LONGITUDINAL CORTICAL SPLIT SIGN AS A POTENTIAL DIAGNOSTIC FEATURE FOR CORTICAL OSTEITIS

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Septic cortical osteitis is a rare but distinct type of bone infection that is characterized as a hematogenously seeded infection predominantly or exclusively limited to the cortex. Diagnosis is difficult and often delayed. Combination of clinical and laboratory findings together with the typical radiological findings consisting of vertically orientated cortical osteolysis, the 'cortical split sign' and the predominantly cortical disruption at the periosteal side of the cortex may lead to the correct diagnosis.

Key-word: Bones, infection.

In adults, subacute and chronic forms of osteomyelitis are frequently encountered, usually secondary to an open injury to bone and surrounding soft tissue. Septic cortical osteitis – however – is a rare subgroup of bone infection caused by hematogenous spread that is predominantly or exclusively limited to the cortex of long tubular bones and usually affecting adolescents and young adults.

Diagnosis can be difficult because of the nonspecific clinical findings and sometimes misleading imaging features mimicking a focal bone lesion. The purpose of this paper is to present three cases of septic cortical osteitis, in which the cortical split sign may be a potential useful clue to the correct diagnosis. The literature on the subject is reviewed.

Case report

Case 1

A 17-year-old, otherwise healthy male student and football player, presented to his general practitioner complaining of a continuous pain in the left leg, causing limping and awaking him at night. Symptoms started abruptly one month previously, without any history of trauma. Clinical examination revealed a firm, slightly painful area at the postero-lateral aspect of the left thigh. Laboratory investigation was unremarkable except for a slightly raised C-reactive protein (CRP).

Plain radiographs showed a 12 cm long cortical defect paralleling the long axis of the middiaphyseal femur with a central linear density and faint periosteal reaction (Fig. 1A). Computed tomography (CT) demonstrated a longitudinal osteolysis with central density and partial cortical disruption at the periosteal side of the cortex (Fig. 1B-C). Magnetic Resonance (MR) imaging showed an irregularly delineated cortical defect on the posterior side of the femur, surrounded by edema in the bone marrow and peri-osseous soft tissues. The cortical defect was slightly hyperintense to muscle on T1- and T2-weighted images, with a central linear hypointense focus on both pulse sequences, representing a sequestered bone fragment (Fig. 1D). After administration of intravenous gadolinium contrast, the cortical lesion showed peripheral rim enhancement as well as enhancement of the adjacent soft tissues and bone marrow (Fig. 1E).

The diagnosis of chronic cortical osteitis was suggested and surgical debridement was performed. Culture of the obtained tissue was positive for Staphylococcus aureus and histology confirmed the diagnosis of cortical osteitis. Post-operatively, a 6-week course of antibiotics was initiated. The further follow-up was uneventful.

Case 2

A 38-year-old female presented to the emergency department with pain in her left upper limb. The symptoms were of insidious onset without history of trauma, getting worse in recent days. Plain radiographs of the left femur showed a 7 cm long longitudinal intracortical lucency on the anteromedial side of the left femur with preservation of the internal cortical lining, irregular thickening of the outer cortex and disorganized periosteal reaction (Fig. 2A). CT confirmed a longitudinal osteolytic area in the anteromedial cortex containing internal central linear densities, representing calcifications or bone fragments. There were multiple radiolucent defects in the outer cortex and an interrupted periosteal reaction but there was absence of intramedullary extension (Fig. 2B-C). T2-weighted, fat-saturated MRI showed increased signal intensity in the cortex with focal outer cortical disruption and associated soft tissue and bone marrow edema (Fig. 2D-E). Surgical debridement was performed and histological and laboratory findings confirmed the diagnosis of cortical osteitis. After a 3 month period of antibiotic treatment, there was a complete clinical recovery.

Case 3

A 30-year-old male recreational football player presented with gradually worsening pain in his right upper leg since three weeks. As in the two previous cases, plain radiographs and CT revealed a 6 cm long longitudinal cortical osteolysis with a central linear density surrounded by faint periosteal reaction (Fig. 3A). Partial cortical disruption at the periosteal side of the cortex was better evaluated on CT (Fig. 3B-C). MRI showed an irregularly delineated cortical defect, surrounded by extensive bone marrow edema and peri-osseous soft tissue edema (Fig. 3D). Histological examination after surgical debridement confirmed the diagnosis of cortical osteitis.

