A 74-year-old male with a prior left total knee arthroplasty presented with deformity, loosening, pain, and stiffness associated with multiple raised, erythematous, cutaneous nodules about the anterior knee. Workup was concerning for infection, but the skin nodules were atypical. The patient was sent for biopsy which revealed cutaneous diffuse large B-cell lymphoma. The revision surgery was delayed, and the patient underwent chemotherapy/radiation with complete resolution of his lymphoma. He then underwent a successful aseptic revision total knee arthroplasty. Proper identification and treatment of rare cutaneous skin lesions about a prior surgical site can limit morbidity and result in more desirable outcomes.

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Introduction

Identification and treatment of postoperative complications after total knee arthroplasty (TKA) are undeniably important. While postsurgical skin changes can indicate infection, skin necrosis, or benign etiologies, it is important to be aware and hence identify unusual manifestations. Improper management without a definitive diagnosis can lead to increased morbidity and poor outcomes [1].

 Upon review of the literature, there are only 11 case reports of postoperative metallic-implant-associated lymphomas [1–13]. Furthermore, only 2 such reports are concerning a TKA. In this case report, we aim to describe a clinical situation where if one exercises an abundance of caution and obtains an accurate diagnosis, patient morbidity can be avoided with an excellent patient outcome.

Case history

A 74-year-old male presented to our clinic after he had a revision left TKA performed by another surgeon 2 years prior. The patient presented with a valgus deformity, pain, range of motion 15–80 degrees, and anteriorly based erythematous skin nodules (Fig. 1). No deep palpable mass was identified. Radiographs demonstrated a revision TKA (Triathlon TS; Stryker Orthopedics, Mahwah, NJ) implant with lucency around the femoral component and valgus malalignment (Fig. 2). The patient's implant before his subsequent revision was a Stryker Triathlon primary knee. We obtained standard laboratory workup for infection including aspiration, complete blood count, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), and bone scan. The knee aspiration was dry, CRP was >10 mg/dl (reported 0–10, >10), ESR 2.0 mm/h, and white blood cell count 6300/mL. Bone scan demonstrated “diffuse uptake about the femoral component concerning for infection.”

At this point, our suspicion for infection was high, but the anteriorly based skin nodules were atypical (Fig. 1). We then sent the patient to a dermatology team who biopsied the nodules which revealed a diffuse large B-cell lymphoma with cells being CD20-positive and CD3-, CD4-, and CD30-negative (Figs. 3 and 4). Computed tomography (CT) of the left lower extremity (LLE) revealed a 14.1 × 8.1-cm soft-tissue mass involving the anterior and posterior distal thigh musculature. A positron emission tomography scan revealed extensive multifocal intraosseous and anterior muscle compartment soft-tissue masses with infiltration into the distal femoral metaphysis (stage III). A bone marrow biopsy at the iliac crest was negative for disease.
The patient was recommended to undergo chemotherapy and treated with Rituxan-CVP (Genentech, San Francisco, CA) for 5 months. He was not a candidate for Adriamycin (Thymoorgan Pharmazie, Goslar, Germany) due to cardiac comorbidities. At the completion of his chemotherapy, a new CT was obtained, which showed the mass shrunk to 1.5 × 1 cm about the anterior distal thigh musculature.

The patient then received radiation therapy (45 Gy in 25 fractions to residual; 36 Gy to prechemo gross tumor volume) over the course of 1 month. Three months after the completion of the radiation therapy, a repeat CT scan revealed that “the previously described lesion in the distal thigh is not delineated in the current study.” At this point, the patient was deemed clear of disease and a candidate for revision surgery.

Repeat examination of the patients’ left knee demonstrated complete resolution of the cutaneous nodules and skin changes (Fig. 5). Repeat ESR, CRP, and complete blood count were normal. Repeat bone scan demonstrated no soft-tissue uptake, but persistent bony uptake in the femur. Repeat knee aspiration was dry. He continued to be symptomatic due to his valgus deformity, pain, and limited range of motion. The patient then underwent left-knee revision TKA to a long-stemmed, cemented femoral component (Stryker Triathlon TS); the tibial side was not revised (Fig. 6). The pathology of bone and soft tissue sent from the surgery revealed no visible tumor/lymphoma. Cultures and frozen sections for infection were negative. The total time from initial presentation to the ultimate revision surgery was 14 months.

At the 12-month follow-up from arthroplasty, there was no recurrence of lymphoma, and the patient returned to all activities of daily living (ADLs). He was working out 6 days a week and pleased with the result. The final range of motion was 3-110 degrees.
The patient was informed the data and images concerning the case would be submitted for publication and provided written consent.

