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Tumoral calcinosis complicating a reverse total shoulder arthroplasty: a case report

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There are three etiologies of soft-tissue calcification that are of importance to the orthopedic surgeon: articular chondrocalcinosis, calcific periarthritis, and tumoral calcinosis (TC).[6] TC is rare, is primarily associated with hyperphosphatemia and hypercalcemia, is often associated with end-stage renal disease (ESRD), and impacts soft tissue because of the deposition of dystrophic calcifications. Treatment for TC usually involves medical management primarily, with surgery used as an adjunct to decrease the pain level and improve quality of life.[1] The purpose of this article is to discuss the development of TC after reverse total shoulder arthroplasty, a complication that has not been previously reported after reverse total shoulder arthroplasty. Patient consent was obtained.

Case report

A 64-year-old right hand dominant Hispanic female with history of rheumatoid arthritis, renal cell carcinoma status after nephrectomy, and ESRD secondary to hypertensive nephrosclerosis originally presented and was hospitalized after sustaining a left proximal humerus fracture after a fall from standing (Fig. 1). Despite having a previously placed brachiocephalic arteriovenous fistula at the injured extremity, she underwent an uncomplicated, cemented reverse total shoulder arthroplasty with greater tuberosity repair on the initial hospitalization (Fig. 2). Despite resorption of the greater tuberosity (Fig. 3), the patient had a relatively uncomplicated postoperative course and was lost to postoperative clinical follow-up after three months.

One year postoperatively, the patient presented to the office with acute onset of severe left shoulder pain, erythema, and swelling of the affected left shoulder after a symptomatic urinary tract infection. Ultrasound-guided aspiration of the left glenohumeral joint returned 30 ml of cloudy fluid with a cell count differential of 2750 nucleated cells and 87% polymorphonuclear cells. Because her presentation was consistent with acute, hematogenous prosthetic joint infection, she was admitted to hospital and underwent débridement with glenosphere and polyethylene liner exchange. The glenoid baseplate and cemented humeral stem were retained. Calcium sulfate absorbable antibiotic beads (Stimulan, Biocomposites, Wilmington, NC, USA) loaded with vancomycin were placed (Fig. 4). Three separate cultures obtained from the left shoulder joint showed no growth for any organism after being held for 14 days. She underwent 6 weeks of treatment empirically with intravenous antibiotics under the care of an infectious disease specialist and had an otherwise uneventful recovery. Subsequently, she was again lost to follow-up for at least 8 months.

Through most of the second postoperative year, the patient had no complaints with regard to pain or function with her shoulder; however, at the end of that year, the patient began developing a mass on her left upper extremity. Because of her fistula positioning, she was referred without the authors' knowledge to a vascular surgeon who explored the area due to concern for a pseudoaneurysm. No pseudoaneurysm or vascular issue was noted in the

Institutional review board approval was not required for this case report.

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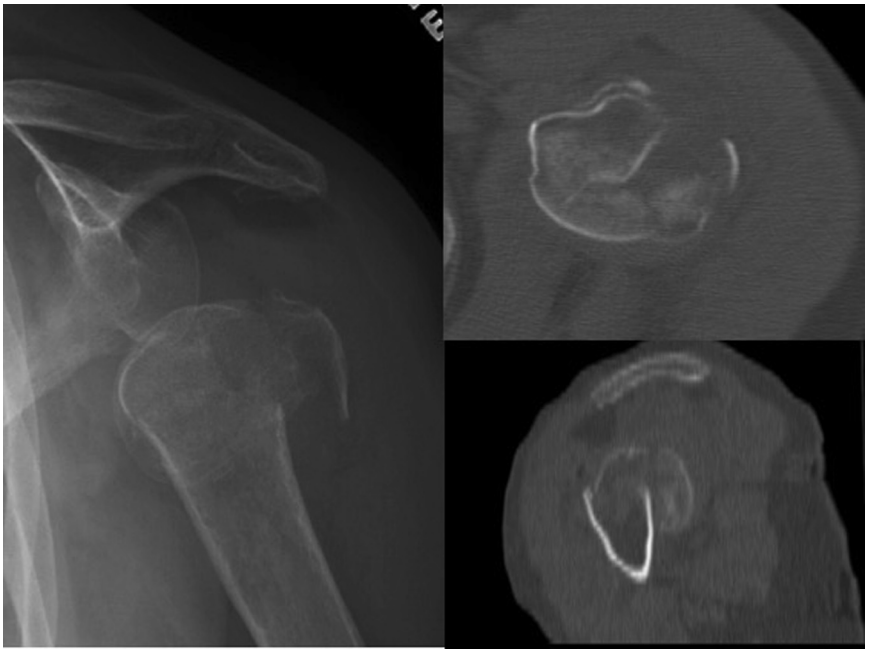

Figure 1 The initial anteroposterior radiograph and computed tomography (axial and sagittal views) demonstrating left proximal humerus fracture.

