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Case Report

Total Knee Arthroplasty and Atypical Cartilaginous Tumor/Enchondroma of the Distal Femur

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ABSTRACT

Enchondroma is a common benign chondrogenic tumor, which typically occurs in the short bones of hands and feet. However, when affecting the long bones, it is difficult to rule out the low-grade chondrosarcoma, called atypical cartilaginous tumor (ACT), because of the highly similar clinical and radiologic features. This study reports 2 patients with advanced knee osteoarthritis, scheduled for total knee arthroplasty, who had a distal femoral lesion on imaging suggestive of ACT/enchondroma. We believe that the treatment of these patients could be a challenge for arthroplasty surgeons. This is because it might be difficult to decide whether a periarticular chondral tumor of an osteoarthritic knee is malignant and changes the plan. In this report, we described our approach to address both knee osteoarthritis and ACT/enchondroma of the distal femur. To the best of our knowledge, this issue has not yet been discussed in the literature.

Introduction

Cartilaginous tumors comprise a benign to malignant spectrum from enchondroma to high-grade chondrosarcoma [1,2]. Enchondroma is a common and benign primary chondrogenic tumor, which occurs in the bones of endochondral origin [2]. It is often asymptomatic, and when it becomes symptomatic, it shows nonspecific symptoms. Most cases are diagnosed only after a pathologic fracture, regional pain, or incidentally on imaging [3]. An atypical cartilaginous tumor (ACT) is the lowest grade of chondrosarcoma, named by the World Health Organization since 2013, which is usually challenging to distinguish from enchondroma. The differentiating features of both have been the subject of many studies so far [4-6].

The treatment of ACT/enchondroma is usually conservative with observation and follow-up; however, when clinical and radiologic features suggest malignancy, surgery is indicated [7-9]. Extralesional resection is the gold-standard treatment for the ACT, as it provides the highest oncological safety [8]. However, as it is usually difficult to distinguish between both entities based on clinical and radiologic grounds, overtreatment of enchondroma and undertreatment of ACT may ensue [10]. Moreover, the pathological differentiation of enchondroma and ACT is exceedingly challenging because of highly similar histological features. Therefore, if there is no evidence of high-grade chondrosarcoma in clinical and radiologic studies, a biopsy is not typically indicated preoperatively. It is helpful for more aggressive, that is, grades 2 and 3 chondrosarcoma [3,11]. The magnetic resonance imaging (MRI) features suggestive of high-grade chondrosarcoma, which entails a preoperative biopsy, include peritumoral edema, periostitis, cortical expansion, cortical destruction, and extension to soft tissue [12].

In this case report, we presented 2 cases of advanced knee osteoarthritis scheduled for total knee arthroplasty (TKA), who had a periarticular ACT/enchondroma. We believe that the treatment of these patients could be a challenge for arthroplasty surgeons, as it might be difficult to decide whether a periarticular chondral tumor of an osteoarthritic knee is malignant and changes the plan. It is especially important in areas where there are not enough musculoskeletal tumor surgeons. In this report, we described our approach to address both knee osteoarthritis and ACT/enchondroma of the distal femur.

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Case histories

Case 1

A 55-year-old woman came to our clinic with left knee pain, which had been compromising her daily activity for 4 years. On examination, she had tenderness, crepitation, and a limited range of motion consistent with severe osteoarthritis. No mass was palpable. She had no marked past medical or surgical history. Radiography showed degenerative changes of the knee and a calcifying mass of distal femoral metaphysis (Fig. 1a). No periosteal reaction or soft-tissue mass was noted. We performed MRI, which showed a 33*22*21-mm intramedullary lesion with the involvement of anterior and medial cortices of the distal femur, endosteal scalloping, no soft-tissue mass, and no involvement of the joint space (Fig. 1b). The bone scan revealed an increased uptake of technetium-99 in the lesion. With a diagnosis of ACT/enchondroma, she underwent TKA and intralesional treatment in one session. The postoperative radiograph is shown in Figure 1c. The postoperative Musculoskeletal Tumor Society score was 96.67%. This scoring system is a well-established measure of pain, emotional acceptance, and functional outcome after musculoskeletal tumor reconstructions. It has been frequently used as a valid end-outcome measure, which is comparable between different studies [13]. The pathology result was reported as enchondroma, as seen in Figure 1d. In a 5-year follow-up, we found no complication or recurrence of the lesion.