Discussion

Primary non-Hodgkin lymphoma in association with an orthopedic surgery is very rare, with only 12 other reported cases in the literature [1–13]. Of these, only 3 occurred after a TKA [1,2,13]. The median latency to presentation was 8 years (range 7 months to 17 years). Only 4 of the 11 cases presented as soft-tissue manifestations, while the remaining 7 presented as bony radiologic findings [1–13]. Many of the cases provided a diagnostic challenge, with symptoms and signs mimicking other conditions such as loosening or infection.

Whether orthopedic implants have carcinogenic potential can often be a concern of orthopedic surgeons. Particle debris from wear has been shown to promote DNA instability through DNA and chromosomal injuries [14–16]. This damage has theoretical risk to induce malignancy of the surrounding tissues. Multiple early studies established a potential link to orthopedic implants and carcinogenic potential [17–19]. In a study of the rates of malignancy following total hip arthroplasties, Gillespie et al. observed a small increase in the overall prevalence of tumors of lymphatic and hematopoietic origin [20]. Despite the aforementioned studies, no epidemiological study has demonstrated a significant increase in the prevalence of malignancy, including lymphoma in patients who have been treated with orthopedic implants [21,22]. We do not believe the lymphoma in this case was caused by the orthopedic implants utilized in the primary or revision arthroplasties. It is more likely that the lymphoma was present at the time of initial revision or developed spontaneously thereafter.

Long discussions about potential complications after a TKA following radiation therapy should also be conducted with patients in this setting. The effects of radiation on bone have been studied extensively and found to cause osteonecrosis [23–25]. This could lead to increased rates of loosening; however, the addition of cement with stems and augments could help provide more reliable long-term results [26]. In this case, we utilized a long cemented stem with a cone in order to mitigate possible osteonecrosis and, hence, loosening. Furthermore, radiation in the preoperative setting can lead to increased wound-healing problems [27,28]. While radiation was necessary to treat the lymphoma, understanding the potential for these complications is important, and one should be prepared for rotation or free flaps as indicated to mitigate wound and soft-tissue healing problems. Lastly, radiation-associated knee stiffness should be discussed with patients. Stiff patients receive significantly higher mean doses to the suprapatellar bursa/anterior suprapatellar fat pad and quadriceps tendon than nonstiff patients. This suggests that radiation-induced shortening of the quadriceps tendon and damage to the synovial membrane lining the anterior suprapatellar fat pad may be contributing factors [29].

Abnormal presentations of postoperative wound complications in the setting of a prior TKA, therefore, should be approached with great caution. In our case, initial presentation was certainly concerning for infection. It would be easy to recommend a revision surgery with rotational or free flap in the acute period given the findings. In the only other similar case report published to date by Eskander et al., aggressive surgical management was undertaken [1]. The high suspicion of infection led to the need for both medial and lateral gastrocnemius flaps with knee fusion after it was discovered the patient had a diffuse cutaneous B-cell lymphoma [1].

Current controversies and future considerations

Whether orthopedic implants have carcinogenic potential can often be a concern of orthopedic surgeons. There are no data at this
time to support this concern. In this case, it is most likely that the disease was present at the time of the initial operations, and we do not believe the implants caused the tumor. Future considerations, however, could be aimed at studying this potential phenomenon.

Summary

Our case had an initial presentation almost identical to that described by Eskander et al. However, by using an abundance of caution, obtaining an accurate diagnosis, and undergoing proper treatment, the patient was able to have a significantly improved outcome with revision TKA and had no need for flap coverage or fusion. To our knowledge, this is the first case report to demonstrate the diagnosis, treatment, and resolution of diffuse B-cell lymphoma in association with TKA prior to undergoing a revision surgery. The case report demonstrates that a proper identification and treatment can lead to good patient outcomes. The limitation of the study is that we did not perform his prior surgeries and, therefore, do not know if this pathology was present at the time of his initial revision. It is our hope that the information presented will provide surgeons with a framework on how to approach this difficult problem.

Conflicts of interest

Dr. Palumbo receives royalties from DJO and Conformis; is a paid consultant for DJO and Conformis; and holds stock or stock options from Amedica and Zimmer; is the editor of the Journal of Arthroplasty; and is on the board of Florida Orthopaedic Society. Dr. Lyons receives and is on the Board of Councilors of American Academy of Orthopaedic Surgeons and Florida Orthopaedic Society. Dr. Wetzel declares no potential conflicts of interest.

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Informed patient consent

The authors confirm that informed consent has been obtained from the involved patient or, if appropriate, from the parent, guardian, power of attorney of the involved patient and that they have given approval for this information to be published in this article.

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