Case report

Subperiosteal abscess in a child. Trueta's osteomyelitis hypothesis undermined?

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ABSTRACT

Subperiosteal abscess formation is almost exclusively seen secondary to underlying hematogenous infected osteomyelitis or secondary as a result of a contagious focus. We present an unusual case of a 9-year-old girl with progressive ankle pain due to an isolated subperiosteal abscess of the distal fibula without concomitant osteomyelitis. The subperiosteal abscess was most likely caused by hematogenous spread to the periosteal region of the distal fibula located above the highly vascularized metaphysis. Remarkably, there were no signs of osteomyelitis on either MRI or during surgical inspection. She was successfully treated with debridement and antibiotic therapy. We hypothesize that subperiosteal abscess formation near the metaphysis originates in the periosteal region and not from outward extension from the sinusoidal veins in the intrametaphyseal area to the cortex and subperiosteal region.

1. Introduction

Pyogenic osteomyelitis leads to inflammation of the medullary cavity, cortical bone and periosteum [1]. Sometimes, subperiosteal abscess formation is observed, which is regarded as a rare complication [1]. In children, the incidence of osteomyelitis complicated by subperiosteal abscess is higher in comparison with adults [2].

Subperiosteal infection is observed in cases of pyogenic osteomyelitis and is thought to result from an outward spread from inside the cortex or as a complication of direct inoculation from trauma or surgery [1]. The leading hypothesis on the pathogenesis of osteomyelitis and subsequent subperiosteal abscess formation is based on the landmark work of Trueta published in 1959. Even today, the Trueta hypothesis is the leading hypothesis for the underlying pathogenesis of osteomyelitis [3,4]. Trueta showed that the age dependent differences in vascular arrangement of the epiphyseal region are responsible for the variation in clinical presentation between different ages [3].

We present a case of a 9-year-old Caucasian girl who developed an isolated subperiosteal abscess of the distal fibula most likely caused by hematogenous spread without evidence of a trauma or skin infection and without concomitant osteomyelitis. We will discuss recent pathophysiological mechanisms concerning the origin of infective osteomyelitis and subperiosteal abscess formation.

Furthermore, we discuss new insight that challenges the current explaining hypothesis, called Trueta’s hypothesis, for explaining the pathophysiology of subperiosteal abscess formation in children.

2. Case report

A 9-year-old Caucasian girl was presented to the emergency department with a two-day of progressive ankle pain and she was unable to walk or bear weight due to severe pain. There was no history of trauma or arthritis. On examination, she had a fever up to 38.3 °C. There was a swelling located 5 cm proximal from the tip of the right lateral malleolus extending to the dorsum of the foot.

Laboratory results showed an erythrocyte sedimentation rate (ESR) 27 mm/hour, a serum C-reactive protein (CRP) of 141 mg/L, and leukocytes 11.5/nanoliter. Conventional X-ray showed no abnormalities. Ultrasound examination revealed a 5-cm subperiosteal fluid collection extending to the area above the cortex of the epiphyseal plate (Fig. 1).

After incision and drainage of the abscess, the cortex above the epiphysis and metaphyseal area of the fibula were intact. Furthermore, there was no weakening of the bone, tested through applying local pressure with a pair of forceps. The subperiosteal abscess of the distal fibula was evacuated and the periosteum was left open, to allow for further draining and thus preventing...
new abscess formation. Perioperative cultures of the subperiosteal abscess grew methicillin sensitive Staphylococcus aureus. Blood cultures showed no growth. We performed no histopathological analysis of the bone because the bone was intact. Postoperatively intravenous flucloxacillin (100 mg/kg/day) was administered for 2 weeks followed by 4 weeks clindamycin (oral 25 mg/kg/day). An additional magnetic resonance imaging (MRI) scan was made directly after the operation to confirm that there were no signs of osteomyelitis (Fig. 2). She was discharged after 5 days and full weight bearing was allowed together with physical therapy training. After 6 months, she visited the outpatient clinic; there was no functional impairment or pain and a conventional X-ray showed a normal growth plate.

3. Discussion

This case was remarkable for the isolated subperiosteal abscess formation located at the distal fibula without evidence for underlying osteomyelitis or contiguous spread [4,5]. In general, most case reports describe subperiosteal abscesses as a complication of sinusitis, the so-called Pott’s Puffy tumor [6]. To our knowledge, very few cases of isolated subperiosteal abscess formation of the distal fibula have been described [4,5]. The fact that the subperiosteal abscess developed without osteomyelitis, questions the Trueta hypothesis which dictates that the origin of osteomyelitis is located within the cortex.

Fig. 1. Ultrasound of the right ankle showing a subperiosteal abscess.

