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Case report

Bilateral glenohumeral septic arthritis secondary to mastitis with subsequent avascular necrosis: A case report

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ABSTRACT

Introduction and importance: Septic arthritis is an orthopaedic emergency. Only 3% of septic arthritis patients present with glenohumeral involvement. Polyarticular disease with shoulder involvement constitutes 1% of this group. There is currently no documented case of bilateral glenohumeral septic arthritis with avascular necrosis secondary to mastitis.

Case presentation: We present a case of a 38-year-old African woman with bilateral glenohumeral septic arthritis after management for mastitis of the left breast. She had left, then right shoulder pain, fever, and reduced range of motion, with multiple arthroscopic washouts and antibiotic therapy instituted at various hospitals by various specialists, leading to the resolution of active infection. The patient developed bilateral humeral head avascular necrosis with complete collapse as evidenced on plain radiography 4 months later, and underwent hemi-arthroplasty of the right shoulder. Biopsy at operation showed no active bone infection, and inflammatory markers were not elevated. She had reduced discomfort for the following 2 years but developed increasing pain on lifting and reduced range of motion. The moderate symptoms in her left shoulder managed non-operatively had remained unchanged. There were no elevated inflammatory markers 2-years post arthroplasty.

Clinical discussion: This case presents an uncommon scenario of bilateral septic arthritis secondary to mastitis, with rapid progression to avascular necrosis in an individual with no classical risk factors for avascular necrosis, despite appropriate antibiotic treatment and surgical interventions. Advanced collapse necessitated hemi-arthroplasty of the dominant right side.

Conclusion: This case highlights the need for close radiographic follow-up in atypical presentations of septic arthritis and the dilemmas in managing advanced bilateral avascular necrosis with arthroplasty in a young patient.

1. Introduction

Septic arthritis has an incidence of 2–6/100,000 [1]. It is an orthopaedic emergency, managed with either open or arthroscopic drainage of the joint with a course of culture-sensitive antibiotics. Mortality rates vary from 4 to 42% [2], with values as high as 50% in polyarticular disease [3]. This occurs in patients with comorbidity or the immunocompromised, such as those with sepsis, liver cirrhosis, HIV, Diabetes mellitus, and Intravenous drug abusers [4].

Glenohumeral involvement constitutes only 3% of all septic arthritis cases, with 1% of this group having a polyarticular presentation [5]. The polyarticular septic arthritis group usually has involvement of the knee joint [6], making isolated bilateral glenohumeral disease exceedingly rare. There is scant literature on bilateral glenohumeral involvement and management.

We present a case of bilateral glenohumeral septic arthritis in a young female treated for mastitis, with subsequent bilateral humeral head avascular necrosis necessitating hemiarthroplasty on the right shoulder. The patient was managed in a private practice setting. This case has been reported in line with the SCARE criteria [7].

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2. Case presentation

2.1. Patient information

Our patient is a 38-year-old African female who presented with a three-day history of atraumatic left shoulder pain. She is right-hand dominant.

Past medical history: Left breast mastitis 2 weeks prior managed with oral Flucloxacillin 500 mg QID for 10 days, and resolved one week before the onset of shoulder symptoms. No previous surgery nor prior steroid use or joint injections and was not on any medication. She has no known food or drug allergy, no adverse drug reactions and her family history is unremarkable for hereditary conditions or malignancy. She is a banker, with no previous or current smoking, and takes a glass of wine a week. Her systemic review was unremarkable.

2.2. Clinical and diagnostic assessment

2.2.1. August 2018

Our patient presented with atraumatic left shoulder pain, of gradual onset, present for 3 days, aggravated by lifting light objects, with no fever or chills, but with restricted shoulder motion. She is right-hand dominant. In her past medical history, she had left breast mastitis managed with oral antibiotics and resolved one week before the onset of shoulder symptoms. She had no comorbidities, no prior steroid joint injections, and no smoking, but occasional social alcohol use. She had used a levonorgestrel based intrauterine hormone-releasing contraceptive device before her symptoms. Her systemic review was unremarkable. Her left shoulder was moderately swollen, tender, with limited active and passive range of motion.

