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A Rare Case Report of Emphysematous Osteomyelitis of the Femur in a Pediatric Patient: Imaging Findings and Review of the Literature

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Abstract **Case Report**

Emphysematous osteomyelitis (EO) is a rare yet life-threatening condition that must be considered in patients presenting with intraosseous gas on imaging. Anaerobic bacteria or members of the Enterobacteriaceae family are typically implicated in EO. The presence of significant comorbidities, such as malignancy and diabetes mellitus, can increase the risk of developing this condition, and high mortality rates have been reported. Prompt diagnosis and management are critical for improving patient outcomes, and radiologists play a vital role in identifying and interpreting imaging findings suggestive of EO.in this article we report the exceptional case of emphysematous osteomyelitis of the femur in a 12-year-old girl with no prior medical history. Microbiological analysis of the collected samples revealed the presence of Staphylococcus aureus.

Keywords: Emphysematous Osteomyelitis, Femur, Pumice stone sign, Radiological diagnosis.

INTRODUCTION

Emphysematous osteomyelitis (EO) is a rare, aggressive, and potentially fatal variant of osteomyelitis; it is an uncommon yet serious bone infection characterized by the presence of gas within the bone. EO should always be considered as a potential diagnosis when intraosseous gas is identified on imaging. This condition is typically caused by anaerobic bacteria or members of the Enterobacteriaceae family, which have the ability to produce gas within infected tissues, it is often associated with significant comorbidities, such as malignancy and diabetes mellitus, which can lead to impaired immune function and increase the risk of developing this condition. It can lead to severe complications, such as sepsis and bone necrosis, and high mortality rates have been reported in severe cases [1].

The diagnosis of EO can be challenging, as the clinical presentation can be nonspecific and imaging findings may be subtle. However, radiologists play a crucial role in identifying and interpreting imaging features suggestive of EO, as this can facilitate early diagnosis and expedite management. Computed tomography (CT) is considered the gold standard imaging modality for the detection of intraosseous gas, as it has a high sensitivity and specificity for this finding. MRI can also be useful in the diagnosis of EO, as it can provide information on the extent of bone and soft tissue involvement [2].

CASE REPORT

We report the case of a 12-year-old girl, with no particular medical history, who was admitted to the pediatric emergency department for a painful swollen left thigh that had been evolving for 12 days. It was preceded by bilateral knee pain, which was followed a week later by respiratory distress associated with abdominal distension but without any other digestive disorders, all of which developed in a context of fever and deterioration of the general condition. Upon clinical examination, the patient was found to be conscious, tachycardic, tachypneic, and febrile at 39 C°. The examination of the thigh revealed a swollen, painful, immobile, and warm left thigh, especially in its anteromedial part and in the inguinal region. Laboratory investigations were performed and showed very high levels of white blood cells at 30,000 predominantly neutrophilic and a C reactive protein level of 349. An

ultrasound of the soft tissues of the swelling revealed an ill-defined, hypoechoic, and heterogeneous collection containing air bubbles, an enhanced CT scan, revealed the presence of multiple deep collections in the left thigh that extended to the homolateral iliopsoas muscle. Furthermore, there was evidence of a joint effusion in the left hip that was subluxated, along with the presence of intraosseous air bubbles at the level of the left upper femoral metaphysis that were associated with a trochanteric detachment Figure 1. In addition, blood cultures were obtained, followed by subsequent aspiration of the soft tissue and the obtained samples exhibited a polymicrobial infection with an abundant amount of gram-positive cocci, wherein Staphylococcus aureus was identified as the predominant organism upon culturing.