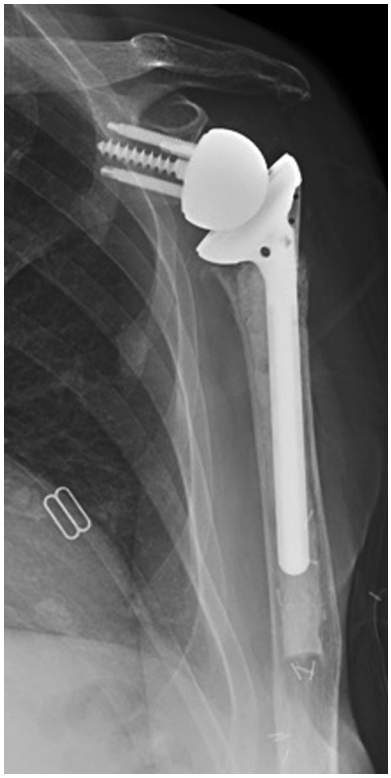

Figure 2 The postoperative anteroposterior radiograph after index procedure showing intact reverse total shoulder arthroplasty.

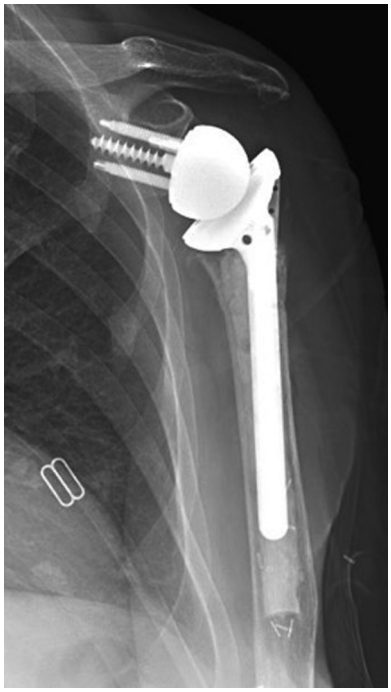

Figure 3 The six-month postoperative anteroposterior radiograph after index procedure demonstrating greater tuberosity resorption.

area. Radiographs and computed tomography were subsequently obtained as the mass persisted. Computed tomography showed a 3.6 x 4.1 x 9.2 cm fluid collection in the anterior aspect of the left upper arm, along with small foci of "heterotopic bone" formation adjacent to the medial proximal humerus metaphysis. Beginning at this time, calcifications were noted on imaging studies (Fig. 5).

At the beginning of the third year from the index operation, the patient presented again to the authors, and chronic prosthetic joint infection was presumed to be causing the painful mass. She consented for revision surgery. During this procedure, the joint was débrided arthroscopically because lifetime antibiotic suppression

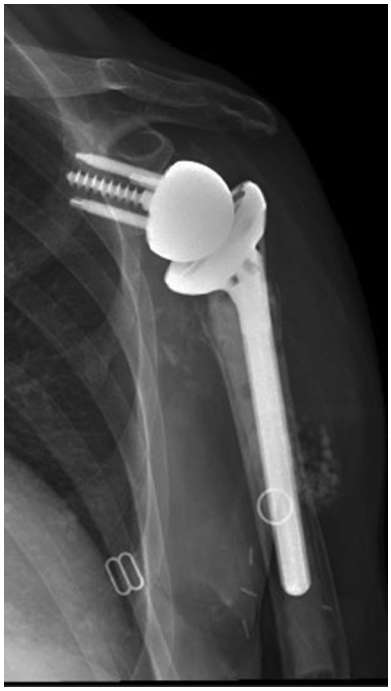

Figure 4 The postoperative anteroposterior radiograph after first revision surgery showing placement of Stimulan beads.

