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Osteomyelitis of flat bones: A report of 20 cases and review of the literature

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ABSTRACT

Objective: Osteomyelitis is frequently localized on the fertile metaphysis of long bones. However, there are other locations such as short or flat bones. The aim of our study was to determine the diagnostic, therapeutic and evolutionary aspects of osteomyelitis of flat bones in our environment.
Methods: It's was a retrospective study conducted from January 2013 to December 2017; Children from 0 to 16 years admitted for osteomyelitis of a flat bone where included.
Results: We followed 19 patients who presented 20 locations of osteomyelitis on a flat bone. They accounted for 9.1% of all osteomyelitis observed during the study period. The average age was 6.7 years (9 months–14 years). The clinical picture most often associated with fever and local inflammatory swelling related to an abscess. The predominant locations were the sternum with 8 cases and the scapula with 4 cases. Eleven patients were homozygous sickle cell patients. The diagnosis was strengthened with standard radiography in the majority of cases. A micro-organism was isolated in 8 cases (40%) and *Staphylococcus aureus* was the predominant germ in 7 cases. Nearly all patients were treated with surgical drainage of the abscess, bone curettage and antibiotics. A typical radiographical evolution has been observed with bone reconstruction in 15 cases with a mean follow-up of 3.3 years.
Conclusion: Flat bones are relatively rare locations of osteomyelitis. Standard radiography remains the first-line examination. Their treatment obeys the same rules as that of osteomyelitis of long bones. There is a successful outcome when the care is early and adequate.

1. Introduction

Osteomyelitis is a common disease in paediatrics with an incidence estimated at 1cas/5000 in the USA.[1] It is defined as haematogenous infection of the bone; *Staphylococcus aureus* remains the most common cause of haematogenous osteomyelitis in children.[1–3] In the black population, sickle cell disease has been described to be an additional risk factor.[4] Osteomyelitis is frequently localized on fertile metaphysis of long bones, especially on the femur and tibia.[1,5] However, there are other rare locations such as short or flat bones. Osteomyelitis of flat bones has already been published in isolated case reports[1,6,7] or short case series,[8,9] most often involving a particular type of flat bone. Very few studies[10] reported cases of osteomyelitis affecting every types of flat bones. We therefore collected each and every case of osteomyelitis of flat bones treated in our department in order to determine the diagnostic, therapeutic and evolutionary aspects.

1.1. Materials and methods

This was a retrospective study of children from 0 to 16 years old admitted in the paediatric surgery department between 2013 and 2017. Cases of osteomyelitis affecting flat bones were included. The study population consisted of 19 out of 220 (9.1%) cases of osteomyelitis hospitalized our department during the study period. These 19 patients presented with 20 different locations of osteomyelitis of flat bones. There were 13 boys and 6 girls, with a sex

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ratio of 2.7 and an average age of 6.7 years (range from 9 months to 14 years). Sixteen patients were transferred from other paediatric units and three from home. The studied parameters were as follow:

- **Diagnostic:** Medical history (history of sickle cell disease, diabetes mellitus), time to admission, history of trauma, pain, functional impotence, swelling characteristics, comorbidities; biological findings (haemogram, CRP, bacteriology); imaging: standard radiography, computerized tomography, magnetic resonance imaging.
- **Therapeutic**: antibiotics, surgery.
- **Evolutionary**: duration of hospitalization, outcome.

Data were collected on medical records and hospital records.

1.2. Results

Diagnostic aspects.

1.2.1. Medical history

Eleven out of 19 patients had homozygous sickle cell disease. There were no diabetic patients or any other favouring factor for osteomyelitis.

- Time to admission

The average duration of disease progression before admission was 10.2 days (range, 7–20 days).

- History of trauma

The notion of trauma was found in 2 patients occurring a few days before the onset of symptoms: One case reporting a 14-years-old girl who had been hit on the scapular region with a small stone; and another case of a 7-year-old boy with history of knee trauma after a fall from standing.

- Clinic

All patients had significant bone pain causing functional impotence of the thoracic limb. Fever was found in almost all patients (18 out of 19) upon admission to the service. It ranged from 38° to 41 °C. Only one patient was apyrexial.

All patients presented with subcutaneous swelling related to abscess or bone deformity. Table 1 shows the most common locations. The swelling was inflammatory in 18 cases and pseudotumoral in 1 case (patient with apyrexia).

- Biology

The blood count showed an average white blood cell 17,850/mm^3 (8100 to 24,600/mm^3). Mean CRP was 68.5 mg/l (6–176 mg/l), mean sedimentation rate was 59 mm/s (range 10–128 mm/s). The case of pseudotumoral osteomyelitis affected the right anterior hemithorax with normal biological findings. Bacteriological samples were taken pre- or intraoperatively in all patients, except in patients with sternal deformity. Cultures isolated a germ in 8 cases (40%), one of which was *Staphylococcus aureus* in 7 cases and one enterobacteria in 1 case. The other 12 samples were sterile.