Case 2

A 65-year-old woman presented with right knee pain for 3 years, not relieving by rest recently. Physical examination showed similar findings consistent with severe osteoarthritis. The past medical or surgical history was clear. Besides degenerative joint disease, radiography revealed a similar distal femoral metaphyseal mass with scattered areas of calcification in favor of a chondral tumor (Fig. 2a). The MRI showed a 38*27*25-mm intramedullary mass with the involvement of anterior and medial cortices of the distal femur and endosteal scalloping (Fig. 2c). The lesion showed increased uptake of technetium-99 in the bone scan, as shown in Figure 2b. With the same diagnosis as that of case 1, she went through the same surgical procedure (Fig. 2d). The pathology result was enchondroma as well. The postoperative Musculoskeletal Tumor Society score was 93.34%. We found no complication or recurrence of the lesion over the 5-year follow-up.

Surgical technique

After appropriate anesthesia and standard prepping and draping, the knee was opened through an anterior longitudinal

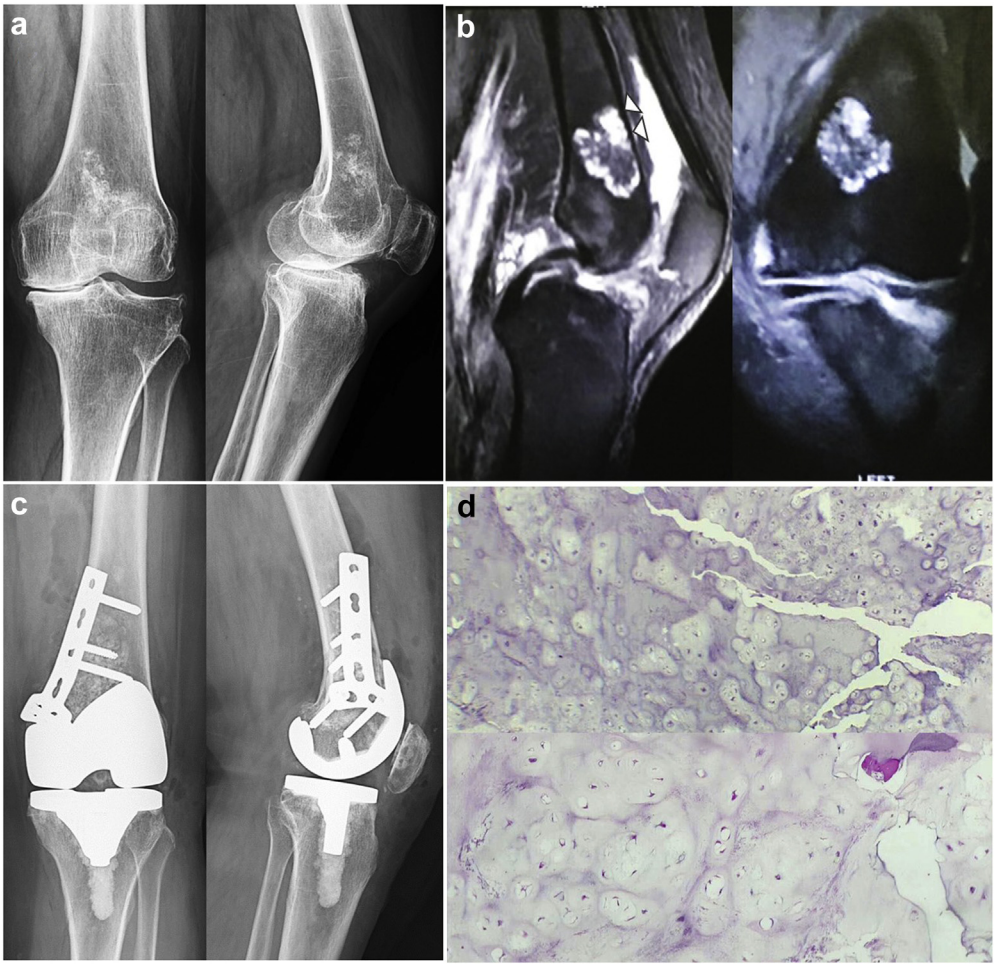

Figure 1. Case 1: preoperative anteroposterior and lateral radiographs of the left knee (a), T2-weighted sagittal and coronal MRI showing the involvement of the anterior and medial cortices of the distal femur with endosteal scalloping (white arrowheads) (b), postoperative radiographs (c), the photomicrograph of the lesion showing chondroid tissue in favor of enchondroma (d).

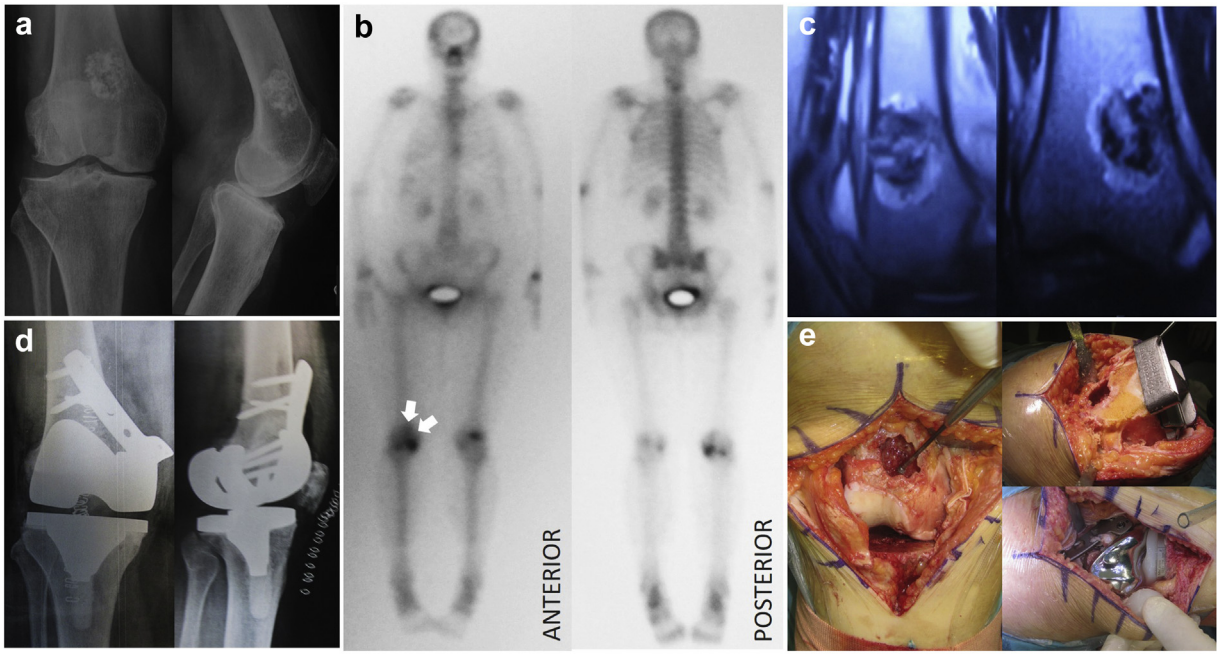

Figure 2. Case 2: preoperative AP and Lat radiographs of the right knee (a), the whole-body bone scan showing the increased uptake of Tc-99 by the lesion (white arrows) (b), T1-weighted sagittal and coronal MRI cuts (c), postoperative radiographs (d), intralesional curettage of the distal femoral chondral mass (left), performing the distal femoral cuts after curettage of the lesion (right, up), the final photograph after prophylactic plating and TKA prosthesis implantation (right, down) (e).

