

Emphysematous osteomyelitis: Report of two cases and review of literature

Sachin Khanduri, Meenu Singh, Aakshit Goyal, Simran Singh

Department of Radio-diagnosis, Era's Lucknow Medical College and Hospital, Lucknow, Uttar Pradesh, India

Correspondence: Dr. Aakshit Goyal, Department of Radio-diagnosis, Era's Lucknow Medical College and Hospital, Sarfarazganj, Hardoi Road, Lucknow - 226003, Uttar Pradesh, India. E-mail: aakshit11g@gmail.com

Abstract

Emphysematous osteomyelitis is a rare condition characterized by the presence of intraosseous gas. A prompt diagnosis is required for this disease to expedite management as it is a potentially fatal condition. Many comorbidities, such as malignancy, diabetes mellitus, alcohol abuse, Crohn's disease, and other etiologies causing immunosuppression, predispose to this condition. The causative organisms are generally anaerobes or members of Enterobacteriaceae family; however, the infection can be mono or polymicrobial. We report two cases affected with emphysematous osteomyelitis due to varied underlying comorbidities. The purpose of this study is to (a) emphasize the importance of computed tomography in diagnosing emphysematous osteomyelitis and (b) to highlight an unusual location of this rare pathology.

Key words: Anaerobes; computed tomography; emphysematous osteomyelitis; *Enterobacteriaceae*; intraosseous gas

Introduction

Presence of intraosseous gas in appendicular skeleton in the absence of a history of surgery, trauma, or degenerative changes is highly suggestive of emphysematous osteomyelitis. However, the presence of intraosseous in vertebral bodies is generally due to a noninfectious cause, most commonly being degenerative process.^[1,2] We present two cases of this rare entity aged 56 years and 25 years diagnosed with emphysematous osteomyelitis of bilateral, mid, distal foot and left hip joint, respectively.

Case Reports

Case 1

A 56-year-old male admitted to our hospital with complaints of burning micturition and severe pain and swelling in both the feet. He had a medical history relevant for Type 2

diabetes mellitus and chronic alcohol consumption. He had an irregular intake of the oral hypoglycemic drugs prescribed from a local physician. No history of trauma or surgery was given.

On examination, he was febrile with body temperature of 38.4°C. Lower extremity examination revealed swelling and tenderness over both dorsal and plantar aspects, more on the plantar aspect. Posterior tibial pulse was normal; however, dorsalis pedis pulse could not be elicited due to severe tenderness and swelling. Laboratory findings revealed leukocytosis ($30 \times 10^9 L^{-1}$ with 80% neutrophils), elevated C-reactive protein (>2.8 mg/L), HbA1c was ~8.3%. Liver and renal function tests were within the normal limits. His urine examination revealed pus cells (15–20), 1+ sugar, trace albumin, and no casts/crystals. Red blood cells were 0–1 per high power field.

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Specimens for urine culture were obtained prior to start of empirical antibiotic – ciprofloxacin. His blood glucose levels were managed with intravenous insulin. Sensitivity testing later revealed growth of *Escherichia coli* after 24-h incubation sensitive to ciprofloxacin, gentamicin, and piperacillin. Gram stain revealed growth of gram-negative bacilli.

He was then referred to the Radio-diagnosis Department for X-ray of both of his feet, which did not reveal any significant abnormality, but his excruciating pain prompted us for further evaluation. Color Doppler study of lower limb was performed, which revealed mild intimal wall thickening of the arteries and normal color flow and spectral pattern in femoral artery, popliteal artery, and anterior and posterior tibial arteries. Dorsalis pedis artery could not be assessed because of excessive swelling over the feet. Further during his course of hospital stay, computed tomography (CT) angiography of lower limb vessels was planned, which did not reveal any vascular pathology. However, multiple gas pockets were seen in tarsals, metatarsals, as well as phalanges of both feet. Air locules were also noted in the surrounding soft tissues. Possibility of emphysematous osteomyelitis with necrotizing fasciitis was considered and the patient was urgently planned for bilateral below ankle amputation [Figure 1A-D]. Patient was started on piperacillin and tazobactam postoperatively. Tissue specimens obtained from the amputated part were sent for gram stain and culture, which again revealed growth of *E. coli*, thus pointing toward possible source of infection being urinary tract. No further complications occurred on follow-up.

Case 2

A 25-year-old female was admitted to the emergency department complaining of left hip pain and difficulty

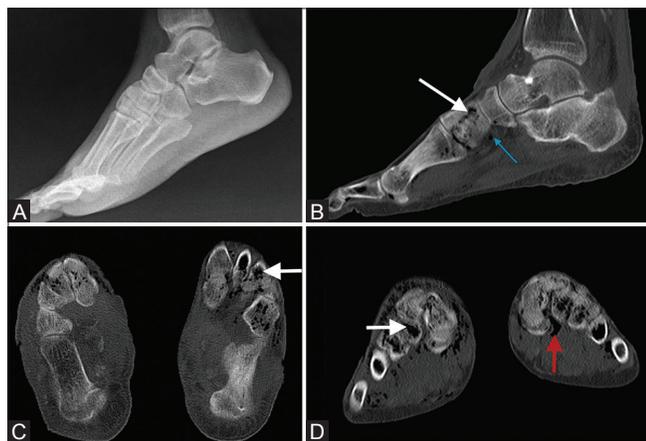


Figure 1 (A-D): (A) Plain radiography of foot (lateral view) of 56 year old male depicting soft tissue thickening along dorsal and plantar aspect. (B) CT lateral view of foot showing presence of gas in metatarsal, tarsal bones (white arrow), phalanges and in soft tissues (blue arrow). (C) CT axial view of both feet depicting presence of intraosseous gas in metatarsal, tarsal bones (white arrow) and in soft tissues. (D) CT axial view of metatarsal bones depicting intraosseous gas (white arrow) and gas in soft tissues (red arrow)

