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Case Report

Osteochondroma after Acute Skeletal Infection: a Primary or a Secondary Lesion?

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Abstract

Secondary or acquired osteochondroma may develop following radiation exposure, trauma or surgery, and osteomyelitis. This report presents a 15-year-old patient with an osteochondroma of the anterolateral left distal tibial metaphysis, who received surgical treatment for an acute suppurative arthritis of the left ankle joint at the age of 4 years. The purpose of this paper is to present the challenging diagnostic enigma of the osteochondroma's etiology in our patient between a primary versus a secondary lesion. After analysing retrospectively the available information in the patient's files, we concluded that the osteochondroma was likely a primary lesion with modified presentation due to infection.

Keywords

primary, secondary, osteochondroma, osteomyelitis, septic arthritis

INTRODUCTION

The categorisation of solitary osteochondromas is according to the etiology into a primary (congenital) and a secondary (acquired) lesion. The primary osteochondroma develops spontaneously with no precipitating event, while the secondary one may develop due to childhood radiation exposure[1-4], trauma[5-7] or surgery[8,9], and osteomyelitis[10,11].

This paper focuses on distinguishing between a primary and a secondary etiology of an osteochondroma of the distal tibial metaphysis in a 15-year-old patient who had been treated for acute suppurative arthritis of the ipsilateral ankle joint at the age of 4 years. The clinical and radiographic findings are presented and discussed.

CASE REPORT

A 15-year-old boy was evaluated for a long history of swelling on the anterolateral side of his left distal tibia. Physical examination revealed a palpable bony mass with no tenderness. A hypertrophic scar was evident overlying the bone lesion ***(Fig. 1)****. His parents reported that he had been treated surgically for acute septic arthritis of the left ankle joint 11 years ago. Plain radiographs showed a broad osseous excrescence along the distal anterolateral left tibial metaphysis, showing cortical and medullary continuity with the underlying bone. The cortex was smooth, without evidence of disruption or erosion, and no periosteal reaction was present* ***(Fig. 2)****. Two-dimensional computed tomography* ***(Fig. 3)*** *and three-dimensional reconstruction views* ***(Fig. 4)*** *were obtained. They also indicated continuity between the marrow space of the underlying bone and that of the bone lesion as well as no irregular calcification, representing characteristics consistent with a typical osteochondroma. The base of the lesion was calculated to be approximately 20×60 mm, while the maximum dimensions in the coronal and sagittal planes were almost 85 mm and 50 mm, respectively. Under general anesthesia, excision of the bone lesion was performed through an anterolateral incision. Surgical excision at its*

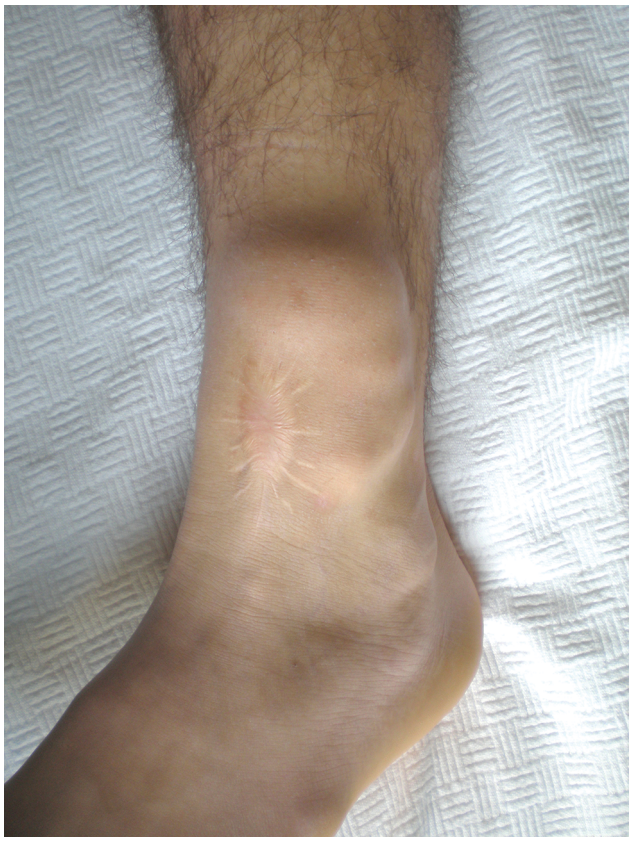

Figure 1. Clinical appearance of the patient. The deformity was related to an enlarging exophytic mass. A hypertrophic scar was evident overlying the lesion.