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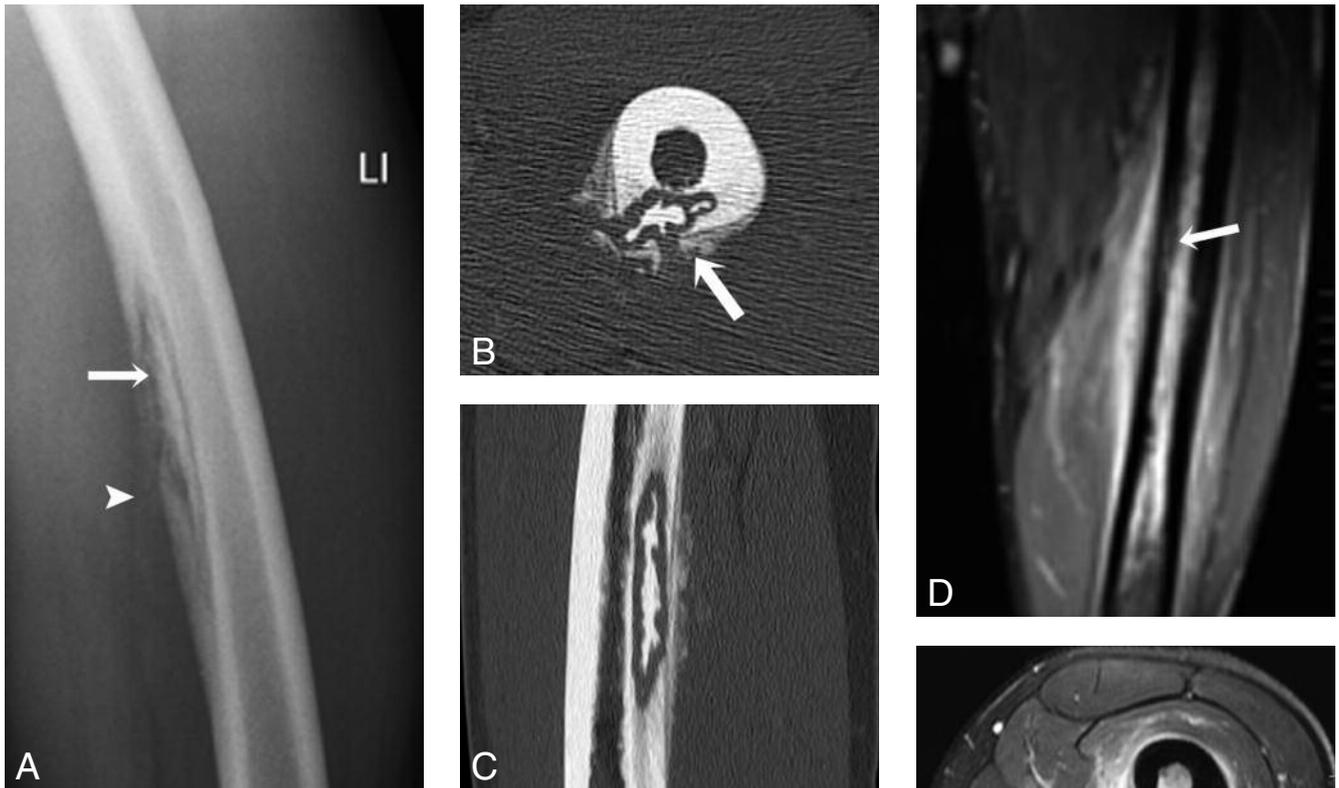
Fig. 1. — A. Plain radiographs of the left femur show a longitudinal cortical defect with a central linear density (arrow) and faint periosteal reaction (arrowhead). B. Axial CT image shows a well defined focal osteolysis of the outer cortex, which contains a central linear sequestrum. There is cortical disruption at the periosteal side with periosteal reaction (arrow). C. Sagittal reformatted CT image demonstrates the typical 'cortical split sign', consisting of a vertically orientated osteolysis with a central linear sequestrum. D. Coronal fat-suppressed T2-weighted MR image of the femur shows a linear hyperintensity in the medial femoral cortex (arrow) and high signal intensity involving the marrow and surrounding soft tissues representing edema. E. Axial fat-suppressed T1-weighted MR image after intravenous administration of gadolinium contrast depicts rim enhancement of the lesion, with enhancing perilesional soft tissue and bone marrow edema.

Discussion

Septic cortical osteitis represents an unusual type of bone infection, first described during the nineteenth century by Gerdy (1). It is characterized as a hematogenously seeded infection predominantly or exclusively limited to the cortex. Only a few case reports have been reported in literature (1, 2).