walking since 3 days. She was diagnosed with acquired immunodeficiency syndrome about 2 months ago and was taking prescribed highly active antiretroviral therapy. She gave no history of trauma or surgery. On examination, she was afebrile with normal vitals. Her neurological examination was normal and her lower limb examination revealed mild tenderness over the left gluteal region; however, deep tendon reflexes and power were within the normal limits. Laboratory examination showed normal blood cell counts. Her CD4+ T-cell count was 300 (within normal limits). She was referred to Department of Radio-diagnosis for plain radiograph of pelvis, which did not reveal any significant abnormality. She was subsequently discharged with a prescription of anti-inflammatory drugs.

She returned a week later with the same complaints. On examination, she was tachycardic and febrile with a body temperature of 38.2°C. Her lower examination revealed pain on flexion and extension of left hip joint. The lower limb pulses were within the normal limits.

She was then referred to Radio-diagnosis Department. Since her plain radiographs were within the normal limits in the initial visit, she was investigated with a CT scan of the lower abdomen. Multiple air locules were noted in the left iliac blade and acetabulum with multiple ill-defined hypodense areas in periarticular muscles of left hip joint with surrounding fat stranding [Figure 2A-C]. A possibility of emphysematous osteomyelitis was considered and she was subjected for a fine needle aspiration cytology, a specimen of which was also sent for culture and sensitivity tests. She was empirically started on metronidazole and piperacillin. Her gram stain revealed gram-negative rod-shaped bacteria. Culture and sensitivity revealed growth of *Klebsiella pneumoniae* sensitive to piperacillin, amikacin, and ciprofloxacin. She was put on piperacillin and tazobactam, and metronidazole was discontinued. No complications were noted on follow-up.

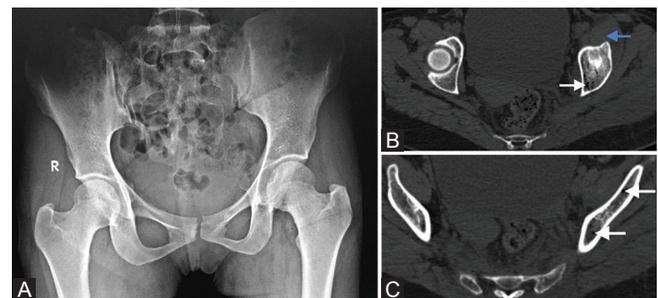


Figure 2 (A-C): (A) Plain radiograph of pelvis AP view depicting no significant abnormality. (B) CT axial of hip joint showing presence of intraosseous gas in left acetabulum (white arrow) and ill-defined hypodense areas in periarticular muscles of left hip joint (blue arrow) (C) CT axial view of pelvis depicting few air locules in left iliac blade (white arrow)

Discussion

Emphysematous osteomyelitis is a rare and potentially fatal condition characterized by the presence of intraosseous gas.^[3] Only 25 cases have been reported in the literature so far with common locations being pelvis, femur, tibia, fibula, thoracolumbar vertebra.^[1] Mautone *et al.*^[4] reported the first case of emphysematous osteomyelitis of midfoot, in 2014, which is an unusual location for its occurrence. To the best of authors' knowledge, we reported the second case of emphysematous osteomyelitis of midfoot with associated involvement of the distal foot. Also, we believe this is the first reported case of involvement of bilateral feet. There is no gender predilection for its occurrence.

Hematogenous dissemination is the most common route of spread of infection.^[5,6] However, McDonnell *et al.* have also reported rare modes of disease spread, such as an extension of intraabdominal infection, intraabdominal, or spinal surgery, or from skin or soft tissue infection.^[2]

Presence of intraosseous gas in intravertebral space is generally a feature of disc degeneration; however, extensive intraosseous gas, bone marrow edema, and/or adjacent fluid collections should raise the possibility of emphysematous osteomyelitis, which was the possible scenario in our second case that showed evidence of intraosseous gas with adjacent fluid collection in the pelvis.^[5]

Monomicrobial or polymicrobial infection by anaerobes or members of Enterobacteriaceae family is generally the cause of infection,^[1,3,7] which was the same in all of our cases.

Certain differential diagnoses, such as penetrating wounds, open fractures, postbiopsy, lymphangiomatosis of bone, need to be excluded before diagnosing a case with emphysematous osteomyelitis.^[7,8]

A radiologist plays a critical role in diagnosing a case of emphysematous osteomyelitis as prompt management is required once a case is confirmed. Plain films can be helpful in detecting the air pockets in bone and soft tissues, but did not provide any clue to point to this diagnosis in both of our cases. CT detection of intraosseous gas was first described by Ram *et al.* in 1981.^[7] Our CT findings diagnosed our cases with sufficient confirmation and also described the entire

extent of the spread. Ultrasonography has a very limited role as it is difficult to diagnose air in soft tissues. Magnetic resonance imaging can also detect the presence of gas, but there is paucity of literature on its correlation with CT in detection of intraosseous gas.

The patients in our study were referred to surgical department for their management, with one of them being managed with combined surgical and medical treatment and the other responded to antibiotic therapy.

Conclusion

Emphysematous osteomyelitis is a rare condition, which if not diagnosed early has significant morbidity and can even prove fatal. CT plays a vital role in the diagnosis and extent of this condition, thus emphasizing indispensable role of a radiologist.

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Conflicts of interest

There are no conflicts of interest.

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