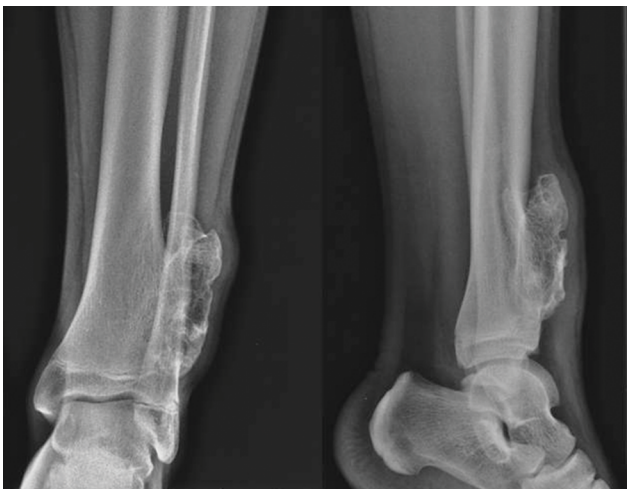

Figure 2. Anteroposterior and lateral radiographs demonstrated a well-defined bone lesion with a broad area of osseous continuity with the parent bone, protruding from the anterolateral distal tibial metaphysis and growing away from the ankle joint. The tumour was composed of cortical and medullary bone, which was in continuity with the tibial marrow cavity.

*base was uncomplicated **(Fig. 5)**. Histological examination revealed typical features of an osteochondroma. No complications were detected in the long-term follow-up of our patient.*

His previous medical record was retrospectively reviewed. It *indicated that the patient at the age of 4 was diagnosed with acute febrile exudative tonsillitis (T max=39°C) and treated with cefprozil monohydrate. He suffered from a painful, acutely swollen left ankle 6 days later. There was limited motion of the ankle joint and the child refused weight-bearing. His parents reported an injury to the left ankle about a week ago. The clinical findings on admission included high fever (T max=40.3°C), left ankle joint swelling, tenderness, local heat, and decreased range of motion. His laboratory studies indicated a raised erythrocyte sedimentation rate of 72 mm per the first hour (normal values 1–20) and an increased serum C-reactive protein value of 6.9 mg/dl (normal value <5) with a white blood cell count of 18.800/mm^3 (normal values 5–15.5×10^9 cells/L), (neutrophils: 60%, lymphocytes: 30.8%, monocytes: 4.2%, eosinophils: 3.6%). His left ankle radiographs on admission indicated no abnormal findings other than a lytic lesion on the anterolateral portion of the distal tibial metaphysis. The lytic lesion was surrounded by a thin sclerotic margin **(Fig. 6)**. A MRI examination was not accepted by his parents because it required transfer to another hospital and sedation. No clinical findings related to a bone protuberance from the distal tibial metaphysis were recorded on his notes during admission. An acute septic arthritis of the ankle joint was diagnosed, which was potentially associated with osteomyelitis of the distal tibial metaphysis. Blood cultures were not obtained. He was treated with an open arthrotomy, drainage of the purulent whitish synovial fluid and lavage of the joint. There were no abnormal findings of the distal tibia to necessitate periosteal stripping and surgical drilling of the metaphyseal bone. The synovial fluid showed high viscosity (>75.000 cells/μl, mostly polymorphonuclear). Histopathology findings were indicative of septic synovitis. The cultures of the joint fluid were negative, most likely due to the preoperative intake of antibiotics. Empiric coverage with intravenous ceftriaxone started and the ankle was immobilised in a below-knee splint. The postsurgical course of the disease was uneventful; the patient was discharged from the hospital without any complications. Full recovery was detected after an 18-month follow-up.*

DISCUSSION

Two case reports have only been encountered so far in the world literature reporting an osteochondroma resulting from acute skeletal infection. The first published case was about a 10-month-old infant suffering from osteomyelitis of the distal third of the femur, who developed an osteochondroma at that site after a subperiosteal abscess had been drained and a cortical window produced.[10] The second case was a premature newborn (gestational age: 28 weeks) with neonatal osteomyelitis of the distal metaphyses of the radius and ulna, treated with drainage of the abscess, and diagnosed with a radial metaphyseal osteochondroma 7 months later.[11]

The etiology of osteochondroma resulting from osteomyelitis is debated. Several reasonable hypotheses have