midline incision and medial parapatellar arthrotomy. By releasing the medial soft tissue, the patella was everted, and the proximal tibia was exposed. We performed tibial cuts using an extramedullary guide. Then, we opened an anterior cortical window on the distal femur by orthopaedic burr and performed intralesional curettage with a 5-millimeter margin. Subsequently, liquid nitrogen was poured into the cavity to ablate the margins. It should be noted that the complete curettage and ablation of the lesion must be performed before the insertion of the femoral intramedullary guide (Fig. 2e). The obtained tissue was sent for pathologic study. As the preoperative clinical and radiologic workup showed nothing in favor of high-grade chondrosarcoma, the frozen section or preoperative biopsy was not performed. Moreover, the histological differentiation of enchondroma and ACT not only is challenging but also did not change the treatment plan. In the next step, we performed the femoral cuts using an intramedullary guide. The lesion cavity was filled with prepared autograft from bone cuts, and prophylactic medial plating was performed by a 7-hole T-shaped plate and 5 conventional screws. Finally, we embedded the femoral and tibial components by cement fixation and inserted the polyethylene liner. The prosthesis was Zimmer NexGen LPS-Flex Knee (Zimmer Biomet, Warsaw, IN) in both patients. After proper lavage and repair of the quadriceps tendon, the joint capsule was sealed, and standard wound closure and dressing were performed.

Discussion

Chondrogenic tumors of the appendicular skeleton, other than those of hands and feet, have been known as a diagnostic dilemma in literature over the past 3 decades. Among the first who tried to find a solution were Murphey et al., who presented a valuable set of clinical and radiologic features to help differentiate between enchondroma and chondrosarcoma of long bones [14]. However, as more of the spectrum of cartilaginous tumors was understood, the studies were more narrowed to distinguish between enchondroma and ACT (the World Health Organization nomenclature for low-grade *appendicular* chondrosarcoma) [7,15,16]. It will be even more challenging if the lesion lies close to another musculoskeletal pathology such as osteoarthritis. Our study reports 2 patients with complaints of knee pain who had both knee osteoarthritis and metaphyseal chondral lesions of the distal femur. Whether the lesion is the source of pain or osteoarthritis and what decision should be made regarding the treatment approach are the questions we are to discuss.

In comparing the clinical features of ACT and enchondroma, pain is the only constant feature deemed in favor of ACT by almost all the studies [14,15,17]. Errani et al. argued pain as a differentiating feature, as they found it more frequent in enchondroma than in ACT. It was because most enchondroma cases were placed beside other painful pathologies, for example, osteoarthritis or tendinopathy [7]. Our patients had both complaints of disabling knee pain. The features in favor of osteoarthritis include short-duration stiffness after inactivity, joint-line tenderness, pain aggravation over range of motion, and crepitation [18]. On the other hand, rest or nocturnal pain and local tenderness over the lesion are the features describing tumoral pain. In the study of Ferrer-Santacreu et al., the frequency of pain on palpation was 58.2% and 88.2% in enchondroma and ACT, respectively. Although the frequency of pain was significantly more in ACT, more than half of the patients with both enchondroma and ACT had pain [15]. The nature of pain also did not show a significant difference. Moreover, both lesions could produce either mechanical (74.6% enchondroma, 58% ACT) or inflammatory pain (25.4% enchondroma, 42% ACT) [15]. Therefore, these features are impractical in differentiating both entities from either each other or osteoarthritis. Pain relief by rest, nonsteroidal antiinflammatory drugs, or intra-articular lidocaine or steroid injection has been suggested to differentiate the pain source. However, these maneuvers can cause misinterpretation, as both enchondroma and ACT might be symptomless. Thus, the pain caused by osteoarthritis may be relieved by injection, although a periarticular chondral lesion still exists. As a result, the differential diagnosis of ACT/enchondroma is usually made on radiologic and not clinical grounds [14,19,20].

