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Case Report

Emphysematous osteomyelitis with acute promyelocytic leukemia: a rare case report

B. Vijay Kumar, G. Vamshi Nandan Rao*, M. Veena

Department of General Medicine, Yashoda Hospitals, Secunderabad, Telangana, India

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***Correspondence:**
Dr. G. Vamshi Nandan Rao,
E-mail: gvnandan2@gmail.com

ABSTRACT

Emphysematous osteomyelitis represents an infrequent clinical entity. The confluence of acute promyelocytic leukemia (APML), immunosuppression, and an elevated susceptibility to opportunistic infections—exacerbated by bacteraemia-may precipitate the pathogenesis of this gas-forming osteomyelitis process. A 63-year elder male presented to our hospital with consistent fever and chills from past one month. Hematological evaluation revealed pancytopenia. Subsequent bone marrow aspiration and flow cytometry confirmed a diagnosis of acute myeloid leukemia (AML), further substantiated by reverse transcriptase-PCR demonstrating a PML/RARA bcr-3 fusion transcript, confirming the diagnosis of acute promyelocytic leukemia (APML). The patient received a contemporary chemotherapy that included all-trans retinoic acid (ATRA) and arsenic trioxide (ATO), leading to a notable enhancement in the results. However, during treatment, he developed bacteraemia with Klebsiella pneumoniae, confirmed via blood and urine cultures. A high-resolution computed tomography (CT) scan unveiled the pathognomonic presence of intraosseous gas, confirming emphysematous osteomyelitis. Selected intravenous antibiotic therapy yielded in marked clinical amelioration, thus obviating the requirement for surgical debridement. This case underscores the imperative role of advanced imaging modalities in detecting rare infectious sequelae and demonstrates that, with prompt recognition and appropriate antimicrobial intervention, conservative management can yield favorable outcomes even in severely immunosuppressed hosts.

Keywords: Emphysematous osteomyelitis, Acute promyelocytic leukemia, *Klebsiella pneumoniae*, Antibiotic therapy

INTRODUCTION

Acute myeloid leukemia (AML) is a distinguishable malignancy noted for the abnormal maturation and clonal proliferation of myeloid precursor cells.[1] Acute promyelocytic leukemia (APML), constituting approximately 5–8% of AML cases, is typified by the malignant expansion of promyelocytic-stage myeloid precursors within the bone marrow.[2] The identification of gas in the extra-axial skeleton, when other causes such as trauma or surgical procedures are excluded, increasingly indicates EO, particularly in cases involving infections with gas-producing microorganisms.[3] Reported cases remain exceedingly scarce, with frequent predilection for the pelvis, femur, tibia, and thoracolumbar vertebrae. Due to its potentially lethal progression, expeditious radiologic identification and empiric antimicrobial therapy are imperative.[4] Given the paucity of comparative data between conservative (medical) and surgical modalities, an interdisciplinary strategy is advocated. We delineate the clinical course of a 63-year-old male with *Klebsiella pneumoniae*–induced EO managed exclusively with antimicrobial therapy, circumventing surgical debridement, thereby underscoring the therapeutic potential of early medical intervention.

CASE REPORT

A 62-year male patient with symptoms like decreased appetite, constipation, weakness, increased fatigue and

fever with chills over the past month had pancytopenia, hypertension, positive for HBsAg and has previously undergone surgery for anal fistula. Additionally, he had a history of road traffic accident 20 decades ago. Upon arrival, the patient underwent evaluation and was diagnosed with pancytopenia. A bone marrow aspiration was conducted, which revealed aspirate smears showing a predominance of blasts, accounting for approximately 55-60% of the differential count. These blasts were 2-3 times larger than small mature lymphocytes, displaying fine chromatin, prominent nucleolus and scant to moderate amount of pale blue cytoplasm. These observations are suggestive of acute leukemia.

