Bursal osteochondromatosis overlaying an osteochondroma in the immature skeleton

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A rare case of bursal osteochondromatosis overlying an osteochondroma located at the proximal third of the left tibia in a 10-year-old boy is described. The treatment was operative by marginal resection of the affected bursa and marginal en bloc resection of the osteochondroma. The patient remains symptom and complication free 8 years postoperatively with no sign of recurrence of both lesions. This is the first reported case of a bursal osteochondromatosis overlying an osteochondroma in a developing skeleton, thus rendering this type of lesion one among the possible differential diagnoses that should be considered when dealing with relative diagnostic dilemmas in immature patients. *J Pediatr Orthop B* 16:160–163

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Introduction

Synovial osteochondromatosis (SOC) is an uncommon metaplastic disorder, involving articular or tendon sheath synovial membranes, which is characterized by the formation of multiple cartilaginous nodules within the synovium. The extra-articular manifestation of SOC is rare and it is known as 'bursal chondromatosis' or 'bursal osteochondromatosis'. The development of an osteochondromatosis in a bursa overlying an osteochondroma is even rarer and it has only been described in adults. This is the first report, according to the best of our knowledge, of a bursal osteochondromatosis overlying an osteochondroma in an immature, developing skeleton. Therefore this lesion should be considered as one among the possible differential diagnoses in younger patients and in adults.

Case report

The patient was a 10-year-old boy, son of a 40-year-old Caucasian woman and a 43-year-old Caucasian man with no significant medical history. He presented at our outpatient's clinic complaining of a painful swelling over the anteromedial aspect of the proximal third of the metaphysis of his right tibia for the last 18 months. The mass came to his attention because of the dull pain that was elicited upon palpation. Since the initial discovery of the lesion, the pain gradually deteriorated and during the last 6 months its characteristics changed as it became more sharp and spontaneous with long periods of self-remission. Two months before his initial medical examination, the patient suffered an injury (a direct kick) exactly at the lesion's area during a soccer game. After this injury, the pain rapidly got worse and forced the patient to seek medical attention.

The physical examination revealed the existence of a well defined, multilobular, circumscribed, palpable mass located at the area of pes anserinous. The mass was approximately 3 cm × 5 cm, and its palpation provoked slight discomfort to the patient. Unrestricted, painless and full range motion of the knee joint, both passive and active was achievable. No varus or valgus deformity was noticed. The standard anteroposterior and lateral radiographic examination of the affected knee (Fig. 1) revealed the existence of a bony projection arising from the medial surface of the proximal metaphysis of the tibia. The radiographic features of this lesion (well defined metaphyseal excrescence of bone with a mottled density) were consistent with that of an osteochondroma. Over this lesion, however, were noted a number of ossified nodules in the adjacent soft tissue, that probably belonged to the more superficial mass.

The treatment was surgical. During the operation, a bursal sac overlying a bony lump was noticed. The bursa was entirely extra-articular, and it was denucleated quite easily, and in one piece, from the surrounding soft tissues. The bone lesion was resected (marginal en bloc resection) with the use of a bone chisel. The opening of the bursa revealed the existence of a cavity. Multiple pedunculated nodules (ranging from 4 to 15 mm in diameter) were attached on the inner wall of the cavity (Fig. 2). The pathologic examination of the specimens that were excised confirmed the originally set diagnosis of

