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Bilaterally Symmetrical Serratia Osteomyelitis of Distal Third Radius in an Adolescent Female with Sickle Cell Disease: A Rare Case Report

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Learning Point of the Article:

We present a case with bilaterally symmetrical osteomyelitis due to Serratia marcescens which being a non-pathogenic organism is an extremely rare cause of osteoarticular infections; however, in immune-compromised patients such as those with sickle cell disease, we should always keep such non-pathogenic organisms and unusual presentations in mind.

Abstract

Introduction: Osteomyelitis as a condition in adolescents is difficult to diagnose as it is. In patients with sickle cell disease, the diagnosis is even more difficult due to the occurrence of Vaso occlusive crisis which being the most common acute clinical manifestation of sickle cell disease in bone may mimic osteomyelitis. No single clinical presentation, laboratory result, or imaging finding can definitively distinguish these diagnoses.

Case Report: A 15-year-old female presented to the outpatient department with mild pain and swelling in both forearms persisting for the past 3 months. Radiographs revealed predominantly diaphyseal expansile osteolytic lesions in the bilateral distal third radius with periosteal reaction along with a small radiolucent area in the right distal radius. She was subsequently admitted for further assessment. On the 3rd day of admission, pus pointing was noted on the volar aspect of the right forearm while the left side only had bony swelling. Surgical intervention (debridement with saucerization) was performed on both sides. Pus culture (collected intraoperatively) showed Serratia marcescens as the causative organism. Culture-sensitive antibiotics were administered intravenously for 3 weeks followed by oral antibiotics for 3 weeks. At 1-year follow-up near complete resolution of bony lesions with full range of motion in both upper limbs was noted.

Conclusion: Subacute osteomyelitis may present with just bony swelling without significant pain or fever and can present a diagnostic dilemma, a high index of suspicion is important to rule out other conditions and diagnose osteomyelitis in such cases. Unusual organisms like Serratia can cause osteomyelitis in immunodeficient patients like those with sickle cell disease. Appropriate management in the form of thorough surgical debridement, saucerization, and appropriate antibiotics can lead to complete resolution of the infection with good clinical results.

Keywords: Osteomyelitis, sickle cell disease, bilateral, serratia marcescens.

Introduction

Sickle cell disease is an autosomal recessive hemoglobinopathy causing chronic hemolytic anemia [1-3]. Osteomyelitis has a higher propensity in patients with sickle cell disease due to impaired immune function and functional asplenia [4-6]. Osteomyelitis usually presents in early childhood and is relatively rare in adolescents. It usually presents with acute pain, fever, and swelling with or without discharge. Patients with sickle cell disease suffer from repeated episodes of Vaso occlusive crisis which can mimic and mask the symptoms of osteomyelitis. Multifocal osteomyelitis has been reported in patients with sickle cell disease; however, bilaterally symmetrical involvement in sickle cell disease has not been reported to the best of our knowledge, unlike the general population where Staph. Aureus is