She had a fever (39 °C), with other vital signs being unremarkable. Her C-reactive protein was elevated at 58.23 mg/l (normal range 0-5 mg/l), with normal total white cell count and differential. A plain x-ray of the left shoulder was performed (Fig. 1a) showing mild inferior displacement of the humeral head. An MRI was performed (Fig. 1b). This showed cystic lesions in humeral metaphysis and diaphysis with joint effusion (Marrow Edema Syndrome), and prompted aspiration of the joint, yielding a yellowish/brown fluid that was sent to the lab for analysis.

Microscopy, culture and sensitivity results yielded pus cells, but no bacterial growth and no Acid Fast Bacilli. She underwent an arthroscopic washout of the left shoulder by a consultant Orthopaedic surgeon, who was 2 years post-training. It was performed at a SafeCare Level 5 hospital. A diagnosis of septic arthritis and left proximal humerus osteomyelitis was made. She was started on intravenous Flucloxacillin 2 g QID and Gentamicin 150 mg OD for 5 days, and was discharged on oral Clindamycin 450 mg QID for 5 weeks, Paracetamol 1 g QID and Tramadol 100 mg BD for 2 weeks. Her C-reactive protein had risen sharply to 289.05 mg/l, 5 days after admission, but dropped to 210.56 mg/l at discharge, 1-week post-admission.

She was re-admitted 5 days post-discharge, with a 3-day history of severe right shoulder pain, with reduced range of motion. MRI of the right shoulder (Fig. 2), revealed glenohumeral joint effusion, and a collection in the subscapularis fossa, consistent with septic glenohumeral arthritis and osteomyelitis.

Arthroscopic washout of the right shoulder was performed the same day, and she was discharged 5 days later, with continued oral

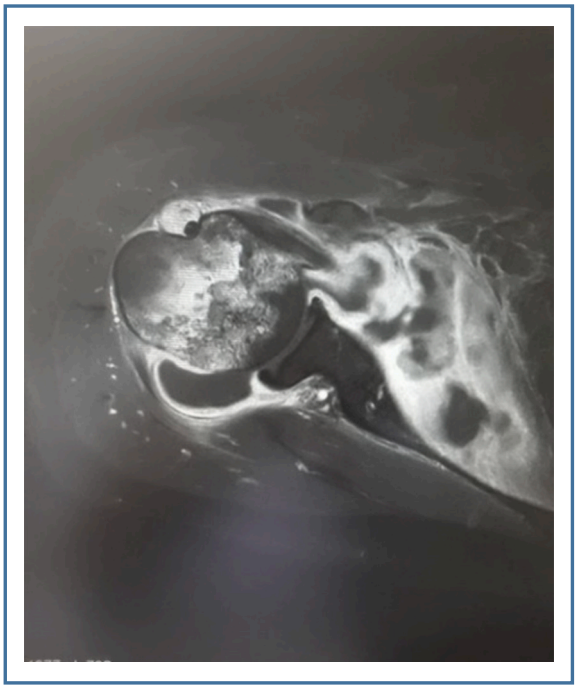

Fig. 2. Axial T1 sequence showing collections (peripherally enhancing) in subscapularis and biceps muscle, consistent with septic arthritis.