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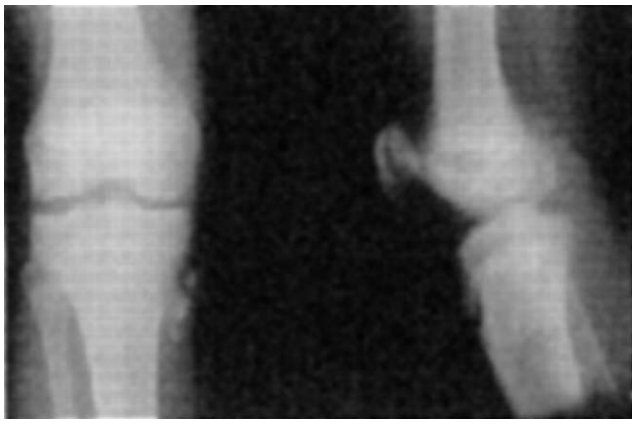
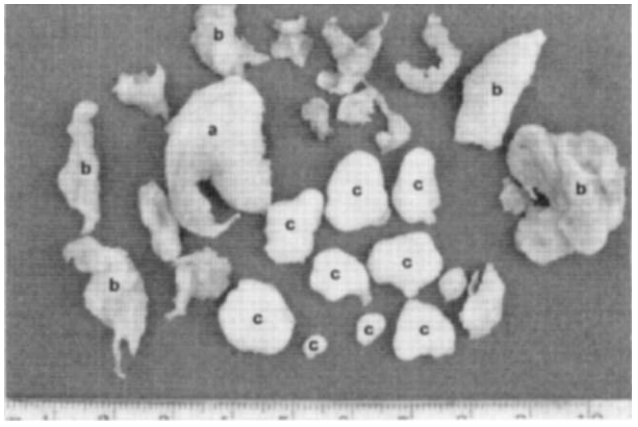
Fig. 1



Anteroposterior and lateral plain radiographs of the knee and the upper tibial metaphysis showing a bony projection arising from the medial surface of the tibial metaphysis with a number of small ossified nodules in the adjacent overlying soft tissue.

Fig. 2



The resected specimens: (a) the exostosis, (b) the bursal wall, and (c) some of the osteocartilaginous nodules that were found inside the bursa cavity.

a bursal osteochondromatosis overlying an osteochondroma (Figs 3 and 4). The patient remains symptom and complication free 8 years postoperatively with no sign of recurrence of either lesion (Fig. 5).

Discussion

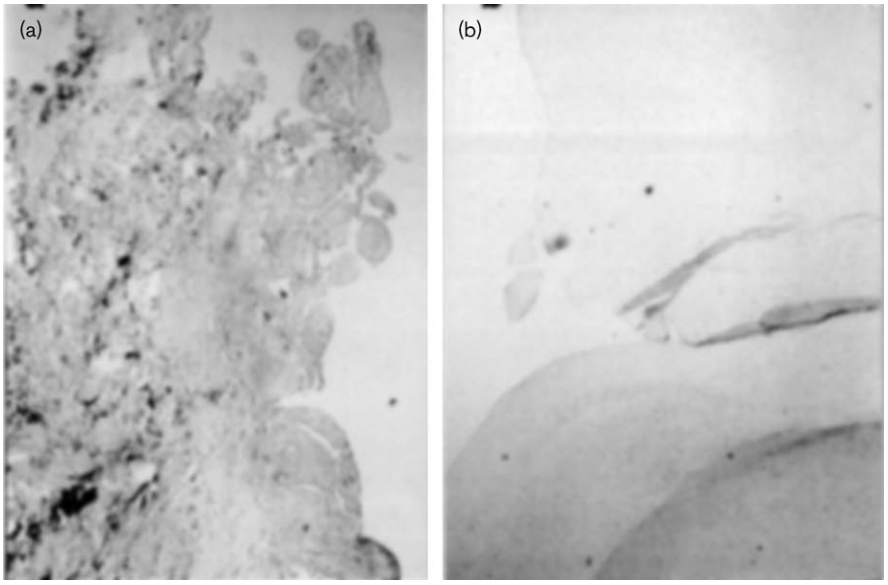
Osteochondroma, otherwise known as exostosis, is by far the most common of the benign bone tumors [1]. Whether single or multiple, an osteocartilaginous exostosis represents, as its name implies, a cartilage-capped bony growth protruding from the surface of the affected bone. Osteochondromas may become symptomatic if there is mechanical irritation of soft tissues, nerve or cord compression, vascular injury, fracture, or malignant transformation. The latter occurs in less than 1% of solitary osteochondromas. A reactive bursa may rarely develop over an osteochondroma, as a result of mechanical impingement upon adjacent structures, such as muscles and tendons [2].