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Author's Photo Gallery


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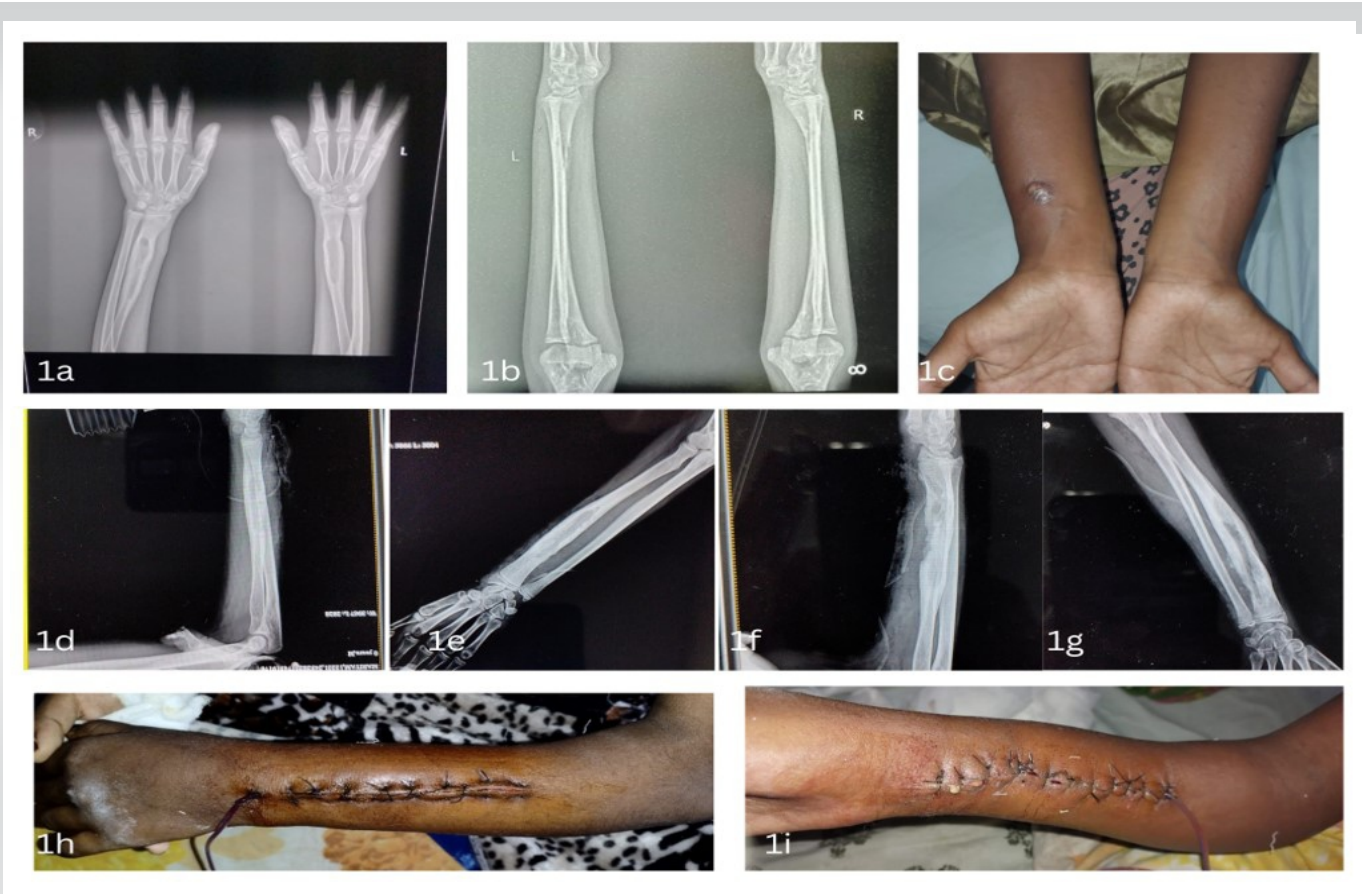

Figure 1: (A and B) Pre-operative radiographs, (C) Rt-sided Pus pointing, (D and E) post-operative radiograph of left side, (F and G) post-operative radiograph of right side, (H) immediate post-operative wound of left side and (I) shows immediate post-operative wound of right side).

the most common organism, in patients with sickle cell disease Gram-negative bacilli like Salmonella are more frequently isolated in patients with osteomyelitis. We are reporting the unusual case of a 15-year-old female, a known case of sickle cell disease who presented with bilaterally symmetrical swelling over the distal third forearm which turned out to be osteomyelitis due to a rare pathogen like Serratia marcescens.