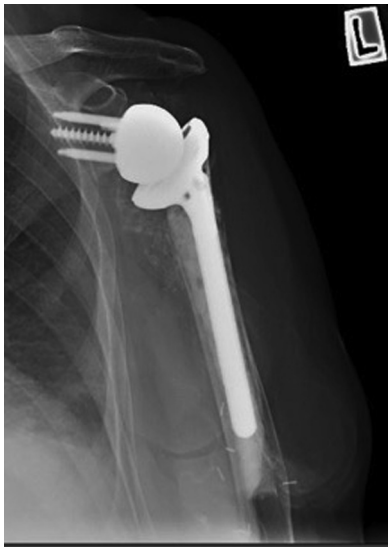

Figure 5 The anteroposterior radiograph demonstrating first signs of heterotopic bone formation.

was planned and the mass was addressed in an open fashion. A copious amount of cloudy white fluid was drained with the fluid appearance resembling a crystalline arthropathy. At this time, a noninfectious process was suspected because the prior operation for infection had not yielded any microbiologic diagnosis. The cysts histologically showed a fibrous capsule lined by granulation tissue with multinucleated giant cells and dystrophic calcifications (Fig. 6). On consultation with pathology (WM), infectious disease, and nephrology (RS) services, the diagnosis of periprosthetic TC was made.

In the fourth and fifth year from the index operation, the patient was medically managed by a nephrologist (RS) with medications

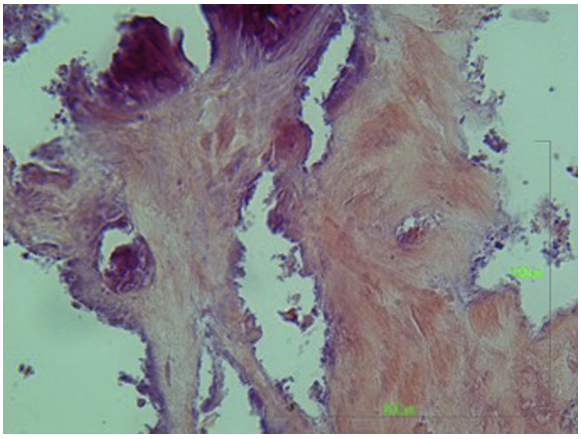

Figure 6 Pathology of specimen taken in OR demonstrating the presence of multinucleated giant cells and dystrophic calcifications.

such as paricalcitol, Parsabiv, Sensipar, calcium acetate, and sevelamer carbonate to control her hyperparathyroidism associated with her chronic kidney disease, administered with hemodialysis.

Late in the fourth year from the index operation, the patient presented again with a painful mass in the upper arm (Fig. 7). At this point, the authors abandoned medical management as a strategy to keep the size of the mass under control. Sequential aspirations were also minimally successful in relieving pain from the growing fluid collection in the upper arm.

In the sixth postoperative year, an orthopedic oncologist (FM) agreed that she was a reasonable candidate to undergo resection of the mass to try to achieve pain control. She underwent excision of the pseudotumor (Fig. 8) with glenosphere exchange to further stabilize the joint through lateralization. Intraoperatively, deep dissection showed that the mass was resting on the outflow portion of the patient's arterialized cephalic vein just distal to the cephalic arch. Pathology specimens showed amorphous calcium salts and calcium hydroxyapatite crystals in periarticular tissues, consistent with an overall picture of TC. There were no areas of malignancy or atypia noted, and cultures were again negative.

At the final follow-up, in the seventh postoperative year, she continues to have some pain and swelling around the left shoulder with acceptable function. Her goal continues to be avoidance of further surgery.

Discussion

One of the key principles of orthopedic practice is to consider alternative categories of disease (eg, oncologic, metabolic) when infection is being worked up and vice-versa. In this case, neglecting this principle resulted in delayed diagnosis and treatment of the true metabolic diagnosis. Considering her initial presentation of acute pain and fever after a urinary tract infection, infection was the obvious diagnosis to rule out; however, inclusion of TC in the differential diagnosis would have been warranted, given her comorbidities. In retrospect, a more comprehensive approach could have been taken early in the patient's presentation to help guide the orthopedic surgeon's treatment plan. Because confined tissue injury is a possible cause of TC formation, this could be a factor in the surgical decision-making process when repeated revisions or palliative surgeries are necessary.

TC complicating a total joint replacement is rare. Orthopedic surgeons must remain diligent and keep an open mind when it comes to working up periprosthetic soft-tissue masses, taking into account all medical, radiographic, and pathologic data. TC should be

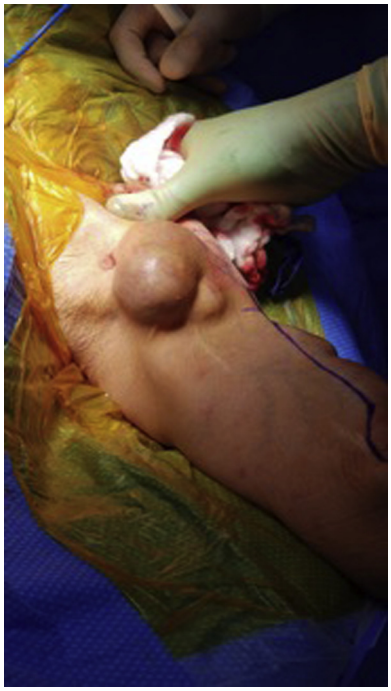

Figure 7 Intraoperative photograph showing extent of tumoral calcinosis mass in left upper extremity.

