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Primary sternal osteomyelitis

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ABSTRACT

Primary sternal osteomyelitis is very rare in children, with less than 100 cases published to date. Its clinical presentation is often non-specific, which results in a diagnostic delay.

Here we describe 2 new cases of primary sternal osteomyelitis. Both referred fever, malaise, chest pain, and refusal to lie down, with pre-sternal erythema in one of the cases. The erythrocyte sedimentation rate and C-reactive protein values were high in both cases. The diagnosis was confirmed by imaging studies; methicillin-sensitive *Staphylococcus aureus* was isolated in the blood culture of one of them. Both recovered without complications with antibiotic treatment.

Primary sternal osteomyelitis should be considered in the differential diagnosis of chest pain, especially if accompanied by fever, local inflammatory signs, intolerance to lying down, or increased acute phase reactants.

Keywords: *osteomyelitis; sternum;* Staphylococcus aureus*; chest pain.*

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INTRODUCTION

Primary sternal osteomyelitis is a very rare condition in pediatrics; its symptoms are usually non-specific, which results in a diagnostic delay. According to a recent review,[1] only 74 pediatric cases had been published between 1864 and 2015. Here we describe 2 new cases and review those published from 2015 to date.

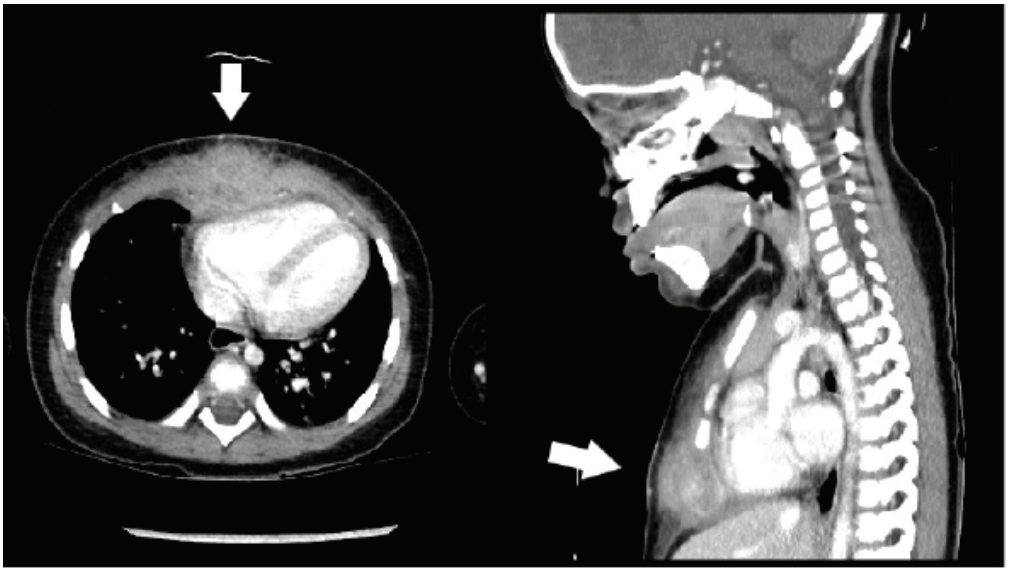
CASE REPORT 1

A 23-month-old girl with Down syndrome consulted because, since 48 hours before, her parents had noticed she was fussy, had labored breathing, and did not tolerate lying down; she did not develop fever. She had been previously assessed twice and had been diagnosed with laryngitis. At the Emergency Department, her vital signs were temperature: 37.7 ºC, heart rate: 136/minute, respiratory rate: 28/minute, and oxygen saturation: 98%; she had continuous grunting with no other signs of respiratory distress. The girl refused to lie down. An erythematous, firm, non-mobile, tender mass was observed in the right parasternal region. The white blood cell count was 13 140/mm^3 (9986 neutrophils/mm^3), with high C-reactive protein levels (61.3 mg/L) and a high erythrocyte sedimentation rate (ESR) (62 mm/h). The chest X-ray showed increased soft tissue in the lower third of the anterior chest wall. The computed tomography of the chest (*Figure 1*) showed increased soft tissue density in front of and behind the xiphoid process and the body of the sternum; the ultrasound of the chest showed that the mass described before extended in a linear trajectory until coming into contact with the sternal cartilages of the xiphoid process (*Figure 2*). The echocardiogram was normal; the blood cultures were negative. She received treatment with intravenous amoxicillin/clavulanic acid and also clindamycin for the first 4 days. After 24 hours of treatment, the patient no longer had grunting and tolerated lying down; in the following days, the swelling gradually decreased, both in clinical terms and as observed in the echocardiogram, without ever observing a drainable collection. After 9 days of intravenous treatment, oral cefuroxime was started until completing a total of 25 days of antibiotic treatment. Four months later, the patient was asymptomatic, with no sequelae.