Case Report

We present a case of a 15-year-old female child who presented in the Outpatient department (OPD) with minimal pain and swelling over bilateral distal forearms for 3 months. The swelling was insidious in onset, gradually progressive and it was associated with mild tenderness over both forearms. The pain was minimal and did not hinder any daily activity. The patient was a known case of sickle cell disease SS pattern with a history of hospital admission for cholecystectomy around 18 months back. The patient had no history of fever. The patient had an average build and was well nourished. On examination, she had bilateral swelling and mild tenderness over the distal third radius. There was no rise in local temperature, erythema. There was no axillary lymphadenopathy. The range of motion of both wrists, fingers, and elbows was normal. X-rays revealed predominantly diaphyseal expansile osteolytic lesions in bilateral distal third radius with periosteal reaction and a small radiolucent area was seen on the right distal radius. Subsequently on the 3rd day of admission, the patient developed pus pointing on the right forearm volar aspect which confirmed our clinical suspicion of osteomyelitis. We then decided to go for surgical intervention bilaterally in the form of thorough surgical debridement with saucerization. As the pus pointing was on the volar aspect on the right side, we utilized a volar approach for the right forearm and on the left side, we went through the dorsal aspect of the forearm (Fig. 1). We collected intraoperative pus culture samples from both sides and sent them for culture and antibiotic sensitivity testing (AST). The Culture and AST report from both sides showed the presence of Serratia Marcescens which was sensitive to Tigecycline, Imipenem, Meropenem, Piperacillin/tazobactam, and Cefoperazone/Sulbactam. The culture plate and microscopy slide are shown in Fig. 2 and 3. After consultation with an infectious disease specialist, the patient was given 3 weeks of IV