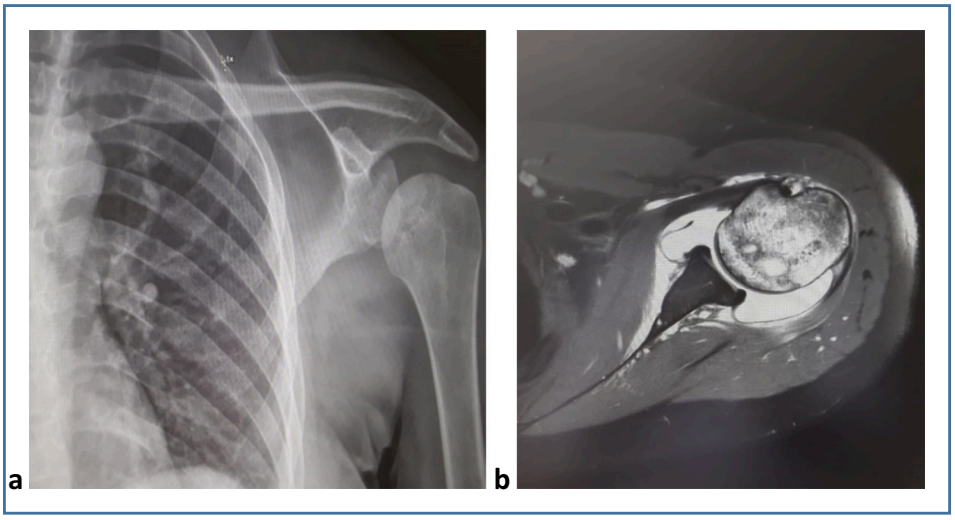

Fig. 1. (a) AP radiograph of the left shoulder with mild inferior displacement of the humeral head, suggestive of joint effusion. (b) A T2 weighted axial cut showing cystic lesions in the humeral metaphysis, an intact cortex, and a large glenohumeral joint effusion.

Clindamycin 500 mg QID for 5 weeks, Ibuprofen 400 mg PO TDS, Dihydroycodeine 60 mg PO BD and Lactulose 10 ml PO BD for 1 week.

Two weeks later (1 month from initial presentation) she experienced purulent discharge from her anterior right shoulder surgical site and was advised on having an urgent arthroscopic washout, to which she declined, preferring self-administered expression with dressing at home. Drainage ceased within a week, and the sinus healed. Three weeks later, she presented with a slow-growing, painless mass in the right armpit. An MRI of both shoulders (Fig. 3) was performed a week later, revealing a right glenohumeral joint effusion and periarticular collections – consistent with right glenohumeral septic arthritis.

Bilateral shoulder arthroscopic washouts were performed, with aspirate specimens taken for microscopy, culture and sensitivity. The specimens grew *Streptococcus* spp. and *E. coli*. She was discharged home 5 days later. On subsequent routine clinic visits, she had limited bilateral shoulder range of motion, impaired activities of daily living, inability to sleep on her sides, but was afebrile and had no discharge from surgical sites on either shoulder.

3. Follow-up and outcomes

She was followed up as an outpatient, and bilateral shoulder x rays were requested 4 months after discharge, showing advanced bilateral humeral head collapse, with sparing of the glenoids (Cruess Stage IV) (Fig. 4). The white cell count was normal, and C-reactive protein had dropped to 3.6 mg/L. Her progression from septic arthritis to advanced bilateral avascular necrosis took 8 months.

Meanwhile, her activities of daily living were getting severely limited. After a long discussion and consultation, she was offered the option of a hemiarthroplasty of her right shoulder, as it was her dominant and more painful one, which was done. Her pre-operative evaluation showed normal C-reactive protein levels (3.2 mg/L) and a Procalcitonin of <0.05 ng/ml (normal 0.0–2.0 ng/ml). A bone biopsy specimen of the right humeral head was analysed to rule out infectious causes for avascular necrosis, including Tuberculosis, showing areas of necrosis and granulation, but no inflammatory cell infiltrates, and no epithelioid granulomas. No metastatic cell lines were identified.

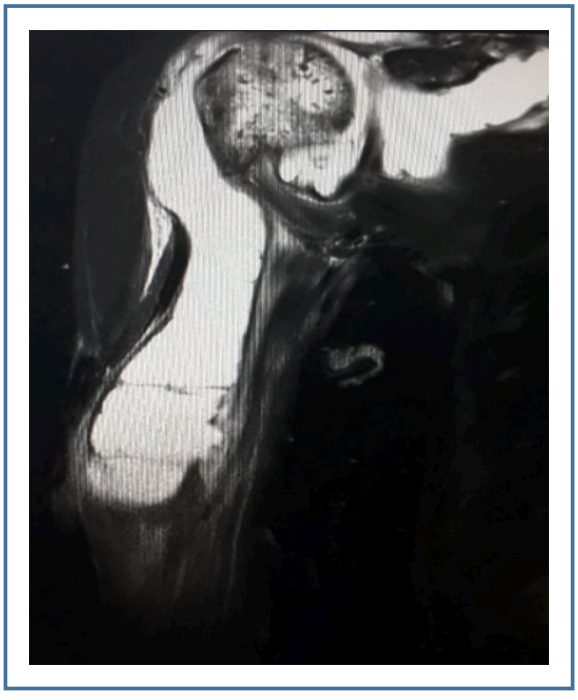

Fig. 3. T2 weighted MRI right shoulder coronal cuts with glenohumeral joint effusion, and peri-articular collections (subscapularis, infraspinatus, supraspinatus) communicating with joint effusion.