Table 1
Swelling location.

Swelling location	N
Presternal swelling with hyperthermia and pain	6
Scapular swelling with hyperthermia and pain	4
Scalp swelling with hyperthermia and pain	2
Presternal abscess fistulisation	2
Buttocks swelling with hyperthermia and pain	2
Swelling with hyperthermia and pain of the anterior face of the knee	2
Firm and painless mass of the anterior face of the right hemithorax	1
Painless deformity of the sternum	1
Total	**20**

- Imaging

The standard radiography strengthened the diagnosis in 19 cases, showing either osteolytic lesions or sclerogeodic lesions (Figs. 1–3). One patient had a pseudotumor of the anterior chest wall with a near-normal radiograph; The MRI provided more exact anatomic localization of osteomyelitis in the anterior arch of the 7^{th} rib (Fig. 4). The above-described clinical and radiographic elements made it possible to establish the diagnosis of osteomyelitis of a flat bone. Table 2 describes its different locations.

Sternal osteomyelitis was predominant in children under 5 years of age (5 out of 7 children had sickle cell disease), while other bone locations were seen in older children and adolescents (Table 3).

- Other associated infectious locations

In the majority of cases, osteomyelitis of a flat bone was diagnosed; only 4 sickle cell patients who had sternal osteomyelitis presented a progression of infection towards phalangeal osteomyelitis, tibial osteomyelitis and pyothorax and bilateral septic arthritis of the hip. An 11-year-old homozygous sickle cell patient presented skull and patellar osteomyelitis.

1.2.2. Treatment

The treatment consisted of a probabilistic parenteral antibiotic with anti-staphylococcal Oxacillin or cephalosporin of 2nd or 3rd generation for 10–14 days, associated with an aminoglycoside; with oral relay after 4–6 weeks depending on the outcome except for 3 sickle cell patients whose clinical outcome was marked by persistence of fever and/or suppuration, thus justifying longer duration of treatment and hospitalization. These were the 2 cases of pelvic osteomyelitis and one case skull and patellar osteomyelitis. Consequently, after identification of the infective bacteria,

Fig. 1. Osteolytic lesion of the patella in a 7-year-old boy.

Fig. 2. Disseminated multiple geodes of the iliac bones and proximal femoral epiphyses in a 9-year-old girl with sickle cell disease.

Fig. 3. Sternal nuclei geodes with subluxation and osteonecrosis of the 3rd sternal nucleus in a 5-year-old girl with sickle cell disease.

antibiogram-guided antibiotherapy seemed to be the most reasonable treatment option.

Almost every patient was received at the stage of abscessing, which required debridement, and when needed, bone curettage or bone trepanation under general anaesthesia. There was only one case of a 5-year-old girl with an osteomyelitis with sternal deformity due to a septic dislocation of the sternal arches (Fig. 1). There was no abscess, and the patient had not undergone surgery.

Table 2
Distribution of the different osteomyelitis topographies of flat bones in relation to all osteomyelitis (N = 220).

Topographies	n	%
Sternum	8	3,6
Scapula	4	1,8
Skull	3	1,3
Patella	2	0,9
Pelvis	2	0,9
Rib	1	0,5
Total	**20**	**10**

Table 3
Patient distribution of osteomyelitis topographies according to age.

Age (years old)	0–5	5-10	10–16	Total
Sternum	7	1	-	8
Scapula	1	–	3	4
Skull	-	1	1	3
Patella	-	1	1	2
Pelvis	-	1	1	2
Rib	-	1	-	1
Total	8	5	7	20

Functional impotence of pelvic limb was found in patients with scapular, pelvic and patellar osteomyelitis.

1.2.3. Evolution

The average duration of hospitalization was 20 days (10–60 days). There was a successful outcome in 15 cases with bone reconstruction ad integrum (average follow-up of 3.3 years). Stiffness of the ipsilateral hip was present in 2 of our patients who presented with osteomyelitis of the iliac bone. The treatment was traction and kinesitherapy. Two patients were lost to follow-up.