The term synovial chondromatosis or SOC denotes the relatively rare condition in which foci of cartilage develop in the synovial membrane of a joint, apparently through metaplasia of the sublining connective tissue of the membrane [3]. This type of lesion usually affects the knee joint. SOC may develop within: (i) an otherwise normal synovial joint (primary lesion), (ii) a joint that is affected by some other pathological entity (secondary lesion), (iii) tendon sheaths, and (iv) within extra-articular bursae. The extra-articular type of osteochondromatosis that is developed within bursae or tendon sheaths is exceptionally uncommon [4]. It is most frequently located in hands or feet; therefore its unusual presentation in other parts of the body may pose a difficult diagnostic problem. The very existence of this type of lesion is evidence that the lining cells of joints, bursae, or tendon sheaths may possess the ability to respond to some type of induction in this manner, hence, supporting the theory that this type of lesion is a reactive process [3]. Radiographically, extra-articular SOC appears as a soft tissue mass with multiple small calcified or ossified densities that show diffuse mottled mineral deposition within the lesion. Occasionally, there may be only several calcified bodies that conglomerate, thus giving a multilobulated appearance. The plain radiographs are usually adequate for preoperative evaluation.

The development of an osteochondromatosis in a bursa overlying an osteochondroma is even rarer, as it has been only described in eight patients, all of whom were adults [2,4]. Peh *et al.* [2] considered that the actual cause of the development of a secondary osteochondromatosis over an osteochondroma is the shedding of osteochondroma fragments into a bursal or synovial cavity that induces synovial chondrometaplasia and hence SOC formation. Among the possible differential diagnoses that must be considered when dealing with SOC overlying osteochondromas, the most possible ones are: post-traumatic haematoma, pseudomalignant myositis ossificans, periosteal chondroma, and soft-tissue or juxtacortical osteogenic sarcoma [2,4].

In the case herein presented, the injury of the patient should be probably considered as coincidental; nevertheless, it served well in obliging the patient to seek medical attention. As far as the pain was concerned, it should probably be attributed to the developed impingement between the bursa, the osteochondroma, and the overlying soft tissues.

Fig. 3

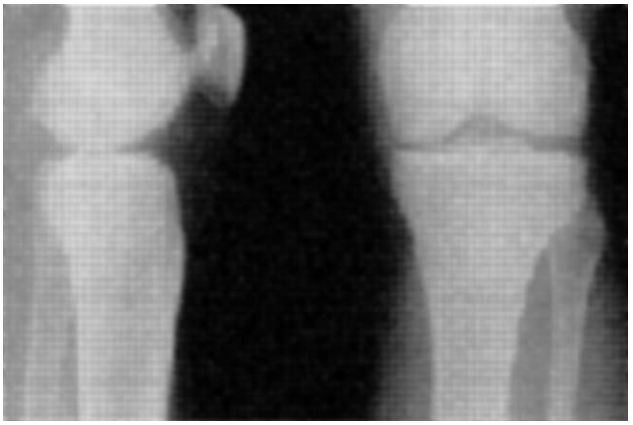


Macroscopic (a) and microscopic (b) cut sections of the resected bursa. Note the granulomatous texture of the inner wall of the bursa (stain: hematoxylin–eosin).

Fig. 4

Microscopic cut section of a free osteocartilaginous body that was found inside the resected bursa. Note the cartilaginous texture combined with multiple areas of calcification (stain: hematoxylin–eosin).

Fig. 5



The radiographic appearance of the same area 8 years postoperatively. The growth plates have been ossified and the proximal third of the metaphysis appears to be normal with no sign of recurrence.

This is the first, according to the best of our knowledge, report of a bursal osteochondromatosis overlying an osteochondroma in the developing skeleton. Considering that, our patient's history (pain for the past 18 months), clinical (palpable masses), and radiological findings (soft tissue mass with multiple small calcified densities overlying an osteochondroma) could have been very easily misinterpreted as belonging to a malignantly transformed osteochondroma. This could have led to more radical decisions as far as the treatment options were concerned. Therefore, the orthopaedic surgeon must always keep in mind the fact that a bursal osteochondromatosis may be overlying an osteochondroma even in younger patients, especially when dealing with such diagnostic dilemmas and particularly when planning treatment modalities and strategies. This type of lesion is more likely to occur in osteochondromas that are not fully and adequately covered by soft tissue, therefore allowing external factors (such as injuries or friction caused by

adjacent tendons or fascia) to exert their potential 'stimulus effect'.

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