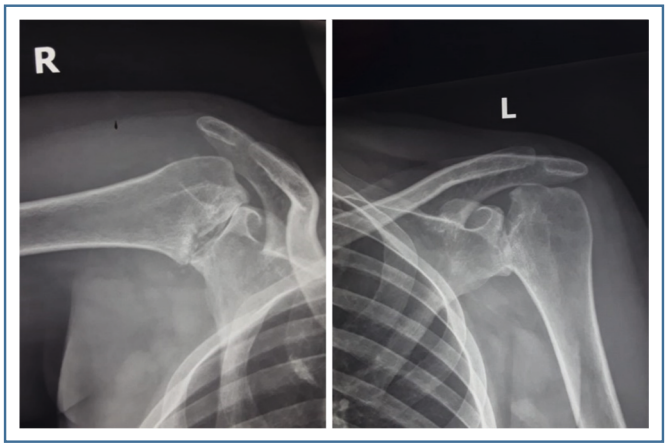

Fig. 4. Antero-posterior plain radiographs showing complete erosion and collapse of the humeral heads bilaterally, with spared glenoids (Cruess Grade IV) (eight months after initial presentation).

Post-op radiographs (Fig. 5) show a right hemiarthroplasty. Fig. 5a. is the immediate post-op radiograph with features suggestive of deltoid inhibition, a classic post-op finding after this procedure. Fig. 5b. was performed 2 years post-op, with no signs of loosening or subluxation. She had a good return of shoulder function after physiotherapy, with increased range of motion, reduced pain, and ability to perform daily overhead activities. She returned to work 3 months post-op and was asymptomatic for the subsequent 2 years. The left shoulder symptoms (non-operatively managed) remained unchanged.

At 2 years post right hemiarthroplasty, she presented with initially intermittent, then constant right shoulder pain of gradual onset, aggravated by carrying moderately heavy loads, with a sense of giving way, and difficulty in performing activities of daily living. Her left shoulder symptoms had not changed (intermittent pain and reduced range of motion). She has no fever or chills, no shoulder swelling, and no discharge from any point on the shoulder.

On examination, her deltoids had subtle wasting, with well-healed surgical scars. There was point tenderness at her right acromial tip and the deltoid insertion. She had bilateral shoulder lateral abduction to 95°, bilateral forward flexion to 95°, and internal rotation on the left to T10, on the right shoulder to L5. There was painful active and resisted range of motion, with relative pain-free passive range of motion. There was good stability on anterior, posterior, and inferior examination. She had no apprehension or relocation, and had good strength on rotator cuff testing – but painful. C-reactive protein levels were not elevated at 1.13 mg/L, with a normal white cell count with differential, and a normal ESR of 11 mm/h.

4. Discussion

Unilateral glenohumeral septic arthritis occurs in 3% of patients, with only 1% having polyarticular disease [5]. Isolated bilateral glenohumeral septic arthritis is exceedingly rare since polyarticular septic arthritis usually includes knee involvement [6]. There are no current guidelines or outcome studies on managing patients with bilateral shoulder septic arthritis [8]. Comorbidities are present in as many as 87% of patients with unilateral glenohumeral involvement [9]. However, other than the preceding case of mastitis with suspected resultant sepsis seeding the joints in this patient, no comorbidities classically associated with this condition and outlined in other studies were present in this patient. A literature search on the significance of her hormonal contraceptive use did not find any associations between levonorgestrel and mastitis or avascular necrosis.