CASE REPORT 2

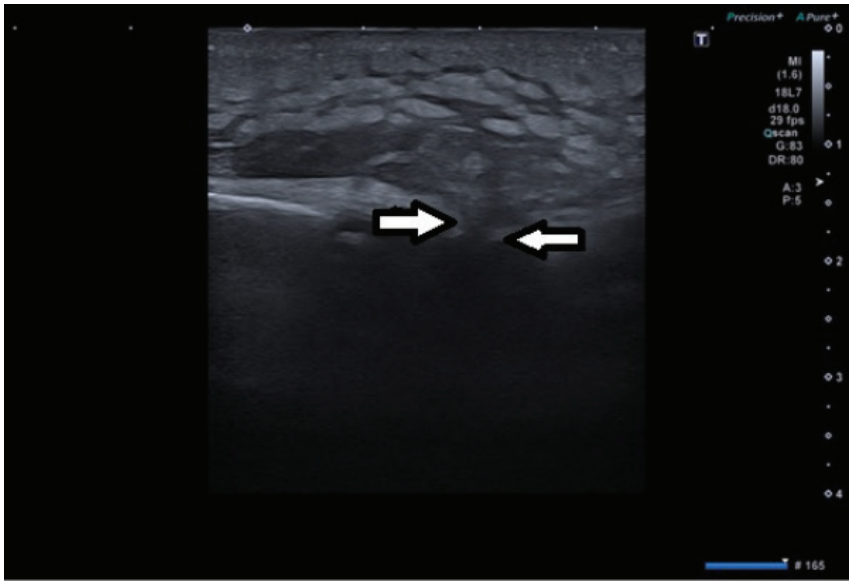
A 12-year-old girl with no history of trauma consulted due to chest pain in the middle third

Figure 1. A (left) Computed tomography. Axial section below the manubrium of sternum. B (right) Sagittal reconstruction



In the lower third of the anterior chest wall, the fat is occupied by a discretely heterogeneous soft tissue density of 5 × 3 × 5 cm (transverse, anteroposterior, and craniocaudal), which is located in front and behind the body and sternal xiphoid process towards the rib cage (arrows).

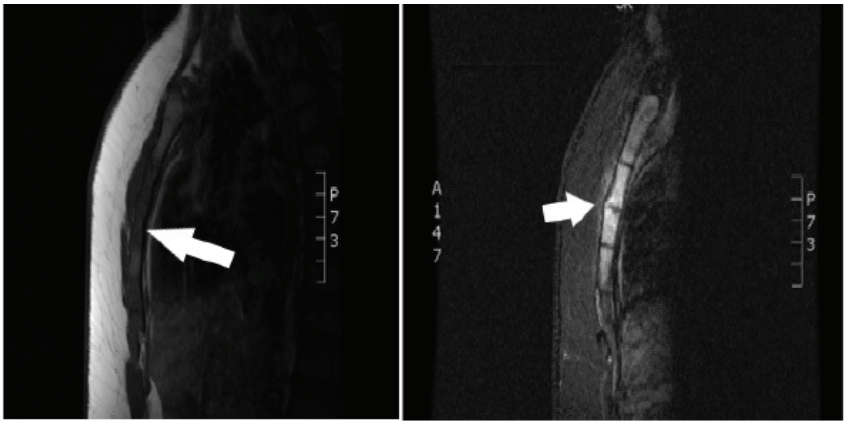
Figure 2. Ultrasound of pre-sternal region



Pre-sternal ultrasound. Image of cobblestone-like appearance in the subcutaneous cellular tissue compatible with cellulitis. Discretely heterogeneous and hypervascularized collection, predominantly hypoechoic, located in the anterior chest wall at the level of the last sternal cartilages, with an image of prolongation towards them in depth (arrows).

of the sternum that worsened with movement and when lying down. Twenty-four hours later, she developed fever (39.5 ºC) so she went to the Emergency Department, where her electrocardiogram and chest X-ray were normal. She was discharged with an oral ibuprofen prescription. She consulted again 48 hours later due to increased pain. She was in a good general condition and her vital signs were normal. The only finding on examination was point tenderness in the mid-sternum, with no erythema or swelling at that level. The white blood cell count was normal (6570/mm^3), but the C-reactive protein (98.8 mg/L) and ESR (55 mm/h) values were elevated. The magnetic resonance imaging (MRI) showed diffuse signal alteration, hypointense signal in T1 with hyperintensity in the sternal marrow in STIR, which suggested bone edema (*Figure 3*). The blood culture was positive for methicillin-sensitive *Staphylococcus aureus*.

