femur wide resection and reconstruction using a DFR (GMRS; Stryker, Kalamazoo, MI) and an all-polyethylene tibial component. Her early postoperative course was unremarkable. At the 3-year follow-up visit, plain radiographs of the knee showed a well-circumscribed, benign-appearing lytic lesion just posterior to the tibial tubercle (Fig. 5). The tibial stem was intact without any signs of loosening. A bone scan demonstrated no evidence of a malignant recurrence, and blood markers were not suggestive of an infectious process. As there was a low clinical and radiological suspicion of an aggressive process and the patient remained asymptomatic, it was decided to follow up this lesion. The lytic defect slowly enlarged over the subsequent 6 years, while the patient remained asymptomatic. However, the patient developed an acute onset pain to her knee after a lower energy trauma. A plain radiograph showed a nondisplaced fracture of the tibial tubercle and progression of the lytic lesion involving the proximal tibial metaphysis anteriorly (Fig. 6a). A subsequent CT scan showed extensive osteolysis around the proximal tibia with a small soft-tissue mass breaching the anterior cortex (Fig. 6b). In view of our previous experience, the decision was made to proceed with curettage of the granuloma after intraoperative histological confirmation and ruling out an infection or tumor, followed by bone allografting and conversion of the all-polyethylene tibia into a cemented metal-backed, long-stemmed tibial component (Fig. 6c). Intraoperatively, the granulomatous lesion was identified anterior to the old all-polyethylene tibial stem that was found stable and well-fixed. The surgical specimen of granuloma showed multiple irregular tiny fragments representing osteolysis and extensive foreign body giant cell reactions to polyethylene macroparticles, confirming the diagnosis of polyethylene granuloma (Fig. 7). Three and a half years after curettage (Figs. 6c), and 12 years after her index procedure, the patient remains asymptomatic and returned to her original baseline function. The range of motion of her knee was 0 to 100°, and the strength of the extensor mechanism has returned to the preoperative level.

Discussion

In joint arthroplasty, it is well recognized that accumulation of wear debris from prosthetic hardware can cause macrophage-driven foreign body reaction, which in turn leads to periprosthetic osteolysis [6-10]. Specifically, polyethylene particulate debris created by chronic abrasive wear of polyethylene components can cause massive periprosthetic osteolysis in arthroplasties of the hip and knee [6-10]. In some cases, foreign body reaction to polyethylene can also lead to the formation of a tumor-like soft-tissue mass, termed polyethylene granuloma or pseudotumor, often detectable by conventional imaging modalities such as plain radiograph and CT [6-10]. This is particularly notable in total hip arthroplasty where the formation of polyethylene granuloma on the femoral side has been more commonly reported [9-14]. Histologically, polyethylene granuloma is characterized by histiocyte infiltration and evidence of polyethylene particulates engulfed by macrophages, circumscribed by multinucleated foreign body giant cells [8-10,15].

Unlike the femoral side [3], tibial side failure appears a less commonly reported complication after DFRs. Despite its frequent use in various designs of DFR, there is limited literature reporting complications specific to all-polyethylene tibial components [3,4]. Interestingly, aseptic loosening of implants has been reported as a culprit source for revisions [1]. In a recent retrospective review, a total of 15 (5.2%) mechanical failures of all-polyethylene tibial components were identified [16]. Of these, 6 were due to aseptic loosening, and 9 were due to structural failure [16]. Others have reported 2 revisions in all-polyethylene tibial components, one due to aseptic loosening and the other due to failure of the rotating hinge [17]. In another study using a different DFR prosthesis of similar design, authors did not report any aseptic loosening on the tibial side with all-polyethylene tibial components [18]. Nevertheless, given that the DFR prosthesis used in this study is from a different manufacturer, thus having unknown biomechanical properties, it is difficult to draw direct comparisons. To our knowledge, no literature to date has reported a case of polyethylene granuloma formation in DFR with an all-polyethylene tibia. Similarly, a limitation of our case report is the low number of cases,

