Idiopathic Acute on Chronic Osteomyelitis Mimicking Metastatic Lesion in Humeral Head: A Case Report

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Introduction

Osteomyelitis is a common disease process that can manifest in a multitude of ways. We present the case of a 69-year-old male who was admitted to a hospital for deconditioning, acute renal failure, and volume overload. However, the limited range of motion of his left shoulder led to radiographic evaluation, which revealed unclear findings. Extensive workup revealed idiopathic acute on chronic osteomyelitis with the causative pathogen being identified as methicillin-resistant *Staphylococcus Aureus* (MRSA), the third most common cause of acute osteomyelitis in the United States.[1]

The annual incidence of osteomyelitis in the United States is estimated to be 90 in 100,000 people.[2] Acute osteomyelitis normally presents as soft tissue swelling, joint space changes, bony destruction, and periosteal reaction on plain film.[3] Chronic osteomyelitis may show areas of sequestrum, involucrum, and cloaca.[3] Chronic osteomyelitis is classified as recurrent acute osteomyelitis with large areas of ischemia, necrosis, and bone sequestrum. Clinically, chronic osteomyelitis can present both acutely with fever, swelling, and pain or more insidiously with subacute to chronic pain in the affected bone. Osteomyelitis in adults most commonly presents in the epiphysis and subchondral regions of the bone. The variation in the location of hematogenous osteomyelitis is believed to result from differences in the vasculature as bone ages. The lower extremity is most frequently affected, followed by the vertebrae, sacroiliac joint, and radial styloid.[4] The most common mechanism of osteomyelitis in adults is the polymicrobial contiguous spread of infection after trauma or secondary to decubitus/diabetic ulcers.[5] The pathogenesis of hematogenous osteomyelitis is believed to be due to a disruption or slowing of blood flow to the bone, allowing for the deposition and proliferation of infection.[5]

This patient's pathology is exceedingly rare when combining its absence of known trauma to the area except during adolescence; unique appearance on imaging; rare presence in the humeral head; and hematogenous spread. It is unclear what mechanism may have led to this unique presentation. Here we present a case of an unfortunate gentleman with atraumatic, hematogenous, acute on chronic osteomyelitis presenting as suspiciously metastatic appearing lesions on imaging.

Case Report

The patient is a 69-year-old male with a past medical history significant for type 2 diabetes, diabetic retinopathy, diabetic nephropathy, HTN, HLD, and gout who presented to an outside hospital with progressive weakness and falls. On initial evaluation the patient was found to have a urinary tract infection, acute renal failure, new onset ascites and was subsequently transferred to our hospital due to insurance concerns. The day following admission the patient was working with therapies when patient was noted to have decreased mobility in the left shoulder. A shoulder radiograph was obtained which showed: cortical irregularity of the greater tuberosity with probable lytic lesions in the humeral head concern for pathologic fracture (Figure I). A non-contrast CT of the left shoulder was obtained which showed: expansile lucency in the humeral head, neck, and proximal diaphysis with pathological fracture indeterminate for primary neoplasm versus metastatic/myelomatous disease. Additionally, CT showed soft tissue thickening and joint effusion with underlying septic arthritis unable to be excluded (Figure II). Furthermore, an MRI of the shoulder showed osseous erosions, numerous prominent intraosseous cysts/abscesses with osteomyelitis with septic arthropathy being the favored differential diagnosis, however unable to completely exclude an atypical malignant process (Figure III).