Organisms that seed the joint cause irreversible damage by

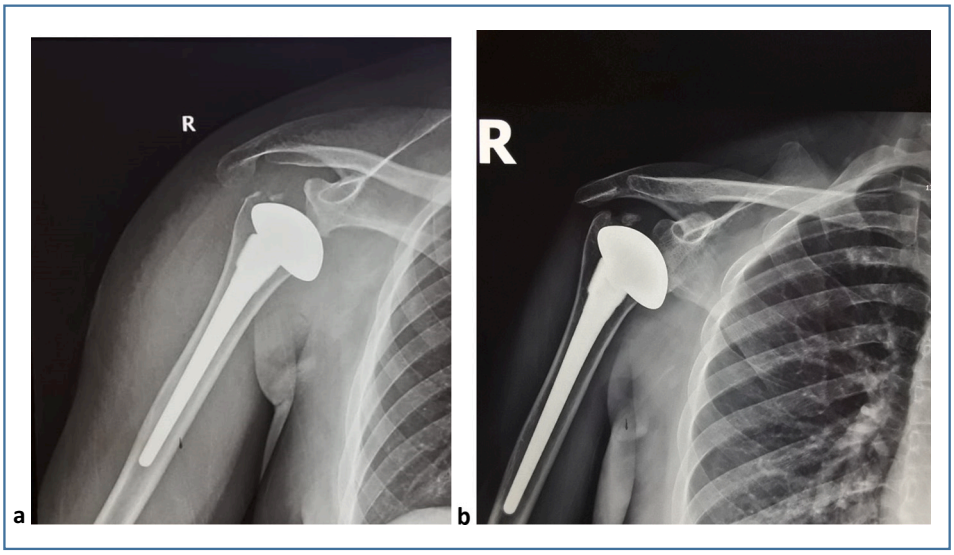

Fig. 5. A right shoulder hemiarthroplasty. a. Immediate post-op. b. 2 years post-op.

activating the release of proteolytic enzymes from activated inflammatory cells [1]. An exudative build-up in the joint space has a tamponade effect on surrounding vessels with resultant ischemia of the synovium and eventual anoxic damage to joint cartilage [1]. However, literature drawing a direct link between septic arthritis and avascular necrosis of the shoulder is scant. Humeral head trauma, long term corticosteroid use, radiation, chemotherapy, chronic heavy alcohol use, Caisson's disease, and sickle cell hemoglobinopathies are the leading causes of atraumatic avascular necrosis of the humeral head [10].

Intervals between pain onset and humeral head collapse vary, ranging from 6 months in sickle cell disease [11], to 10 years in long term corticosteroid use [12]. The interval in our patient between resolution of active infection and avascular necrosis was 8 months. Osteonecrosis as an indication accounts for only 5% of shoulder arthroplasty cases [13]. The challenge is the younger age at which patients with atraumatic osteonecrosis present, given the limited lifespan of the prosthesis, and a need for later revision [13]. Patients presenting with late-stage disease (Cruess IV and V), invariably have progression, necessitating arthroplasty [14]. Hemiarthroplasty is employed in Cruess Stage IV disease [10], with total shoulder arthroplasty reserved for stage V, in which glenoid involvement is present [15].

This case report's main strength is its elaboration of the need for close radiological surveillance for signs of avascular necrosis in patients with atypical septic arthritis. There is currently no documented case of bilateral glenohumeral septic arthritis with avascular necrosis secondary to mastitis. Its weakness lies in some of the delays in imaging and surgical interventions, which were outside the control of the attending orthopaedic surgeons. This being an unusual presentation, a few questions arise. Normally, septic arthritis would result in joint destruction of both joint surfaces. Why were the glenoid surfaces spared in this instance? Furthermore, it is unclear why neither the hip nor the knee was involved, as is typical in septic polyarthritis. Her presentation has no clear explanation at the moment. Despite this, we continue to follow her up.

5. Conclusion

The patient manifested a rare case of bilateral glenohumeral septic arthritis post left breast mastitis, a pattern of infection not described in the literature so far. Furthermore, the rapid progression to bilateral avascular necrosis with advanced collapse is notable, given the absence of classic predisposing conditions. This case highlights the need for close radiographic follow-up in atypical presentations of septic arthritis.

6. Patient perspectives

The patient expresses concern for her current difficulty in performing activities of daily living due to bilateral shoulder pain. She is open to operative management to resolve her right shoulder symptoms.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

The patient has provided both verbal and written consent for the publication of this article and was involved at every stage of this case report. It was made sure that her identity will be kept a secret at all levels.

If there are any questions or concerns regarding the ethics of this patient, please contact Dr. Vincent Mutiso at mutiso@uonbi.ac.ke.

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Guarantor

Vincent Mutiso.

Research registration number

Not applicable.

CRediT authorship contribution statement

Both authors were involved in the researching, writing, and editing of the manuscript.

Samora Maranya: Writing - Original draft preparation**. Samora Maranya:** Visualization. **Vincent Mutiso:** Supervision and approval of the final version. **Vincent Mutiso:** Writing - Reviewing and editing.

All authors read and approved the final version to be published.

Declaration of competing interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

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