1.3. Discussion

1.3.1. General aspects

Osteomyelitis of flat bones is a rare disease. In our study, it represented 9,1% of all osteomyelitis. We have found a predominance of children with sickle cell disease, the association between sickle cell disease and osteomyelitis being well established.[4,11,12]

Although osteomyelitis is most often seeded hematogenously, some authors[7,13] point out the possibility of traumatic origin; in our series, we had 2 cases of post-traumatic osteomyelitis: scapular and

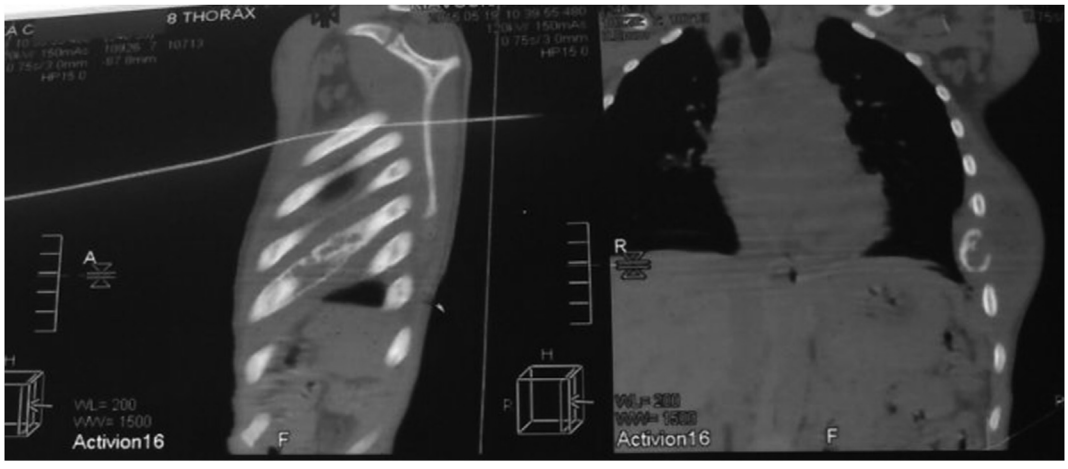

Fig. 4. Osteolytic lesion of the anterior arch of the 7th right rib with soft tissue swelling in a 10-year-old boy without sickle cell disease.

patellar osteomyelitis. This could be explained by the fact that these bones are superficial and therefore exposed to trauma.[7]

Our clinical findings were similar to what many authors have reported[1,8]: first, localized pain, hyperthermia, followed by local inflammatory swelling due to abscess; laboratory findings suggestive of biological inflammatory syndrome.[1,8] In some cases, the clinical and biological inflammatory syndrome are absent.[7,13] In our study, nearly all patients were admitted at the stage of abscess.

In our study, a micro-organism was isolated in 8 cases out of 20 bacteriological findings because some germs are not always identified; according to some authors, culture is negative in 22–50% of cases.[1] Eleven cultures were negative in our series. This can be explained by the fact that the majority of patients came from other paediatric units and have been treated with antibiotic prior to admission. *Staphylococcus aureus* was found in 7 cases including one multi-resistant staphylococcus. It is the most commonly implicated germ[1–3] and identified in about 40% of osteomyelitis cases according to the literature.[14] However, other germs are also incriminated.[15,16] In almost half of children with acute osteomyelitis, a bacterial etiology is never established,[17] the germ has been identified in 40% of cases in our study.

The medical imagery is very helpful in the diagnosis of osteoarticular infections.[18] Standard radiography is systematic and may be sufficient to make the diagnosis at an advanced stage, which has been the case for the majority of our patients. MRI is not systematic, but frequently used in case of deep abscess (pelvis, spine), in case of risk of epiphysiodesis, to analyse the extension of a chronic osteomyelitis, and in case of suspicion of osteomyelitis with normal radiographs. This last indication was found in the patient who had rib osteomyelitis with pseudotumoral aspect without biological inflammatory syndrome. The CT scan has few indications[18]: it is carried out in search of a bone sequestrum in chronic forms and in case of impossibility of access to MRI in difficult anatomical areas (spine, pelvis, scapula). Scintigraphy is only performed in the case of normal radiographs and absence of a clinical symptoms.[18]

1.3.2. Specific aspects according to the location

Sternal osteomyelitis is the most common location in our series with 3.6% of all osteomyelitis. In the literature, its frequency is estimated at 0.3% of all osteomyelitis in children.[19] Children under 5 years old were the most affected. Most often it is primitive, but may also be secondary to mediastinitis or pyothorax.[9,19,20] In our series, it was primitive in 7 cases and secondary to a pyothorax in 1 case. The clinical picture includes pre-sternal pain and fever; at the stage of abscessing, there is an inflammatory swelling over the sternum. In all cases, standard radiographs were sufficient to make the diagnosis by showing osteolysis of the sternal nuclei.