Based on these unclear imaging findings of malignancy versus infection, oncology was consulted, and the patient underwent further hematologic malignancy workup including serum and urinary protein electrophoresis of which results are shown in (Table 1). Based on these findings, the patient underwent a bone biopsy that was negative for any malignant process. Orthopedic surgery was involved in the patient's case after the oncology workup was completed and malignancy was excluded as the source of left shoulder imaging findings. Left humeral debridement with bony resection and placement of an antibiotic spacer was performed. Intraoperatively the rotator cuff complex was found to have eroded secondary to infection with pus spontaneously exuding from the entirety of the humeral head. The biopsy of resected bone showed both acute and chronic osteomyelitis with intraoperative cultures growing MRSA. The patient was subsequently started on IV Vancomycin with tentative plans for a 6-week course followed by a drug holiday then joint aspiration, and if no signs of residual infection, then proceed with a total shoulder replacement. The patient was discharged to acute rehab for a several-week stay. Unfortunately, shortly after returning home from rehab, the patient had a fall resulting in hemothorax complicated by hypotension requiring an ICU admission with vasopressor support. As of this writing, no further surgical intervention has been planned due to this recent complication.

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Figures

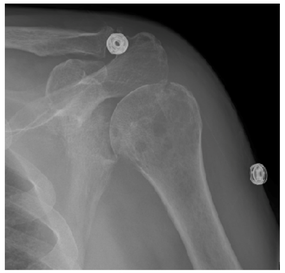

Figure I. X-ray of left shoulder showing cortical irregularity of the greater tuberosity with probable lytic lesions in the humeral head concern for pathologic fracture.

Figure II. Non-Contrast CT of left shoulder showing soft tissue thickening, joint effusion, as well as expansile lucency in the humeral head, neck, and proximal diaphysis with pathological fracture.

Figure III. MRI of left shoulder showing osseous erosions, numerous prominent intraosseous cysts/abscesses with osteomyelitis.

Table 1: Multiple Myeloma Work-Up

Serum Light Chains	8/6/2023	8/11/2023
Free Kappa LC, Quant (3.3-19.4 mg/L)	248.8 H	150.0 H
Free Lambda LC, Quant (5.7-26.3 mg/L)	99.8 H	145.3 H
Free Kappa/Lambda Ratio (0.26-1.65)	2.49 H	1.03

Discussion

Osteomyelitis, while a common disease, can have abnormal presentations, delaying definitive treatment and prolonging hospital stays. Our patient had multiple abnormalities in his clinical presentation. The first is the idiopathic nature of this patient's deep-seated and locally devastating infection. The patient had no known inciting event or devices/hardware in the shoulder and was relatively asymptomatic. The patient's only risk factor was diabetes mellitus; however, he reported no history of diabetic ulcers. The patient also denied any prior history of osteomyelitis. The patient had no other infections complaints, raising the question of where the infection could have seeded from. Due to the lack of inciting event, his infection is best characterized as idiopathic. The patient had no positive blood cultures while inpatient at our hospital. While blood cultures were negative, this is not uncommon with studies showing blood cultures to be positive 40-60% of the time in osteomyelitis.[6] Osteomyelitis of the humeral head is also a rare entity. Interestingly a study showed humeral osteomyelitis cases have a prevalence of 3.4% of their database of osteomyelitis, with *Staph Aureus* being by far the most common infectious agent.[7]

The second aspect is the elevated free kappa and lambda chains found on SPEP. It is uncommon to have elevated free lambda and kappa chains in the setting of infection, with this finding classically being associated with multiple myeloma. To our knowledge, there is little literature regarding this phenomenon, with no known mechanism. The only available report on this topic presented a case that led to the patient being started on cytotoxic medication before biopsy showed the lesions to be infectious in nature.[8] In our case, oncology was brought on board for a concern of multiple myeloma but fortunately the patient's biopsy returned before the patient was started on treatment malignancy. Seeing the patient met no other criteria for multiple myeloma, and his clinical status improved with broad-spectrum antibiotics and diuresis, there is a low index of suspicion for multiple myeloma in this patient.