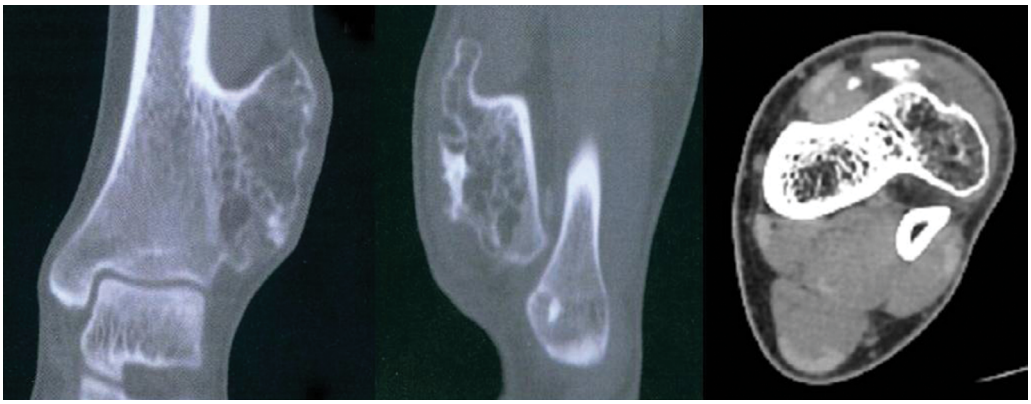

Figure 3. Two-dimensional computed tomography views of the distal tibia at sagittal, coronal, and transverse (axial) planes revealed marrow and cortical continuity between the tumour and the tibia.

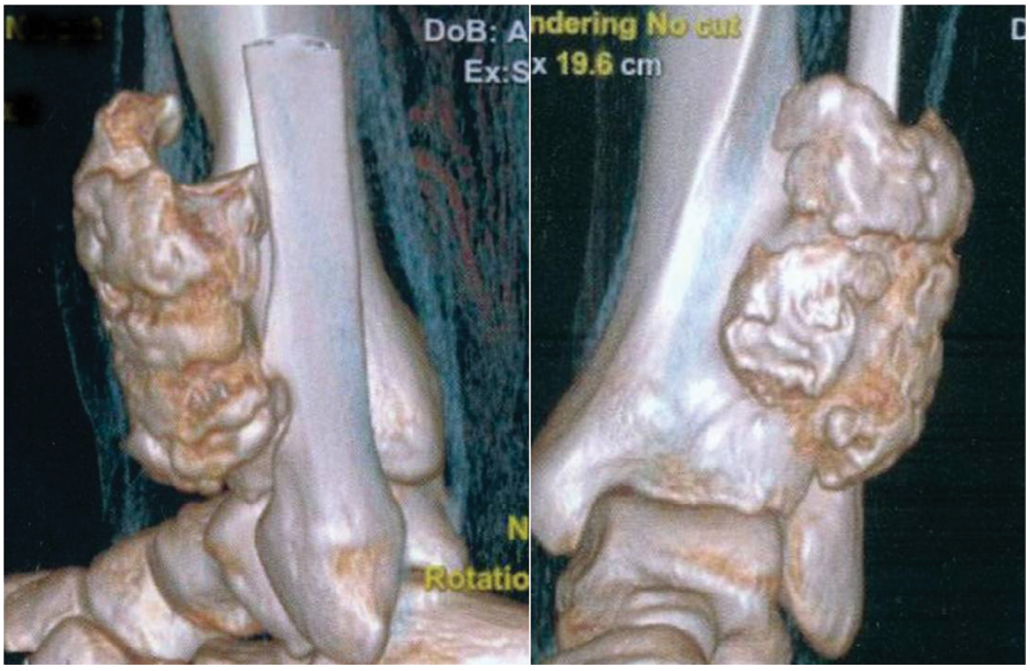

Figure 4. Three-dimensional reconstructed cone beam computed tomography images of the lesion. A cortical pressure indentation on the anterior portion of the distal fibular metaphysis resulting from the bone lesion was noted.

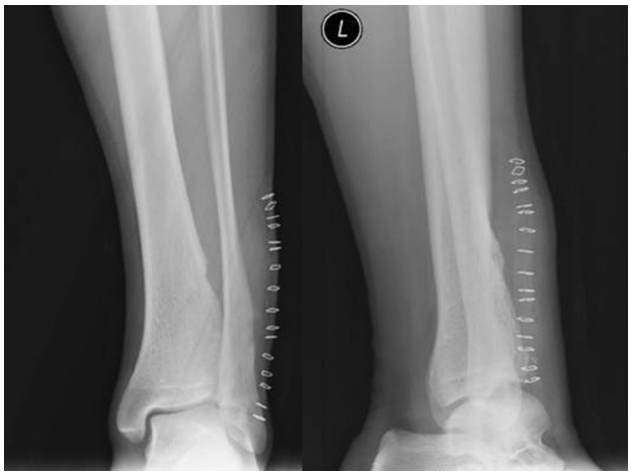

Figure 5. Post-operative anteroposterior and lateral radiographs.