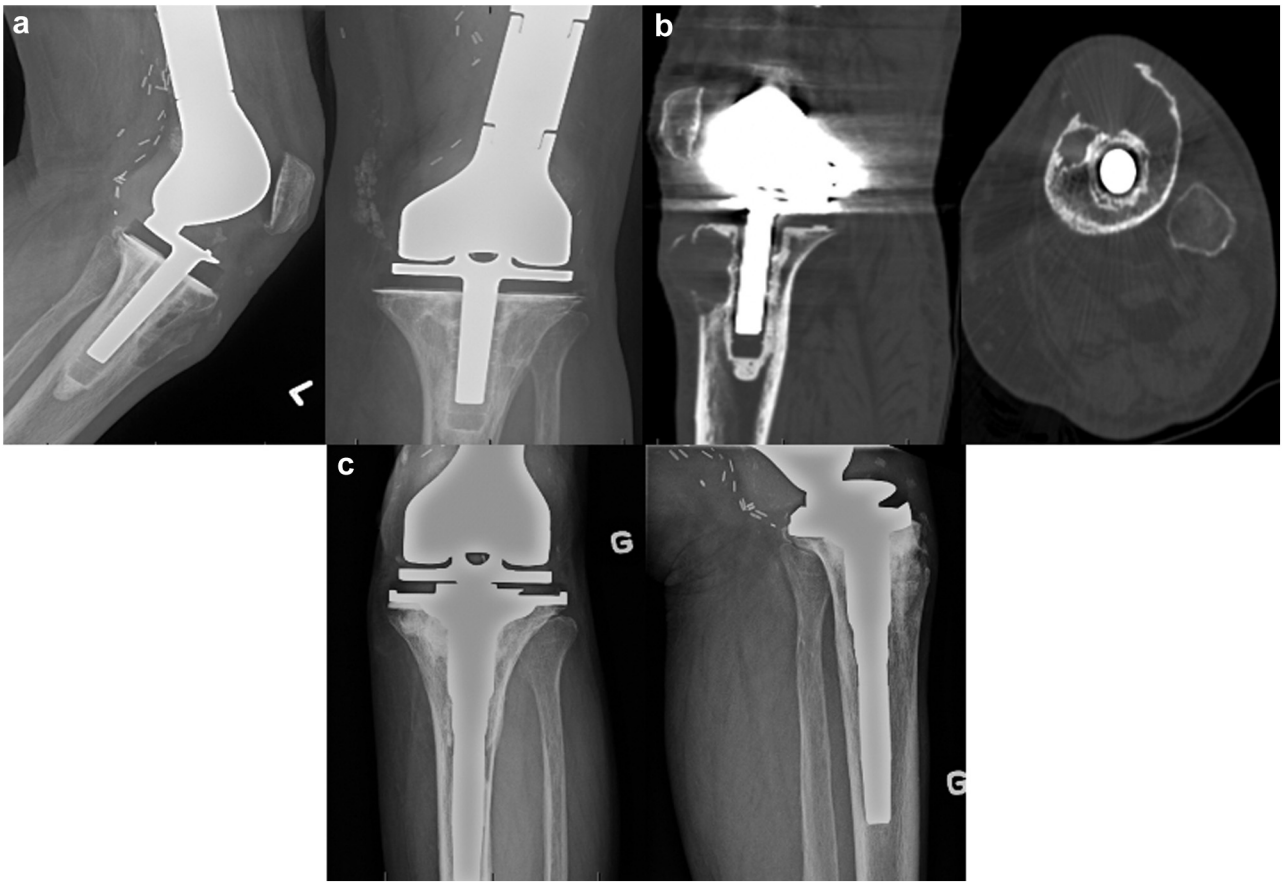

Figure 6. Patient B, left knee. (a) Plain radiographs demonstrating a nondisplaced fracture of the tibial tubercle and a progressive osteolysis. (b) CT images demonstrating periprosthetic osteolysis with a soft-tissue mass breaching the anterior cortex. (c) Plain radiographs 3 and a half years after curettage of the granuloma, revision of the all-polyethylene tibial component to a long press-fitted stem with cemented base plate and impact grafting of the proximal tibia.

which prevents estimating the true incidence of this devastating complication. Nevertheless, from our experience of over 150 cases with all-polyethylene tibial implants with such knee design performed over a 30-year period, these were the only 2 cases identified with a failure from the tibial side specifically. Therefore, the tibial component appears to be an unusual site for implant failure after a DFR.

The tibial components used in DFR reconstructions are either cemented metal-backed or all-polyethylene implants. It has been suggested that there were no differences in the tibial component failure rate, infection rates, functional outcome, or the total implant survival between the 2 tibial component options [17]. However, periprosthetic fractures may be more prevalent in patients with a metal-backed tibial component because of its higher rigidity and thus absorbing less mechanical stress than an all-polyethylene tibia [17]. A 3-dimensional biomechanical analysis comparing all-polyethylene tibia and metal-backed tibia demonstrated that, although the shear stress on the bone-cement layer around the all-polyethylene tibial component is relatively high, the stress distribution at the proximal cancellous and cortical bones was superior and more uniform than that at the metal-backed tibial component [19]. Nevertheless, factors such as bone quality, cementing techniques, and surrounding soft tissues needed to be controlled for in the analysis. An additional advantage of an all-polyethylene tibia is