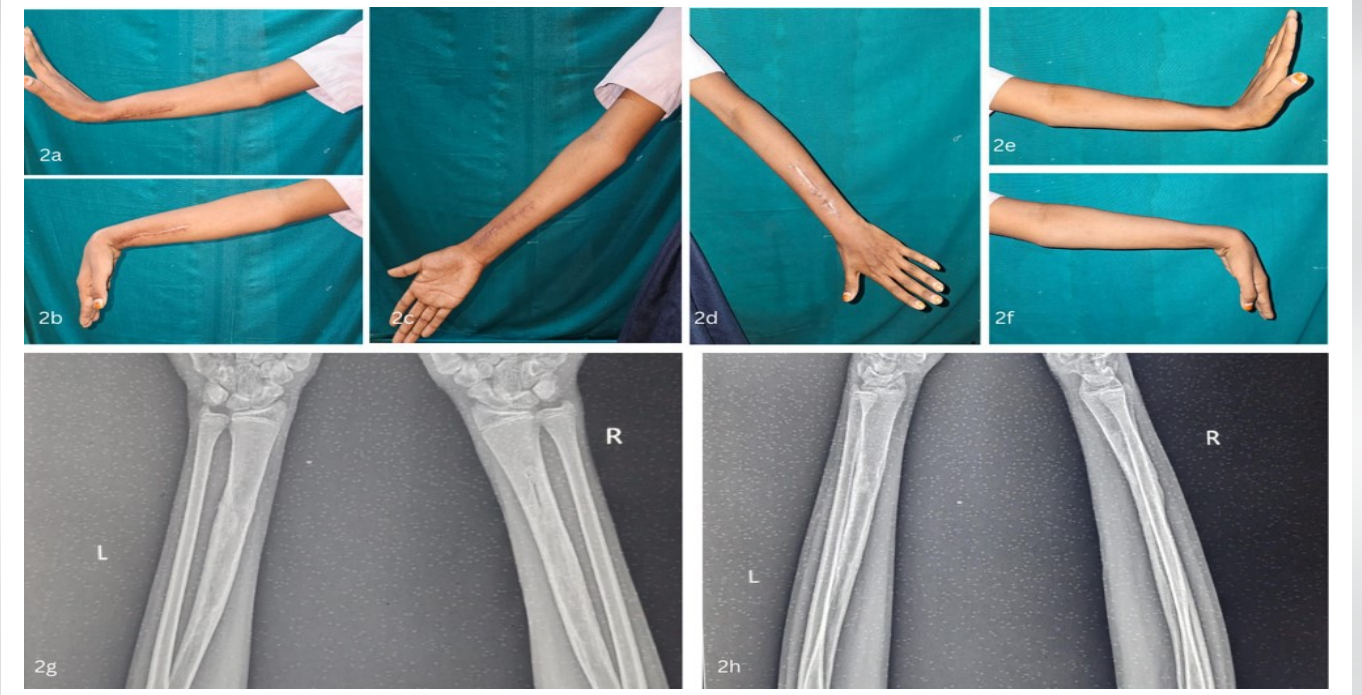

Figure 2: At 1-year follow-up. (A and B) Range of motion of right wrist while 2C shows well healed scar of the right side. 2D shows a well healed scar of the left side while 2E and 2F show range of motion of the left wrist. 2G and 2H show 1-year follow-up radiographs of both forearms in anteroposterior and lateral views, respectively

antibiotics (Piperacillin/tazobactam) and then 3 weeks of oral antibiotics (Faropenem) postoperatively. The patient was followed up in OPD at 15 days for suture removal, at 6 weeks and every 3 months thereafter. The latest follow-up (1 year) shows well-healed scars of bilateral forearms with no swelling or tenderness. On 1-year follow-up radiographs, we can see the resolution of the lytic lesions with remodeling to almost near normal appearance of the bone along with a complete range of motion of both the fingers, wrists, and elbows (Fig. 2).

Discussion

Multiple studies have demonstrated that patients with sickle cell disease are more susceptible to infection. The various mechanisms responsible for this are the presence of infarcted bone due to vaso-occlusion, impaired immune response due to functional hyposplenism and deficient activity of alternative pathways of the complement system [7-9].

Although acute osteomyelitis is generally associated with constitutional symptoms like fever, severe pain, and pseudo-paralysis, subacute osteomyelitis may present with bony swelling with minimal pain as seen in this patient which can present a diagnostic dilemma.

Unlike the general population where staphylococcus aureus is the most common organism associated with osteomyelitis, Gram-negative bacilli like salmonella are more commonly associated with osteomyelitis in sickle cell disease patients [10]. It is postulated that patchy ischemic infarcts of the bowel which occur due to vaso-occlusive episodes can lead to mucosal barrier breakdown through which the enteric Gram-negative bacteria can enter into the bloodstream and get lodged into the avascular bone causing osteomyelitis [11].

In our patient, culture revealed the causative organism to be Serratia Marsecens which is an extremely rare cause of osteomyelitis with very few cases reported in the literature. It is usually considered opportunistic and seen mostly in nosocomial infections. However, our patient gave no history of multiple hospital admissions.

Fonk et al. reported Serratia as a cause of osteomyelitis in a 20-year-old male with sickle cell disease. In his case report, he mentioned that the patient had a history of multiple admissions to the hospital for left thigh pain which was considered to be due to vaso-occlusive crisis. On radiographs, changes of osteomyelitis were observed and bone biopsy when cultured revealed the causative organism to be Serratia marcescens. The patient was managed with Intravenous gentamicin therapy at 4 mg/kg/day in three divided doses along with carbenicillin 5 g every 4 h for 6 weeks [12].

Nelms et al. described a case of a 38-day-old infant with Serratia

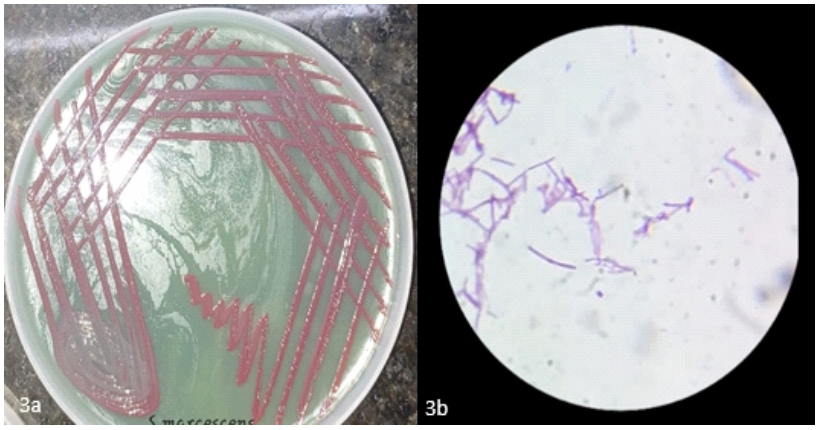

Figure 3: (A) a culture plate of serratia marcescens; (B) microscopy images of Gram-negative bacilli indicating the Possibility of Serratia Marcescens.