To differentiate ACT from enchondroma of long bones, Ferrer-Santacreu et al. have proposed the metaphyseal location, size >5 cm or growth, calcification lysis over time, cortical involvement in computed tomography/MRI, and soft-tissue mass in computed tomography/MRI as radiologic and tecnitium-99 uptake ≥ anterosuperior iliac spine as metabolic aggressiveness features [15], whereas Errani et al. found endosteal scalloping and soft-tissue extension helpful [7]. Both our patients had metaphyseal chondral masses of the distal femur, which showed cortical involvement and endosteal scalloping on MRI and increased Tc-99m uptake in the bone scan. These features in a patient with advanced age require surgical intervention. Our patients also had TKA indication due to osteoarthritis; thus, we planned surgery for them. If no concomitant osteoarthritis was present, current literature would propose intralesional treatment for an ACT/enchondroma of a long bone [6,21]. Nevertheless, to the best of our knowledge, there is no study regarding the treatment of ACT/enchondroma around an osteoarthritic knee with indications of TKA.

According to available literature, 3 approaches could be assumed in the treatment of such lesions: conservative therapy and follow-up, wide extralesional resection and tumor prosthesis implantation, and intralesional treatment combined with TKA [4,6,22,23]. As far as the pain is tolerable and the radiologic features of lesion do not imply aggressiveness, conservative therapy and follow-up seem prudent [4]. Wide extralesional resection and tumor prosthesis implantation are the gold-standard treatment providing the highest oncological safety [22,23]. However, it is not recommended for ACT because of a higher rate of complications and more compromised functional outcomes in comparison with intralesional treatment [24-26]. This paradigm shift in the treatment of ACT has occurred as recent studies have shown comparable oncological safety and better functional outcomes for intralesional treatment [6]. In fact, wide resection is kept for lesions showing radiologic features of high-grade chondrosarcoma such as cortical destruction, periosteal reaction, and soft-tissue mass [14]. Intralesional treatment includes complete curettage of the lesion with or without adjuvant therapies such as cryosurgery, phenol, and polymethylmethacrylate packing. As Chen et al. [6] showed in a recent comprehensive meta-analysis, it does not increase the risk of local recurrence, hence its oncological safety. In case of a large cortical defect after intralesional curettage, prophylactic plating or the use of a stemmed femoral component should be considered to provide stability. However, stemmed components are usually more expensive, at least in our institute. Furthermore, in the same way as intramedullary guides, intralesional curettage and ablation should be performed before stem insertion to avoid spreading possibly malignant cells. We propose that intralesional treatment could be performed combined with TKA for periarticular ACT/enchondroma when knee osteoarthritis coexists. However, the surgical technique is determining as it might lead to the iatrogenic spread of probably malignant cells if performed improperly. No previous report of the technique was found in the literature. It must be noted that the intramedullary guide used for doing femoral or tibial cuts should be inserted only after complete curettage and ablation of distal femoral or proximal tibial lesion, respectively.

In our review of literature, only 3 cases were found, in whom distal femoral ACT/enchondroma and concomitant knee osteoarthritis were treated. In a 2-year follow-up study of 108 patients undergoing intralesional curettage for the ACT, 5 cases had local recurrence. One of them, who also had knee osteoarthritis, underwent combined TKA and repeated intralesional curettage for the distal femoral metaphyseal lesion. The pathologic diagnosis was not reported [27]. In another radiologic follow-up study of 49 patients with untreated ACT/enchondroma of long bones, eight patients needed surgery. The indication of surgery was knee osteoarthritis in 2 cases, who underwent TKA with intralesional curettage of distal femoral lesions (size: 2.7 and 3.2 cm). The pathologic diagnosis was enchondroma for both cases [4]. The pathologic diagnosis was also enchondroma for our cases. Although both patients of our study underwent prophylactic plating, it was performed in none of the literature cases. No follow-up was reported for the literature cases; however, in our 5-year follow-up, no complication or recurrence was seen and both patients had good functional outcomes.

Summary

On approaching toward a concomitant periarticular knee ACT/enchondroma and osteoarthritis, if TKA is indicated, one can perform intralesional treatment simultaneously with TKA. However, we recommend that the complete curettage and ablation of the lesion are performed before inserting the femoral or tibial intramedullary guide to avoid spreading malignant cells if present. Nevertheless, further studies are required to fully establish the outcomes of this surgical technique and other possible treatment approaches.

Conflict of interest

The authors declare there are no conflicts of interest.

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