Figure 3. Magnetic resonance imaging of the chest, sagittal T1 (left) and sagittal STIR (right) before treatment



Hypointense signal in the sternal body marrow in T1 with hyperintensity in STIR, suggestive of bone edema with mild anterior periostitis (arrows).

Treatment was started with intravenous cefazoline with a favorable course; after 6 days, it was replaced by oral cefadroxil for a total of 24 days. Three months later, she was asymptomatic and the control MRI was normal.

DISCUSSION

Sternal osteomyelitis accounts for less than 1% of bone infections in childhood.[2] Primary sternal osteomyelitis, resulting from hematogenous dissemination, with no relation to a penetrating trauma or local extension from an adjacent organ, is even more uncommon. A comprehensive review of the bibliography up to 2015 described 74 cases, 13 of them in the pre-antibiotic era.[1] Since then, 25 new cases have been reported, including the 2 cases described above.[3–10] Due to its low incidence, the rate of suspicion is low, which is often a reason for a delay in diagnosis and treatment.

The age of onset shows a bimodal distribution, with a peak in early childhood—the median age is 1 year—and a less marked peak around adolescence, although half of the children over 10 years of age were diagnosed in the pre-antibiotic era.[1] Eleven had sickle cell anemia as underlying disease;[1,9] the rest were previously healthy children, except for 1 case of systemic lupus erythematosus.[1]

The most frequent clinical findings include swelling, tenderness, and erythema in the pre-sternal region.[1] Chest pain, present in 1/4 children, was the initial symptom in our 2 patients, with the characteristic that it increased when lying down and improved when sitting up, a finding that had not been previously described and which raises the differential diagnosis from other conditions, such as pericarditis. Fever, present in 60% of cases, is less frequent in *Kingella kingae* infections.[1,8]

The white blood cell count is often normal, but other markers of inflammation, such as ESR and C-reactive protein, are usually elevated,[1] although they may be normal, especially in *K. kingae* infections.[8]

The most frequently isolated microorganism is *Staphylococcus aureus* (40%), followed by *K. kingae* (16%) and *Salmonella enteritidis* (8%).[1,3–9] An increase has been observed in cases caused by *K. kingae* (up to 30% in the past 5 years), especially in children under 3 years of age, probably due to the increased performance of new diagnostic techniques, such as polymerase chain reaction.[3,8,11] *S. enteritidis* causes infection primarily in children with sickle cell disease, although cases have recently been described in infants without hemoglobinopathy.[5] The performance of blood culture in primary sternal osteomyelitis is 30%, similar to that described in acute osteomyelitis in general.[1,2,12]

A plain X-ray helped to make a diagnosis in more than one-third of reported cases.[1] Given that radiologic changes in osteomyelitis are not evident until 10–21 days after the onset of infection,[2] this is indicative of a delayed diagnosis. The median duration of symptoms until hospitalization is 7 days and it is common that, as in our 2 cases, they sought medical attention before.[1]

An MRI is the imaging study that provides the most information for the diagnosis of osteomyelitis because it can detect anomalies early[2] and has a high sensitivity and specificity, although its use in young children is limited due to sedation requirements. Some international guidelines consider that it may not be necessary in certain situations in which other findings are highly suggestive of diagnosis.[2] An ultrasound, useful to rule out the presence of abscesses or soft tissue abnormalities, is less sensitive for the diagnosis of acute osteomyelitis,[2] although an experienced technician may notice periosteal elevation a few days after the onset of symptoms.[5] According to a review, the diagnosis was made by ultrasound, without other imaging studies, in 22% of the cases.[1] In our first case, the fistulous tract communicating the inflammatory tissue with the sternal cartilages was visible; as the course was rapidly favorable and the child would have required sedation, it was decided not to perform an MRI.

Half of the reported cases resolved with antibiotic treatment alone.[1] Empirical treatment should always cover *S. aureus*, the most prevalent microorganism, with coverage against methicillin-resistant strains when their prevalence in the community is higher than 10%. In children younger than 3–5 years, the potential presence of *K. kingae* should be taken into account and it should be remembered that antistaphylococcal penicillins are not the best option against this microorganism.[2] In children with sickle cell disease, the treatment must cover *Salmonella*. If an abscess is present, surgical treatment is mandatory. However, despite the delay in diagnosis in many cases and the fact that abscesses occur in slightly more than half of them, the course is good in most patients, with only 2 cases of mediastinitis or mediastinal abscess[7,13] and only 1 fatal outcome in the post-

antibiotic era, in a severely immunocompromised child.[1]

CONCLUSIONS

Primary sternal osteomyelitis should be taken into consideration in the differential diagnosis of children with chest pain, especially when it worsens when lying down or when there are signs of inflammation or increased acute phase reactants. ■

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