osteomyelitis affecting all digits of the upper and lower limb. In this case, the primary differential was salmonella dactylitis as seen in sickle cell disease but the infant was tested negative for sickle cell disease. According to antibiotic sensitivity, testing treatment was started with parenteral chloramphenicol at 100 mg per kg/day and kanamycin at 40 mg per kg/day for a total period of 6 weeks [13].

Hadid et al. presented a case of a 72-year-old who presented with worsening right ankle pain which progressed to tense swelling and inability to bear weight on the ankle. He had a past medical history of uncontrolled type 2 diabetes and coronary artery disease. Surgical history showed 9-year-old distal fibula open fractures managed with ORIF. Occupation required barefoot walking for prolonged periods. He was managed with extensive serial debridement of the ankle joint, implant removal, and antibiotic cement bead placement which were removed at 4 weeks. Pus cultures showed Serratia marcescens. Intravenous antibiotic therapy with Ertapenem was given for a total duration of 5 weeks [14].

Lau et al. reported a case of a 51-year-old man with acute onset back pain, chills, and fever. The patient was a known case of psoriasis and hepatitis c. On MRI, osteomyelitis and discitis at c3-c4 with prevertebral abscesses at c1-c6 and l4-s1 along with bilateral psoas abscesses. Blood culture showed Serratia marcescens. The patient was managed with intravenous meropenem 2 g 3 times a day for 6 weeks followed by 6 weeks of oral ciprofloxacin 750 mg twice a day [15].

Multifocal osteomyelitis has been reported in sickle cell disease patients [16] but to the best of our knowledge, bilaterally symmetrical osteomyelitis as seen in our patient has only been reported once.

Burgner et al. described the case of a 24-year-old male who was a long-term intravenous drug abuser and had an episode of acute hepatitis 4 months prior who presented with pain in the right knee joint. On radiographs, they report symmetrical involvement of both the distal femora and the right proximal tibia which was managed with curettage and long-term antibiotic therapy duration of which has not been mentioned in the study. In contrast to this, our patient had no other associated risk factor other than sickle cell disease and had no acute phase symptoms like pain or fever [17].

Considering the age and clinical history of the patient, fibrous dysplasia was a differential diagnosis, but it was ruled out as polyostotic fibrous dysplasia commonly presents with unilateral bone involvement and café-au-lait spots are associated with 35% of cases [18]. Another possibility considered was that of bone infarcts at an early stage but the localized periosteal reaction seen in our patient made us rule out this possibility. Metastatic histiocytic lymphoma can rarely present with bony swelling, but this is usually seen late in the course of the disease and thus was ruled out [19].

Conclusion

This case demonstrates a rare presentation of bilaterally symmetrical osteomyelitis in an adolescent with sickle cell disease. Subacute osteomyelitis may present with just bony swelling without significant pain or fever and can present a diagnostic dilemma, a high index of suspicion is important to rule out other conditions and diagnose osteomyelitis in such cases.

Unusual non-pathogenic organisms like Serratia can cause osteomyelitis in immunodeficient patients like those with sickle cell disease. Appropriate management in the form of thorough surgical debridement, saucerization, and appropriate antibiotics can lead to complete resolution of the infection with good clinical results.

Clinical Message

With the help of this article, we intend to showcase the possibility of such an unusual presentation of osteomyelitis which to our knowledge has not been reported in literature elsewhere. It is important to keep a high index of suspicion for osteomyelitis in immunocompromised patients like those with sickle cell disease.

It is also important to keep in mind that nonpathogenic organisms can also cause osteoarticular infections in sickle cell patients and these should be treated with appropriate culture sensitive antibiotics for required duration.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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