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Scientific letter

Emphysematous osteomyelitis of the hip and iliac bone: Serious infection with a characteristic radiographic finding

Osteomielitis enfisematosa de cadera y hueso ilíaco: infección grave con una imagen radiográfica característica

Emphysematous osteomyelitis (EO) is an extremely rare infection caused by gas-producing bacteria.[1] It has high morbidity and mortality rates, which means it requires an early and aggressive therapeutic approach.[2] We present a case of EO of the hip with poor progress in a patient who presented with septic shock. We show a characteristic radiological sign which was key to establishing the suspected diagnosis.

This was a 52-year-old woman with a history of type II Chiari malformation with secondary paraplegia who was brought to Accident and Emergency due to disorientation and low level of consciousness for a number of hours.

On arrival at the hospital she was stuporous, tachypnoeic and tachycardic. Examination revealed the presence of a pressure ulcer in the right gluteus developed in the previous month and managed at home with topical wound care, despite which it had increased in extension and depth.

The analysis revealed leucocytosis (19,200 leucocytes/μl) and elevated C-reactive protein (CRP) (>25 mg/dl). Brain computed tomography (CT) and chest x-ray showed no abnormalities.

The patient's level of consciousness deteriorated and she became haemodynamically unstable, requiring admission to the ICU.

Given the suspicion of sepsis with a focus on the skin and soft tissues, an CT of abdomen and pelvis with contrast was requested (Fig. 1), which showed findings compatible with EO.

The Orthopaedics and Trauma Department was contacted, and they performed urgent surgery with resection of the right femoral head, cleaning and debridement.

Initially, she was given empirical antibiotic therapy with meropenem, linezolid and amikacin. The following were isolated in blood cultures without antibiotic pressure: *Streptococcus anginosus, Streptococcus constellatus, Bacteroides fragilis* group, *Clostridium ramosum* (2/2). Cultures of bone and periarticular tissue after surgery grew: *Escherichia coli, Streptococcus anginosus, Streptococcus constellatus, Eikenella corrodens, Clostridium* spp., *Prevotella intermedia, Parvimonas micra* and *Bacteroides caccae.*

After the above isolations, the antibiotic therapy was adjusted to piperacillin/tazobactam, ciprofloxacin and daptomycin. Given the probable involvement of anaerobic bacteria (*Clostridium* spp. and *Bacteroides* spp.), metronidazole was added to the combination after performing antibiotic sensitivity tests.

The patient made very slow progress, with the need for drainage of collections and debridement of pressure ulcers. After more than four months in hospital, the focus was under control and the patient was discharged.

The presence of intraosseous gas was described for the first time in 1981.[3] When it is identified in the vertebral bodies, it is usually related to non-infectious causes.[1] In contrast, in the extra-axial skeleton, it is a finding highly suggestive of EO.[4]

EO is a serious bone infection with very few cases described in the literature.[1] It is generally produced by haematogenous or contiguous spread. In the first case, monomicrobial infections predominate, while in the second, polymicrobial infections.[2] When it spreads by contiguity, it is usually derived from an intra-abdominal infection, spinal surgery, or skin and soft tissue infection (SSTI).[5]

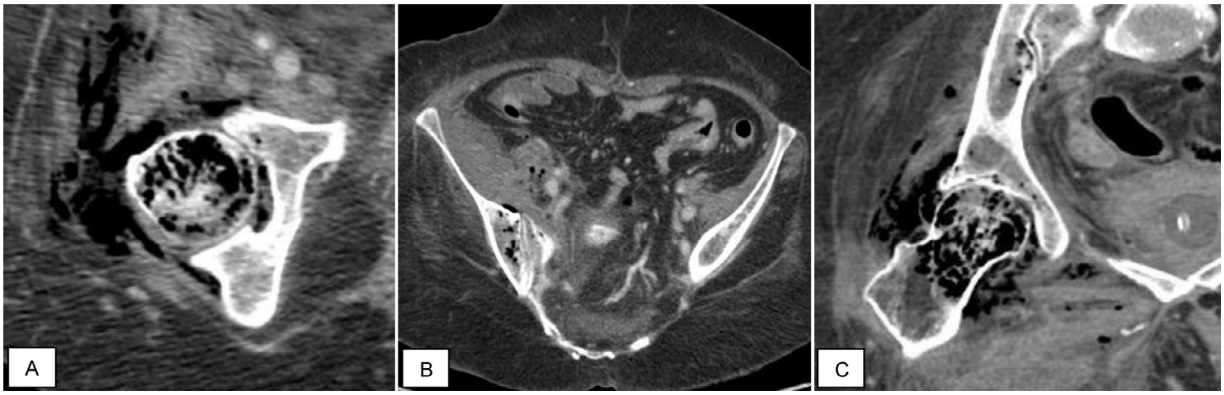

Figure 1. Extensive inflammatory process mainly involving the components of the right hip, with the presence of gas in the thickness of the bone of the femoral head (¨pumice stone sign¨) (A and C), in the iliac acetabulum and in the right femoroacetabular joint (B). The presence of gas can also be seen in the adjacent soft tissues (A and C).