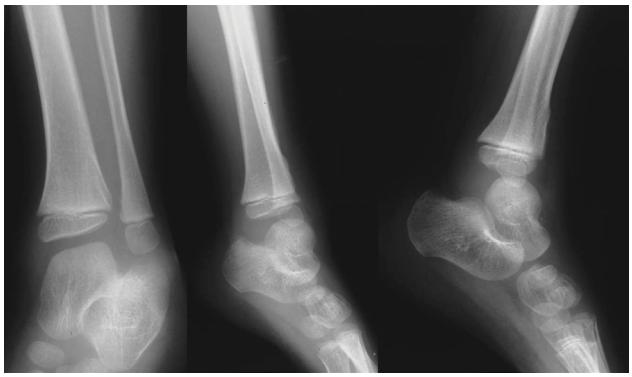

Figure 6. Plain anteroposterior, oblique, and lateral radiographs on admission at 4 years of age. A lytic lesion surrounded by a thin sclerotic margin was noted on the anterolateral portion of the distal tibial metaphysis.

been proposed regarding this origin. It has been confirmed that experimental ischemia could provoke injury to the perichondrial ring in the area corresponding to Ranvier's groove with the production of an osteochondroma.[12,13] In addition, it has been shown that proteolytic enzymes are generated by the infectious process, which may also be responsible for the destruction of the perichondrial ossification groove of Ranvier.[14,15] The severity of the infection and the prematurity of the one case were considered as presumably important factors in the final poor outcome. In both cases, a potential pathogenesis of the osteochondromas could be physeal plate injury either due to iatrogenic trauma during drainage of the abscess or due to the associated inflammatory process.[11]

At our institution, we have treated only a single patient with an osteochondroma, who was diagnosed with a previous acute skeletal infection at the same site. The purpose of this paper was to consider the primary or secondary nature of the osteochondroma in our patient.

The radiographic appearance of a small metaphyseal lucent defect, within the anterolateral cortex of the distal tibial metaphysis, surrounded by a thin rim of sclerosis, with no periosteal reaction and no involvement of the underlying medullary cavity, in our patient indicated several differential diagnoses. The most likely bone tumour was a fibrous cortical defect. All potential diagnoses of a bone tumour may be securely excluded retrospectively since the long-term follow-up in our patient indicated the subsequent development of an osteochondroma with an identical localisation with the lytic lesion. However, it could be a true matter of divergence whether the lytic lesion is consistent with the early phase of the sessile osteochondroma that subsequently developed in our patient or whether it was due to an acute hematogenous bone infection.

It has already been shown that although acute septic arthritis has a predilection towards children under the age of 4 years, the foot and ankle are uncommon sites of bone and joint infections in children.[16] In addition, the frequency of coexisting unsuspected osteomyelitis in children suffering from acute septic arthritis has been indicated, emphasising the importance of diagnosing concurrent osteomyelitis in patients with acute septic arthritis to avoid delay or undertreatment.[17-20]

Therefore, it would be reasonable that the diagnosis of a coexisting osteomyelitis of the distal tibial metaphysis should have also been seriously considered in our patient during his admission. However, the apparent thin sclerotic rim surrounding the lytic lesion was not consistent with the diagnosis of acute hematogenous osteomyelitis. The lack of periosteal reaction was not a valuable radiographic evidence to exclude acute bone infection since it may take a couple of weeks to appear following the acute phase of osteomyelitis. On the other hand, the lack of a surgical manipulation to the distal tibial physeal plate and metaphysis would exclude a primary iatrogenic damage to the physeal plate or a secondary one from stripping of the periosteum, which could be responsible for initiating the growth of an osteochondroma.

These data suggest that the formation of the osteochondroma pre-existed to the acute suppurative arthritis of the ankle joint in our patient, indicating a primary osteochondroma rather than a secondary lesion. The radiographic appearance of an osteochondroma may be obscure in children younger than 4 years of age, leading to a demanding radiographic diagnosis. However, the effect of an acute bone or joint infection on the size of a coexisting osteochondroma may be significant due to the local increased blood supply. Therefore, the volume of the osteochondroma in our patient could have been severely affected by the presence of the acute septic arthritis of the ankle joint and the subsequent surgical treatment. In conclusion, it may be prudent to consider that our patient suffered from a primary osteochondroma with modified presentation due to the local acute skeletal infection.

Conflict of Interest

The author certifies that he has no commercial associations (such as consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted article. The author received no financial support for this study.

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Остеохондрома после острой скелетной инфекции: первичное или вторичное поражение?

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Резюме

Вторичная или приобретённая остеохондрома может развиться после радиационного облучения, травмы или хирургического вмешательства, а также остеомиелита. В данном сообщении представлена больная 15 лет с остеохондромой переднелатерального дистального метафиза большеберцовой кости слева, перенёсшая хирургическое лечение по поводу острого гнойного артрита левого голеностопного сустава в возрасте 4 лет. Цель этой статьи – представить сложную диагностическую загадку этиологии остеохондромы у нашего пациента между первичным и вторичным поражением. Проанализировав ретроспективно доступную информацию в документации больной, мы пришли к выводу, что остеохондрома, вероятно, была первичным поражением с изменённым представлением из-за инфекции.

Ключевые слова

первичный, вторичный, остеохондрома, остеомиелит, септический артрит