4. The classic pathogenesis of osteomyelitis and subsequent subperiosteal abscess formation

The pathogenesis of osteomyelitis and subsequent subperiosteal abscess formation is known as Trueta’s hypothesis [3,4]. Trueta showed that the clinical features of osteomyelitis are different according to the different age groups which results from differences in vascularity between infants, children and adults [3]. In children, vascularisation of the growth plate and epiphysis consists of arteries from the epiphyseal and metaphyseal region. Inside the metaphyseal area of the distal fibula, capillaries and sinusoidal veins vascularize the proximal area of the growth plate and support ossification of the growth plate. During this ossification of the growth plate, metaphyseal capillaries atrophy. Capillaries adjacent to the growth plate, at the metaphyseal side, end as so-called capillary loops which together form a system of large sinusoidal veins responsible for haemopoietic activity [3]. According to the Trueta hypothesis, the infection of bone presumably starts in the metaphyseal sinusoidal veins, where blood flow is slow and abundant and the cortex the thinnest. Infection in the metaphysis is contained by the growth plate. This is probably the reason that the joint is most often spared. Trueta’s hypothesis dictates that hematogenous infected bone originates at metaphyseal sinus and spreads laterally, subsequently breaching through the cortex and lifting the loose periosteum to form a subperiosteal abscess [2,3].

5. A new theory?

In 2010, a new theory of the pathogenesis of acute osteomyelitis was postulated by Labbé et al. [4]. In a case series of over 450 children from the area of New Caledonia with osteomyelitis, the fibula was affected in 7.2% of cases. All children underwent ultrasonography to determine the presence and size of periosteal elevation. In this study, inflammation of the periosteum and subsequent abscess formation was observed in 84% of cases by the second day of admission. These results indicated that the origin of osteomyelitis was frequently located subperiosteal instead of intrametaphyseal [4]. This new hypothesis is more in line with our observation. We think that this case represents the first phase of developing osteomyelitis in which the infection started in the most vascularized part of the fibula and that if left untreated, this subperiosteal abscess would have resulted in osteomyelitis. Labbé et al. also emphasized that trauma of the periosteum can induce edema or hematoma and that the combination of subsequent bacteriaemia and the high vascularization of the periosteum lead to infection [4]. This new hypothesis is different from Trueta’s hypothesis, with the idea that the softer cortex and intrametaphyseal septic diffusion into the cortex outwards are the explanation for the reason why subperiosteal abscesses are almost exclusively have been observed in children [4] (Table 1).

In this case, the indication for surgical intervention was based on the subperiosteal abscess seen on ultrasound. These results emphasize their importance of excluding the presence of subperiosteal abscess with ultrasonography. Where there is debate on the optimal timing and extent of surgical interventions other than biopsy in isolated osteomyelitis [7], in case of subperiosteal abscess formation, there is a general consensus for early surgical debridement [8–11].

We acknowledge that our interpretation of the pathogenesis of infection in this girl is based on the idea of hematogenous spread and bacteriaemia and we could not exclude direct inoculation as the cause, although in this case, there was no history of trauma or skin lacerations.

We performed an MRI after the operation. This is a limitation of our study and we regret the difficult interpretation of the MRI image.
Table 1

<table>
<thead>
<tr>
<th>Route of infection</th>
<th>Trueta</th>
<th>Labbé</th>
</tr>
</thead>
<tbody>
<tr>
<td>Origin of infection</td>
<td>Internal metaphyseal route</td>
<td>External periosteal route</td>
</tr>
<tr>
<td>Role of age</td>
<td>Infection develops in capillary loops</td>
<td>Infection starts in the periosteal area</td>
</tr>
<tr>
<td>Role of trauma</td>
<td>Not described</td>
<td>Direct bone trauma may cause hematoma and secondary infection</td>
</tr>
<tr>
<td>Role of septicemia</td>
<td>Septicemia causes seeding of bacteria that get stuck in capillary loops</td>
<td>Severe septicemia may cause seeding of bacteria and secondary infection of hematoma</td>
</tr>
<tr>
<td>Subperiosteal abscess formation</td>
<td>Edema causes expansion towards the surface of the bone across the cortex which raises the periosteum from the cortex</td>
<td>Periosteal inflammation with subsequent subperiosteal abscess formation</td>
</tr>
</tbody>
</table>

6. Conclusion

We hypothesize that this case of isolated subperiosteal abscess in a child represents the first phase of a developing osteomyelitis with the origin of infection in the most vascularized part of the fibula. If left untreated, the subperiosteal abscess probably would have progressed to osteomyelitis. These results are confirmatory to a new theory postulated by Labbé and oppose the leading hypothesis made by Trueta in 1959. Whether osteomyelitis in children originates intrametaphyseal from hematogenous spread from sinusoid veins or from hematogenous spread through trauma induced subperiosteal edema or hematoma is still not clear. The classic Trueta hypothesis does not support recent observations in children such as our case. Early surgical intervention is warranted in the case of subperiosteal abscesses. It is therefore important to investigate the presence of a subperiosteal abscess when the diagnosis of osteomyelitis in children is made.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References