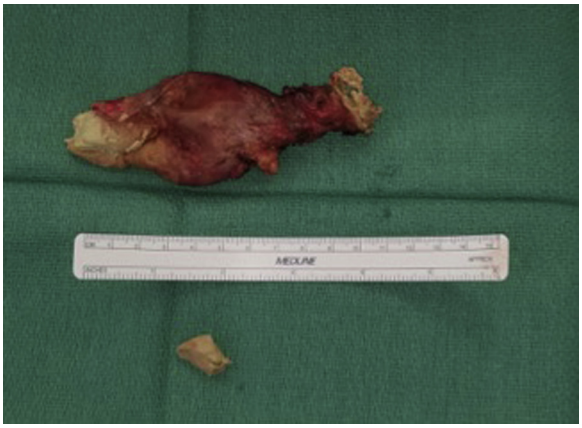

Figure 8 Photograph demonstrating mass 10 cm in length.

included in the differential diagnosis for a periprosthetic soft-tissue mass in the setting of chronic hemodialysis. This case report illustrates the great challenges of this perplexing problem in diagnosis and treatment. Even with an integrated approach, including advanced medical and surgical interventions, TC can only be symptomatically managed and will be a chronic source of pain and disability.

TC is a complicated disease process of unclear etiology that was first reported in 1943.[4] Renal disease, especially of long-term duration, has been implicated as a precursor for pathologic calcification. Although the exact mechanism of extra-articular calcium phosphate deposition remains imprecisely understood, several risk factors play a role including hyperphosphatemia, vitamin D intoxication, confined tissue injury, and secondary hyperparathyroidism.[3] TC has been reported in wide-ranging settings; however, the finding of TC after arthroplasty is a rare occurrence. One case report described an elderly Japanese man who formed TC after a total knee arthroplasty, with no causal relationship formed.[5]

Two different forms of TC have been reported in the literature.[2] The first type includes two subtypes, both with a familial predisposition, differentiated by either hyperphosphatemia or normal phosphate levels. Tiwari et al described a case of hyperphosphatemic familial TC in which a patient had symptomatic left hip pain and concomitant elevated phosphate levels.[9] The patient was started on a combination of phosphate-restricted diet, phosphate binders, and oral acetazolamide with limited effectiveness in reducing phosphate buildup. The second type is more closely related to ESRD and the ensuing metabolic derangements caused by hyperparathyroidism.[2] Smack et al previously had differentiated TC based on a pathogenesis system, with three distinct types: primary normophosphatemic TC, primary hyperphosphatemic TC with no identifiable cause, and secondary TC.[7] This system has faced controversy and was based on whether or not the patients had concurrent disease and the makeup of their electrolyte derangements rather than pathogenesis.[2]

Unlike in other calcification disease entities, TC has greater propensity for para-articular formation than periarticular with a propensity for shoulders, elbows, and hands primarily.[6] TC also has classic findings that are seen both radiographically and histologically. Steinbach et al described the radiologic findings in 12 patients with TC. The homogenous, calcified growth that has a cobbled appearance due to translucencies within the mass is consistently seen on radiographs.[8] Histologically, the multiloculated, cystic masses that form will be nodular in nature with a fibrous stroma that surrounds cavities containing multinucleated giant cells and dystrophic calcifications.[5] If left untreated or mismanaged, it can significantly impair the patient's mobility.

Treatment for TC can include both medical management and surgical resection. Although surgery historically has been used as a secondary means of pain relief, there has been a growing movement toward the management of the components in dialysis as a means of lowering phosphate. Medical therapy has been shown to be first-line treatment for patients with TC secondary to renal failure because of the metabolic derangements responsible for the calcium deposits. Medical management can include a host of treatment options, ranging from phosphorous chelation, phosphate restricted diet, to decreased calcium in the dialyzate.[1] All these options are effective at lowering serum phosphorus. Surgery can be performed to remove any mass in cases where decreased function of the affected joint, discomfort, persistent infection, and skin blistering are present; however, recurrence rates are high because of the extensiveness of soft-tissue invasion that these tumor-like masses exhibit.[2] As such, surgery should be a last resort for TC secondarily caused by other autoimmune or medical processes.

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Patient consent: Obtained.

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