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The identification of polymicrobial bacteraemia should alert to the presence of an uncontrolled focus, generally intra-abdominal.[6] In our patient, we suspect that the most likely mechanism was the deep extension of an SSTI due to her history of pressure ulcer with adjacent pyomyositis and arthritis. This case illustrates the importance of adequate management of chronic wounds and pressure sores, as they can be a focus of polymicrobial bacteraemia.

From the microbiology point of view, anaerobic bacteria and enterobacteria are usually isolated[7] In our case it was a polymicrobial infection with predominance of *Clostridium* spp. and *Bacteroides* spp. Given the severity of anaerobic bacteraemia, antibiotic sensitivity tests must be performed to confirm the sensitivity of these species to metronidazole, piperacillin/tazobactam and carbapenems, as sensitivity can vary between different species and strains.[8]

Contrast-enhanced CT is the most sensitive test for diagnosing EO, showing the "pumice stone sign" in more than 90% of cases. Other typical radiographic findings are emphysema in the surrounding soft tissues and the absence of destruction of the cortical bone.[1]

Treatment is combined with parenteral broad-spectrum antibiotic therapy for at least four weeks and surgical control of the focus.[1,4,9]

It is important to define the optimal duration of antibiotic therapy and the need for surgical intervention in all cases, as multiple or complex surgical interventions can increase mortality rates.[9]

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Conflicts of interest

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First characterization of a *Klebsiella pneumoniae* clinical isolate producing VEB-1 and OXA-436 in Spain

Primera caracterización de un aislado clínico de Klebsiella pneumoniae *productor de VEB-1 y OXA-436 en España*

Sir,

Extended-spectrum β-lactamases (ESBLs)-producing Gram-negative bacilli are a major cause of resistance to third-generation cephalosporins. The genes encoding these beta-lactamases are often carried in plasmids, which have allowed them to spread horizontally worldwide, and are currently endemic in community and hospital-acquired Enterobacterales. These enzymes hydrolyse third-generation cephalosporins and aztreonam, but are not active against cephamycins, and they are inhibited by clavulanic acid and by tazobactam. Currently, CTX-M, SHV and TEM family of ESBLs are the most common in most geographical areas worldwide and specifically in Spain. However, there are other less frequent families such as PER, GES or VEB.[1]

VEB (*Vietnamese extended-spectrum β-lactamase*) is a family of ESBLs, that has so far localised in class 1 integrons.[2] It was first detected in Southeast Asia and subsequently in African and American countries.[3,4] This *bla* gene has been found in Enterobacterales, *Achromobacter xylosoxidans*, *Pseudomonas aeruginosa* and *Acinetobacter baumannii*. In Europe it has been found in both *Klebsiella pneumoniae* and in non-fermentative Gram-negative bacteria since 2001.[5–10] Recently, a clinical case of VEB-producing *K. pneumoniae* was detected in our hospital of Seville and, to date, this is the first description of VEB-1-producing *K. pneumoniae* in Spain.

In addition, the incidence of clinical infections caused by carbapenemase-producing organisms has increased in the last years.[11] A rare plasmid-mediated variant of OXA-48 called OXA-436 has been described.[12] The enzyme has been shown to be a class D carbapenemase similar to OXA-48 in terms of substrate specificity. It has a higher activity at higher temperatures, resembling a human infection scenario, whereas OXA-48 has activity at lower temperatures, indicating an environmental scenario. However, no significant difference is shown in antimicrobial susceptibility profiles *in vivo*.[13] It was initially discovered in an *Enterobacter asburiae* isolate from a patient admitted to a hospital in the capital of Denmark and subsequently detected in other enterobacteria (*Citrobacter freundii*, *K. pneumoniae*, *Escherichia coli*) in different Danish hospitals.[11,12,14] OXA-436 has also been identified in a strain of *Shewanella putrefaciens* from a Pakistani hospital.[15]