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Case Report

Spontaneous emphysematous osteomyelitis of the spine: A case report ☆,☆☆

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ABSTRACT

Spontaneous emphysematous osteomyelitis of the spine is a very rare but severe condition caused by gas-forming microorganisms. We present the case of a 41-year-old obese male patient presenting at the Emergency Department with diabetic ketoacidosis, fever, and difficulty in walking. Computed tomography and magnetic resonance imaging of the spine revealed bone marrow edema and intraosseous gas collections at the level of the fourth and fifth lumbar metamer, suggesting an osteomyelitis sustained by gas-forming microorganisms, which was then confirmed by the isolation of *Klebsiella pneumoniae* in blood culture.

Imaging plays a central role for the diagnosis of emphysematous osteomyelitis: the presence of multiple intraosseous gas collections of variable size, especially in patients with specific risk factors, is highly suggestive of an infection caused by gas-forming pathogens. Being familiar with this peculiar neuroradiological appearance is essential to ensure an early diagnosis and a timely antimicrobial therapy, which can considerably ameliorate the prognosis.

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Case Report

A 41-year-old obese man was admitted to the Emergency Department in a comatose state due to diabetic ketoacidosis. He had a 7-days history of pyrexia and worsening difficulty in walking, without pain or evidence of trauma.

His medical history was remarkable for chronic alcoholic hepatopathy and uncontrolled diabetes mellitus.

Computed tomography (CT) of the chest revealed pneumonia of the left basal lobe with pleural effusion, prompting the initiation of an empirical antibiotic therapy with levofloxacin and ceftriaxone.

Head CT was unremarkable.

Abbreviations: EO, Emphysematous Osteomyelitis; SPP, Species.

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Fig. 1 – A: CT Scan (sagittal reconstruction) showing the presence of multiple irregular foci of intraosseous gas in vertebral bodies L4 and L5 and in sacral metamer (white arrow), extending to intervertebral disc and epidural space (black arrow). **B:** Axial reconstruction showing the bubbly pattern of intravertebral gas at the level of L5.

In the 24 hours following hospitalization, the patient developed acute paraplegia, with mild bilateral arm weakening. The neurologic examination revealed weakness and hypoesthesia of the upper limbs and flaccid paralysis and anesthesia of the lower extremities, prompting the execution of spine imaging.

CT of the spine showed emphysematous lesions in the spinal canal, along with intraosseous gas collections involving L4, L5, (as well as the interposed intervertebral disc) and the first sacral metamer, with no evidence of vertebral fractures (Fig. 1).

Subsequently, the patient underwent spine magnetic resonance imaging (MRI) which revealed areas of signal alteration mainly encompassing the 2 adjacent vertebral bodies at L4-L5 and the interposed disc, with high signal on T2w images and corresponding low signal on T1w sequences, suggestive of bone marrow edema. Presence of intraosseous gas was also detectable as nuclei of reduced signal in all sequences [1,2] (Fig. 2).

Laboratory tests indicated anemia (red blood cells count = $3.84 \times 10^5/\mu\text{L}$), thrombocytopenia (platelet count = $11 \times 10^3/\mu\text{L}$), neutrophilia (77.3% relative value), hypoalbuminemia (1.4 g/dL), high C reactive protein level (16.6 mg/dL), and hyperglycemia (glucose = 269 mg/dL) with high glycated hemoglobin level, indicative of uncontrolled diabetes.

Blood culture was positive for *Klebsiella pneumoniae* spp.

Due to the absence of discrete abscessual lesions, drainage was not required, but an intravenous antimicrobial therapy with tigecycline and ceftazidime was immediately started,



Fig. 2 – A: MRI exam shows elevated signal intensity in T2-weighted sagittal images at L4-L5 (white arrows) and corresponding low signal in T1-weighted sagittal images (white arrows). **B:** At the same level, the intervertebral disc is involved with narrowing of the correspondent intervertebral space. There is no evidence of epidural or soft-tissue abscesses.

along with supportive measures, leading to a nearly complete resolution of the symptoms. The patient was finally discharged with the advice of follow up and physical therapy because of persistent mild bilateral leg weakness.

Discussion

Spontaneous emphysematous osteomyelitis (EO) represents a rare but severe infectious disease sustained by gas-forming pathogens, associated with high morbidity and mortality [3].

Intraosseous gas collection in the extra-axial skeleton without history of trauma, orthopedic or surgical procedures or penetrating wounds is considered pathognomonic of this condition and was first described by Ram et al. in 1981 [4,5].

To the best of our knowledge, to date, only 35 cases have been reported in English literature, including our case, and only 10 showed spine involvement.

No sex deviation has been described, with a median age at presentation of 51 years [6]. The most common predisposing risk factors consist of congenital or acquired immunodeficiency conditions, including diabetes mellitus, corticosteroid therapy, hematological diseases, malignant neoplasms, HIV infection, and alcohol abuse [7,8].

The clinical differential diagnosis between emphysematous and nonemphysematous osteomyelitis is challenging due to the overlapping of signs and symptoms, while radiological exams are usually decisive [9].

Intra-osseous vertebral gas collections are most frequently due to noninfectious disorders, such as degeneration, malignancies, osteonecrosis, fractures, and major trauma [10].

In rare cases of infectious etiology, the causative organism usually belongs to the bacterial families of Enterobacteriaceae or Anaerobes, similarly to those responsible for other gas-forming infections, with the possibility of poly-microbial infections [8,11].

Typically, in patients with spinal involvement and specific underlying comorbidities (eg, diabetes mellitus), pathogens are identified in organic cultures, with a reported prominent role for species like *Klebsiella Pneumoniae* or *Fusobacterium Necrophorum* [6,7].

In our case, the EO was sustained by *Klebsiella Pneumoniae*, probably due to the hematogenous spread of a pulmonary infection, in a patient with diabetic ketoacidosis and alcohol abuse. Interestingly, patients with diabetes mellitus and uncontrolled glycemic status are known to be more susceptible to infections sustained by capsulated bacteria, due to an impaired neutrophilic activity [8].

Imaging plays a central role for an accurate diagnosis. CT is able to detect and define the extension of intraosseous and soft tissue gas collections, and also to reveal the possible presence of abscesses. In particular, the pattern of gas distribution can aid the differential diagnosis: a linear and well-defined pattern of gas distribution without alteration in adjacent tissue suggests degenerative changes (eg, intraosseous pneumatocysts, subchondral cysts, Schmorl's nodes), while a bubbly and irregular pattern can be considered as pathognomonic of EO [12].

Small et al. in 2018 showed that the presence of multiple foci of intramedullary gas of variable size, defined as a "pumice stone pattern", is suggestive of emphysematous infection [13].

MRI can also reveal clusters of intraosseous gas and is more accurate in detecting signal abnormalities of bone marrow and epidural envelopment, as well as subtle soft tissue alterations and possible neurological complications.

Author contributions

All the authors contributed to this article.

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None.

Patient consent

The patient has consented to the submission of this case report to the journal.

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