Scapular osteomyelitis comes in second place after sternal osteomyelitis in our series with 1.8% of cases. According to the literature, its frequency ranges from 0.5% to 2.6%.[21,22] In our series, teenagers were the most affected (13–14 years) with no case of sickle cell disease. The clinical features vary according to the stage of evolution: at the beginning, there is a simple pain with painful mobilization of the shoulder being able to simulate a septic arthritis of the shoulder, then a swelling well localized with the scapular region being able s extend to the ipsilateral shoulder with scoliosis.[21] Our patients were all admitted at the stage of abscessing. A drainage of the pus was performed. The standard radiography had shown sclerosis and geodic images of the scapula, and even in some cases with glenohumeral lesions testifying the chronicity of the disease.

Skull osteomyelitis, even rarer with 1.3% of cases of all osteomyelitis, is often secondary to postoperative infections (craniotomy, external malignant otitis, chronic mastoiditis and sphenoidal sinusitis).[23] However, it may also be favoured by systemic diseases by decreasing the vascularity of the bone such as sickle cell disease, or by modifying the host's defence mechanism.[24] In our series, 2 patients who presented skull osteomyelitis are sickle cell patients. One had only one location (skull osteomyelitis), while the other had 2 locations (skull and patellar osteomyelitis). Clinically, both patients had swelling of the scalp with inflammatory symptoms. A drainage of the pus was performed. Standard radiography helped to make the diagnosis by showing osteolytic images of the skull.

Pelvic osteomyelitis represents 0.9% of cases in our series. It is a severe condition with an incidence varying from 6.3% to 20% according to the literature.[1,25,26] The clinical picture is consisted of pain, fever, lameness or functional impotence of the pelvic limb. However, the diagnosis is not easy because the symptoms are not specific; the pain may be of variable locations including the hip, buttock or even the abdomen respectively in 58%, 31% and 11% of cases.[26] Depth and possible locations of infection help to mislead or delay diagnosis.[25,26] Also, two of our patients presented different symptoms. The first had a clinical picture of septic arthritis of the hip and the second a gluteal abscess. In both cases it is in front of persistent symptoms in spite of a well conducted treatment that we performed x-rays which showed sclerogeodic images localized in the iliac bone. Its understandable that according to Weber-Chrysochoou et al.,[26] the iliac bone is most affected with 38% of cases, followed by ischium with 19%, pubis 14% and acetabulum with 12%.

Patellar osteomyelitis is an even rarer condition with 0.9% of cases. The age of onset ranges between 5 and 15 years old according to the literature,[7,27,28] which is the case in two of our patients who are 7 and 11 years old. The notion of trauma reported by some authors[7,27] was found in one of our patients who had cutaneous excoriations next to the knee following a fall. One sickle cell patient had suppurative septic knee osteoarthritis, while the second had a prepatellar inflammatory swelling with clear citrine content. During patellar osteomyelitis knee effusions can be bacterial or reactional; and may be associated with septic arthritis of the knee.[7,27] The standard radiography was sufficient for the diagnosis by showing, in both cases, geodic lesions with irregular patellar edges.

Rib osteomyelitis represents 0.5% of cases in our series, and less than 1% of all osteomyelitis in the literature.[29] It usually manifests by hyperthermia associated with an abscess and/or persistent fistula in the chest.[30] Our patient presented a pseudotumoral mass of the anterior face of the right hemithorax. We hypothesized a tumour and performed an MRI that showed osteolytic lesions related to osteomyelitis with abscess from the anterior arch of the 7th right rib. A similar case to ours has been reported by Basa et al.[30]: in front of a tumoral mass sitting beneath the right breast in a 2-year-old girl, a biopsy is indicated; intraoperatively, they found an abscess and concluded osteomyelitis.

1.3.3. Treatment

Therapeutically, the treatment of osteomyelitis of flat bones obeys the same rules as that of osteomyelitis of long bones.[21,22] The patient is treated with antibiotics directed against Cocci Gram positive by the parenteral route to reach rapidly high serum concentrations. We used a bi-antibiotherapy: Oxacillin or cephalosporin 2nd or 3rd generation associated with gentamicin (up to the regression of clinical and/or biological inflammatory syndrome); then an oral relay with an antibiotic of the same class after 4–6 weeks. Surgical treatment consists of an incision and drainage of the abscess with bone curettage, sometimes bone trepanning in some cases. Resting was required for scapular, pelvic and patellar locations osteomyelitis.

1.3.4. Evolution

We had a successful outcome in 15 patients with bone

reconstruction ad integrum. Therefore, when the diagnosis is early established with appropriate management, the evolution is often favourable as reported by the majority of authors[1,20,21]

2. Conclusion

Flat bones are rare sites of osteomyelitis. At an advanced stage, standard radiography remains the first-line examination. Their treatment obeys the same rules as those of osteomyelitis of long bones. The evolution is often favourable when the care is early and adequate.

Conflicts of interest

The authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jcot.2019.03.022.

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