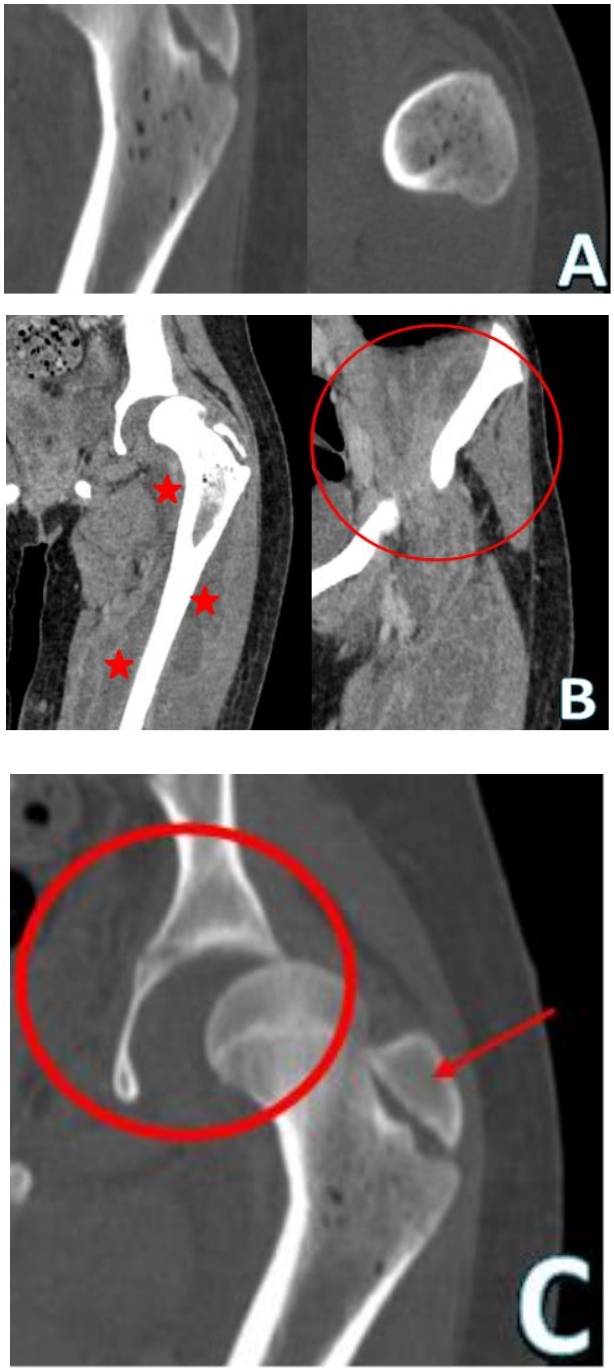

Figure 1: Axial and coronal CT scan zoomed images of the left femur bone demonstrate multiple clusters of intramedullary gas differing in morphology and size (irregularly irregular) and located in the left upper femoral metaphysis (point measurements of CT density averaged - 460 Hounsfield units) (A) the associated adjacent multiple deep collections in the left thigh (stars) that extended to the homolateral iliopsoas muscle (circle).(B) Note the trochanteric detachment (arrow) and joint effusion in the left hip that was subluxated (circle). (C)

Figure 2: Full field view of a pumice stone (A) and Illustration of irregular air locules as seen in emphysematous osteomyelitis, Note the irregular black air locules within the partially intact (white) trabeculations (B)

DISCUSSION

Emphysematous osteomyelitis is a rare but severe condition that has been documented in the literature in only a limited number of cases since its initial description by Ram and colleagues in 1981. The prevalence of this condition has remained low, with reported cases predominantly occurring in various anatomical sites such as thoracic and lumbar vertebrae, sacral bones, femur, pelvis, tibia, or fibula [3].

Emphysematous osteomyelitis is often associated with underlying comorbidities that compromise the immune system, including diabetes mellitus and malignancy. Hematogenous dissemination is the most common mode of infection, although cases of emphysematous osteomyelitis have also been reported as a result of intra-abdominal infections, spinal surgery, or soft tissue or skin infections. In the current case, a soft tissue infection was the likely source of infection, which progressed to necrotizing fasciitis and subsequently emphysematous osteomyelitis. Causative organisms are typically both aerobic and anaerobic bacteria, with anaerobes or members of the Enterobacteriaceae family most commonly identified in mono or polymicrobial reported cases. In this case, a Gram stain indicated a polymicrobial infection with abundant gram-positive cocci, and the culture showed Staphylococcus aureus as the predominant organism [4, 5].

The diagnosis of emphysematous osteomyelitis poses a challenge to clinicians due to the absence of specific clinical manifestations and subtle imaging characteristics. Nonetheless, radiological evaluation is pivotal in determining the nature and magnitude of the infection in affected individuals. The imagery features are typified by the presence of intraosseous gas Figure 2A, also known as the "pumice stone sign" Figure 2B, accompanied by additional indicators of bony and soft tissue infection [2]. The detection of gas locules within the bone and adjacent soft tissues on imaging is pathognomonic for emphysematous osteomyelitis. While these findings can be visible on plain radiographs, CT scan is an excellent modality for identifying intraosseous gas [6, 7]. In the present case,

the CT scan was instrumental in confirming the diagnosis by revealing subcutaneous and deep air, evidence of bone destruction, and air within the femur.

In the field of radiology, the presence of intraosseous gas can be indicative of various pathological conditions such as osteonecrosis, bone malignancy, post biopsy, penetrating wounds, compound fractures, and lymphangiomatosis of the bone. It is important to note that gas can accumulate in any part of the body where there is a significant negative pressure. However, extensive intravertebral gas accompanied by bone edema and/or fluid collections should raise concern for emphysematous osteomyelitis and prompt further evaluation [2, 8]. Radiologists should consider a comprehensive differential diagnosis and exercise caution when interpreting imaging studies to arrive at an accurate diagnosis and appropriate management plan for their patients.

The mainstay of emphysematous osteomyelitis treatment is surgical intervention, which involves amputation, surgical debridement, and intravenous antibiotic therapy. These methods have been successful in managing the infection and halting further tissue loss [9, 10]. Emphysematous osteomyelitis is linked to a high mortality rate (32%) and substantial morbidity, particularly in patients with diabetes [11]. Prompt diagnosis and timely treatment are critical in preventing the severe consequences of the disease.

CONCLUSION

In conclusion, EO is a rare but potentially life-threatening condition that should be considered in patients presenting with intraosseous gas on imaging. The radiologist must be aware of the clinical implications of this finding, as early diagnosis and appropriate management are critical for improving patient outcomes. Clinicians should also be vigilant in identifying and managing underlying comorbidities that may predispose patients to the development of EO. Further research is needed to better understand the pathophysiology and optimal management strategies for this condition.

DISCLOSURE OF INTEREST

The authors declare that they have no conflicts of interest concerning this article.

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