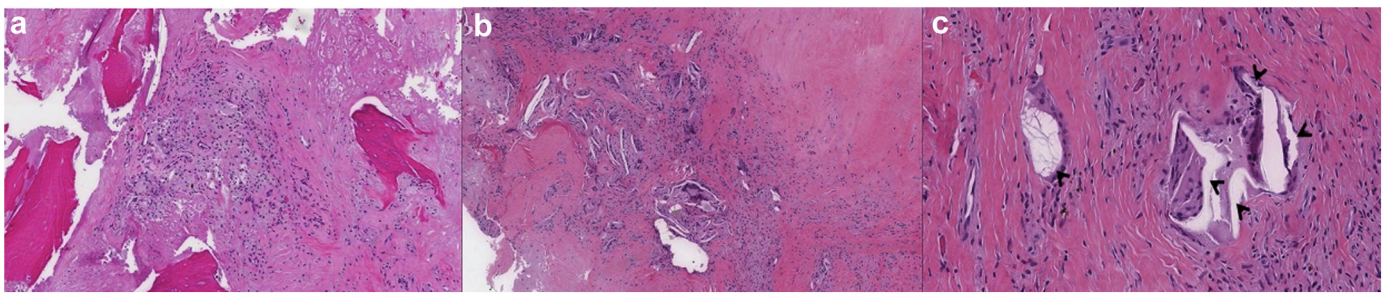

Figure 7. Surgical specimen of patient B. (a) Diffuse lymphohistiocytic infiltrates with osteolysis and necrosis. (b) Foreign body type granulomatous inflammation with polyethylene macroparticles and necrosis ("polyethylene granulomas"). (c) Refractile and frequently curved polyethylene macroparticles (black arrowheads) surrounded by multinucleated giant cells.

its lower cost than a metal-backed tibia [17]. In our first case, we noticed some micromotion in the proximal component of the tibial implant while the stem remained well-fixed distally. This can be due to the cantilever effect on the implant due to poor bone quality proximally. In such cases, metal-backed implants might be a more suitable option to delay catastrophic failure of the implant. Whether one prosthesis design could predispose more to the incidence of polyethylene granuloma is unlikely to be recognized based on the seemingly very low incidence of such complication. In addition, wear debris involved in the process may originate from the plastic hinge parts, such as bushings and bumper, and may not relate to the tibial implant design. Therefore, an all-polyethylene tibia may be the preferred option for primary endoprosthesis reconstruction without tibial bone loss, while the metal-backed tibia with its available modular parts could be used for revisions or poor-quality bone stock [17].

In this study, we reported 2 cases of polyethylene granuloma in patients with primary bone sarcoma who underwent DFR with all-polyethylene tibial components. In both cases, radiological evidence of intraosseous granuloma formation and periprosthetic osteolysis was observed at the anterior aspect of the tibial metaphysis. Interestingly, both cases presented with nondisplaced tibial tubercle fracture from bone insufficiency. The anterior location of both granulomas could be due to the design of the implant, as there is a shorter distance from bone periphery to the central peg anteriorly. Another theory is the chronic rubbing of the patellar tendon over the bumper and/or the anterior aspect of the all-polyethylene tibial component. After ruling out possible infectious etiologies and local malignant recurrence, both underwent an operative debridement of polyethylene granuloma and necrotic tissues followed by a revision to a long-stem, cemented metal-backed tibia and bone grafting.

Despite its significance, polyethylene granuloma formation appears a rare complication, and perhaps more attention should be given to tibial side implant failures in DFR series. In our cases, we reconstructed the defect using impaction allografting. However, porous implants such as metal cones or sleeves may be alternative options to impaction grafting. In these lesions, we suggest an initial thorough assessment to rule out infection and tumor recurrence. Patient symptoms and lesion progression should thus be monitored to counsel patients on the eventual need of a complex revision surgery and its associated risks. We would recommend revising the all-polyethylene tibial component to a metal-backed implant supplemented as necessary with trabecular cone or bone graft if the patient becomes symptomatic, if there is radiographic evidence of implant instability, or if there is evidence of tibial tubercle involvement.

Summary

Two cases of intraosseous polyethylene granuloma formation were observed in patients who underwent DFR with all-polyethylene tibial components. Both cases were associated with an extensive osteolysis and a nondisplaced tibial tubercle fracture. They required an operative debridement of polyethylene granuloma and necrotic tissues, followed by a revision to a long-stem, cemented metal-backed tibia and bone grafting. Our case report highlights polyethylene granuloma as a rare but significant complication of DFR with an all-polyethylene tibia. Patient symptoms and lesion progression should thus be monitored to counsel patients on the eventual need of a complex revision surgery and its associated risks. We would recommend revising the all-poly tibial component to a metal-backed implant supplemented as necessary with trabecular cone or bone graft if the patient becomes symptomatic or there is evidence of implant instability radiographically or if there is evidence of tibial tubercle involvement.

Conflict of interests

The authors declare there are no conflicts of interest.

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