Flow cytometry showed positivity for CD34, CD58, CD33, CD117 and CD13 while showing negativity for CD64, CD14, HLADR, CD2, CD3, CD4, CD8, CD7, CD56, CD10, CD19 and CD20 confirming diagnosis of acute myeloid leukemia. Following this, a PCR-PML/RARA quantitative test was performed using EDTA blood, which yielded a PML/RARA quantitative result of bcr 3-NCN:1.96 (Detected -773 copies). All findings strongly suggested acute promyelocytic leukemia. A consultation with a medical oncologist was sought, and treatment was initiated with a combination of ATRA and ATO, that has significantly improved the prognosis of the patient.

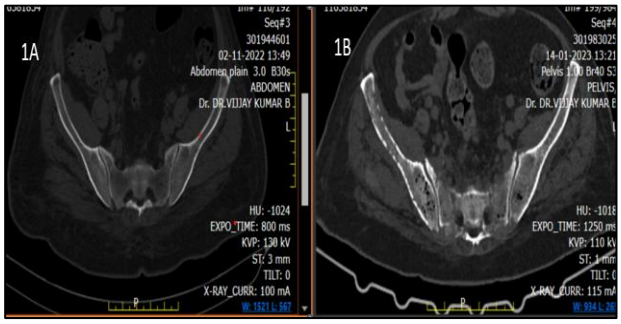

Figure 1: (A, B) MRI image showing gluteal collection.

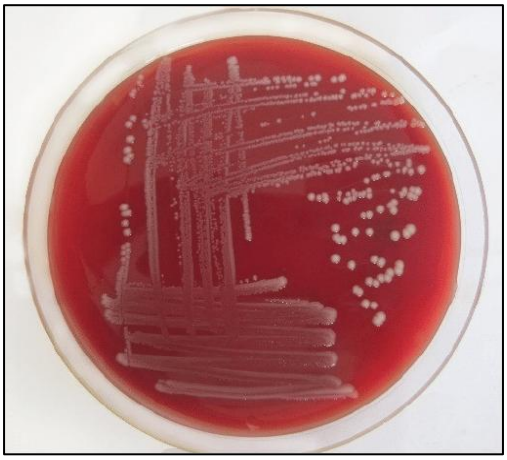

Figure 2: *Klebsiella pneumoniae* growing on blood culture.

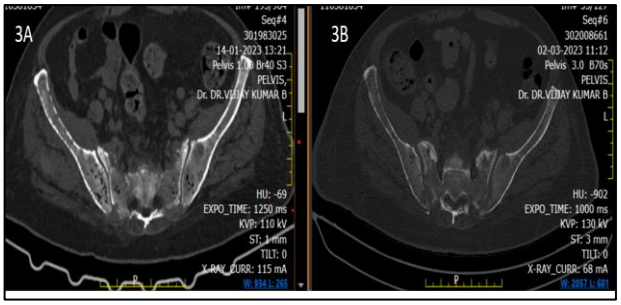

Figure 3 (A, B): MRI images showing steady improvement in patient.

On follow-up examination, patient reported with fever accompanied by chills, severe lower backache and difficulty in walking due to pain. He was evaluated for the complaints. Necessary blood investigations and imaging were done. Treatment was initiated with intravenous antibiotics Magnex forte (IV) and Dalacin (IV). Blood tests indicated negative for malaria, CRP level 22.3 mg/l, haemoglobin 8.6 g/dl, TLC 11,800 and platelet count of 60,000. The MRI showed a right gluteal collection (Figure 1A), where a pigtail drain was placed.

Blood culture, urine culture and aspirated fluid were reported as XDR *Klebsiella pneumoniae* (Figure 2) subsequently CARBA -R was done. The CT scan pelvis indicated emphysematous osteomyelitis (Figure 1B), resulting in the commencement of treatment with Zavicefta 2.5 gm (IV, three times daily) and Aztreonam 2 gm (IV, three times daily) for a duration of eight weeks simultaneously.