The mechanism of bacterial deposition is thought to be caused by hematogenous spread of septic emboli in the periosteal plexus. These plexuses are derived from the arteries of the neighboring muscles and supply the outer third of the cortex (Fig. 4). Any form of end-capillary obstruction, including a small hematoma caused by minor trauma, could produce an area of avascular necrosis that predisposes to infection (3). These anatomic and patho-genetic data may explain why cortical disruption occurs predominantly at the periosteal side of the cortex, whereas the endosteum usually remains intact, such as demonstrated in our three cases. Interestingly, two of our patients were soccer players, which favor the hypothesis that minor trauma may contribute to the pathogenesis.

The long bones of the lower limb are the most frequent locus of infection. Like in other forms of osteomyelitis, Staphylococcus aureus is the most common causative infective organism.

Diagnosis may be challenging because patients present with vague symptoms consisting of local tenderness and minimal or even no fever. Routine laboratory tests are usually nonspecific with normal leukocyte count and slightly elevated CRP. Therefore, the infection is often left untreated initially and can progress to a chronic condition resulting in focal bone loss, sequestrum formation, and reactive bone sclerosis.

Moreover, the radiologic findings are often nonspecific and depend on the time of presentation.

Plain radiographs may reveal a cortical osteolysis along with the long axis of the long bone with a central linear density and limited periosteal reaction. Although ultrasound may demonstrate elevation and/or thickening of the periosteum of more than 3 mm due to pus emanating from the cortex, it provides only limited information about bone changes (3). CT is superior for precise evaluation of the extent of cortical destruction and is the preferred modality for identification of anosseous sequestrum, resulting in the 'cortical split sign'. Cortical disruption – even subtle – at the periosteal side is also better assessed on CT compared to plain radiography. On the other hand, MRI is far superior to demonstrate asso-
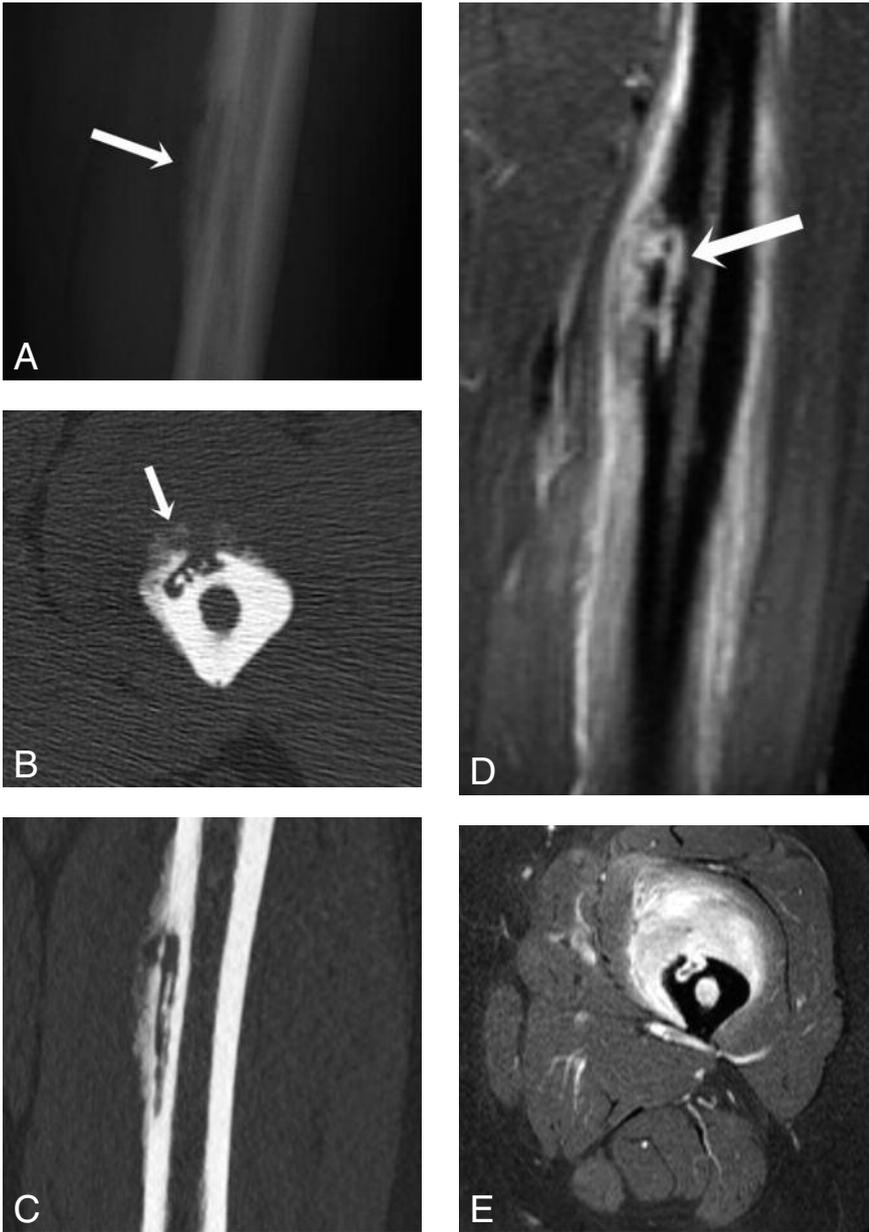
Fig. 2. — A. Plain radiographs of the left femur show an intracortical lucency on the anteromedial side of the left femur with preservation of the internal cortical lining and thickening of the outer cortex. There is a partially disrupted periosteal reaction (arrow). Note the vertical course of the lesion, parallel to the cortical bone. B. Axial CT image shows a focal cortical osteolysis with central punctiform densities. Note the focal cortical destruction and faint periosteal reaction (arrow). C. Coronal reformatted CT image demonstrates the 'cortical split sign'. D. Coronal fat-suppressed T2-weighted MR image demonstrates high signal intensity in the medial femoral cortex (arrow), the marrow and surrounding soft tissues. E. Axial fat-suppressed T1-weighted MR image with gadolinium contrast shows enhancement of the focal cortical destruction and associated bone marrow and soft tissue edema.