The patient's SPEP/UPEP values were not the only diagnostic factor that initially led the team to suspect multiple myeloma. The initial left shoulder radiograph revealed probable lytic lesions concerning for pathologic fracture. There are previous reports of chronic osteomyelitis mimicking bone tumors on radiographs.[9] While osteolytic destruction can sometimes be initially seen on plain radiograph in acute or chronic osteomyelitis, there were no other common features seen including: soft tissue swelling, periosteal thickening, sequestrum formation, or osteopenia.[10] Furthermore, CT of the shoulder showed similar findings thought to be more attributable to a primary/metastatic malignancy or myelomatous process. The patient's CT findings did not show evidence of sequestrum, involucrum, or cloaca that are usually seen in chronic osteomyelitis, further pointing towards a malignant rather than infectious etiology. Not until the MRI of the left shoulder was obtained did the etiology of the shoulder lesion become clearer. The MRI demonstrated bone marrow edema throughout the glenoid and entirety of humeral head, one of the most sensitive imaging findings of acute osteomyelitis. Another MRI finding pointing towards osteomyelitis was the presence of multiple, peripherally enhancing, intraosseous abscesses, referred to as Brodie abscesses, that are seen in the subacute phase of osteomyelitis.[10] Lastly, there was no sign of pathologic fracture on MRI to indicate any evidence of osteolytic/myelomatous process. Due to this patient's lack of classic initial imaging findings seen in osteomyelitis, he subsequently required further evaluation with joint aspiration, bone biopsy, and surgical pathologic review to arrive at the correct diagnosis.

Conclusion

This patient presents an interesting diagnostic conundrum as he had to undergo both extensive testing and a prolonged hospital stay due to delays in diagnosis and treatment. Additionally, the patient was followed by an oncology consult service, diverting crucial healthcare resources. While the adverse outcomes he experienced were minimal, in future patients these complications could be avoided altogether with better recognition of this rather rare radiographic and clinical presentations of a common disease.

References

1. Bury DC, Rogers TS, Dickman MM. Osteomyelitis: Diagnosis and Treatment. Am Fam Physician. Oct 1 2021;104(4):395-402.
2. Henke PK, Blackburn SA, Wainess RW, et al. Osteomyelitis of the foot and toe in adults is a surgical disease: conservative management worsens lower extremity salvage. Ann Surg. Jun 2005;241(6):885-92; discussion 892-4. doi:10.1097/01.sla.0000164172.28918.3f
3. Gaillard F, Rasuli B, Hacking C. Osteomyelitis. Reference article. Accessed November 16 2023, 2023. https://radiopaedia.org/articles/7662
4. Dähnert W. Radiology Review Manual. 2011.
5. Schmitt SK. Osteomyelitis. Infect Dis Clin North Am. Jun 2017;31(2):325-338. doi:10.1016/j.idc.2017.01.010
6. Chen WL, Chang WN, Chen YS, et al. Acute community-acquired osteoarticular infections in children: high incidence of concomitant bone and joint involvement. J Microbiol Immunol Infect. Aug 2010;43(4):332-8. doi:10.1016/s1684-1182(10)60051-5
7. Wu H, Yu S, Fu J, et al. Investigating clinical characteristics and prognostic factors in patients with chronic osteomyelitis of humerus. Burns Trauma. 2019;7:34. doi:10.1186/s41038-019-0173-0
8. Malhotra B, Schulz T, Kallail KJ. When Anemia, Atypical Plasma Cells, and a Lytic Bone Lesion are not Myeloma: An unusual presentation of osteomyelitis. Kansas Journal of Medicine. 11/18 2015;8(4):151-152. doi:10.17161/kjm.v8i4.11536
9. Huang PY, Wu PK, Chen CF, et al. Osteomyelitis of the femur mimicking bone tumors: a review of 10 cases. World J Surg Oncol. Oct 22 2013;11:283. doi:10.1186/1477-7819-11-283
10. Desimpel J, Posadzy M, Vanhoenacker F. The Many Faces of Osteomyelitis: A Pictorial Review. J Belg Soc Radiol. May 11 2017;101(1):24. doi:10.5334/jbr-btr.1300