Patient has symptomatic improvement (Figure 3A and B) after receiving treatment with antibiotics and analgesics, maintains hemodynamic stability and has not required surgical intervention resulting in discharge. In the final follow-up, blood tests indicated a haemoglobin 10.8 g/dl, TLC 5560 cells/µl, platelet count 2.12 lakhs and C-reactive protein level 3.9 mg/l. Additionally, the patient has completely recovered from APML and has not encountered any sepsis.

DISCUSSION

Acute promyelocytic leukemia (APML) is specifically identified through cytogenetic analysis, which reveals the balanced reciprocal translocation (15;17) and the resultant fusion of the PML and RARA genes. Numerous case studies concerning PML/RARA transcripts are frequently documented in adult populations.[5]

In order to diagnose APML using a PML-RARA quantitative test, it is essential to first obtain a sample of either blood or bone marrow. Subsequently, a real-time quantitative polymerase chain reaction (RQ-PCR) is conducted to quantify the PML-RARA fusion transcript within the sample. The detection of the PML-RARA transcript at levels exceeding a specific threshold indicates

the likelihood of APML. Furthermore, the identification of the bcr3 isoform, which arises from a break in intron 3 of PML and exon 3 of RARA, serves as a specific confirmation of APML.[6] In our case, we confirmed the presence of APML by the PML-RARA transcript with the detection of bcr3 isoform.

The advent of all-trans retinoic acid (ATRA), which interacts with RARα to promote the differentiation of blasts, has transformed the treatment landscape for APML, rendering it a highly treatable condition. Arsenic trioxide (ATO) targets and degrades the PML-RARα fusion protein, working in concert with ATRA; current cure rates with the ATRA and ATO combination now surpass 90%.[7] In our case, we also employed the ATRA and ATO combination chemotherapy, and our patient exhibited a favorable response consistent with the documented success rates. Hematogenous circulation is the prime pathway for the develop of infection in emphysematous osteomyelitis.[8]

Nevertheless, McDonnell et al, have documented infrequent methods of disease transmission, including the extension from intra-abdominal infections, complications arising from intra-abdominal or spinal surgeries, or infections originating from the skin or soft tissues.[9] The appearance of intraosseous gas within the intravertebral space is usually indicatory of disc degeneration; even if, noteworthy intraosseous gas, together with bone marrow edema and surrounding fluid collections, should prompt consideration of emphysematous osteomyelitis .[10] Similar findings that exhibited intraosseous gas accompanied by adjacent fluid collection were observed in our case. Infections are usually monomicrobial or polymicrobial, primarily involving anaerobes or members of the Enterobacteriaceae family *Klebsiella pneumonia* which was consistent with our case.[11]

Confirmation of EO of the spine is achieved through a CT scan, which can detect minimal intraosseous gas within the vertebrae that might not be easily seen on standard radiographs.[12] The use of CT imaging to identify emphysematous osteomyelitis in the spine revealed that majority of cases (86.22%) primarily affected the lumbosacral region, with some cases extending into the pelvic bones and thoracic areas.[13] CT imaging, as corroborated by extant literature, was similarly utilized in our case, yielded definitive diagnostic precision for EO.

Precise confirmation and the prompt initiation of antibiotic treatment are required for the proper management of EO. While targeted therapy is ideal, empirical antibiotic treatment was initiated in most of the documented cases. Intravenous antibiotic therapy was commenced in all reported instances, except for those lacking specific details regarding the antibiotic regimen. Commonly reported antimicrobials include penicillin's, intravenous clindamycin and cephalosporins.[14,15] In the current case, Zavicefta and Aztreonam administered intravenously for a duration of eight weeks in view of XDR Klebsiella pneumoniae after which our patient attained hemodynamic stability, reduction in emphysematous osteomyelitis and was subsequently discharged.

CONCLUSION

Potentially fatal outcomes of emphysematous osteomyelitis can be prevented through early diagnosis and timely intervention with suitable antibiotics and surgical procedures when required. Here we used antibiotics and our patient exhibited a positive response without the necessity of surgery.

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