Fig. 3. — A. Plain radiographs of the right femur show a longitudinal osteolysis in the lateral femoral cortex with central density and faint periosteal reaction (arrow). B. Axial CT image shows a ill defined osteolysis of the outer cortex, which contains a central sequestrum. There is cortical disruption at the periosteal side with faint periosteal reaction. C. Coronal reformatted CT image demonstrates the 'cortical split sign'. Note the intact endosteal cortex (arrow). D. Coronal fat-suppressed T2-weighted MR image demonstrates an irregularly delineated cortical defect, surrounded by extensive edema in the bone marrow and peri-osseous soft tissues (arrow).

ciated bone marrow edema and soft tissue inflammation, which is another important clue in the differential diagnosis with noninfectious lesions. Whenever infection is suspected, gadolinium contrast administration is mandatory for evaluation of sinus tracts, fistulas, and soft tissue abscesses.

A bone lesion that consists of a lucent area with a central sclerotic focus has been referred to as the 'button sequestrum sign' (4). This sign was first described as a radiological manifestation of eosinophilic granuloma, a localized form of Langerhans' cell histiocytosis, but this sign may be seen in other

Fig. 4. — Line drawing of the vascular supply of the cortex of a long bone. The outer third of the cortical bone is supplied by periosteal plexuses (1), derived from arteries of neighbouring muscles and soft tissues. These periosteal plexuses form connections with the cortical capillaries (2), derived from medullary branches, which supply the inner two thirds of the cortex.

entities such as osteomyelitis, fibrosarcoma and lymphoma as well.

Other differential diagnoses to be considered in case of a cortical radiolucency with a central density include osteoid osteoma, longitudinal stress fracture and osseous changes associated with calcium hydroxyapatite crystal deposition disease (HADD). Osteoid osteoma (O.O.) appears as a lucent area representing the nidus, which may be uniformly radiolucent or contain variable amounts of calcification, and is surrounded by bone sclerosis. Unlike the osteolysis in cortical osteitis which shows an irregular lining of the inner borders, the lucent area in O.O. has more smooth round margins (5). Moreover, the more extensive length of the cortical osteolysis in our cases argues against O.O.

A longitudinal stress fracture can also present as a linear cortical lucency but on axial CT or MR images, the cortical cleft representing the fracture line is visible on multiple adjacent sections. Other secondary findings of a longitudinal stress fracture are eccentric periosteal reaction and new bone formation, eccentric soft tissue edema and a superomedial location relative to the nutrient foramen (6).

HADD-associated tendinopathy sometimes may produce bone erosion simulating a radiolucent lesion, but this is not often a diagnostic dilemma because of the characteristic site of the calcification and erosion at a tendinous insertion (7).

Treatment of cortical osteitis requires adequate debridement in addition to long-term systemic antibiotic therapy.

In conclusion, cortical osteitis represents a rare subgroup of osteomyelitis. In young patients with non specific complaints, slightly elevated CRP and radiological and CT findings of a vertically orientated cortical osteolysis with the typical 'cortical split sign' and cortical disruption at the periosteal side of the cortex, the diagnosis of cortical osteitis should be strongly considered.

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