

INTRAEPHYPHYSEAL OSTEOID OSTEOMA OF PROXIMAL TIBIAL EPIPHYSIS: A CASE REPORT

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Abstract- Intraepiphyseal location of an osteoid osteoma is extremely rare with few cases reported in the literature. They are often present with various atypical and nonspecific clinical features. Synovitis of adjacent joint is common and along with chronic inflammation could be misinterpreted as rheumatoid arthritis. Initial plain radiographs are often not diagnostic and further radiologic evaluation with CT scan is an essential part of diagnostic work-up. Repeated plain X-ray is very useful in making a correct diagnosis. Although the characteristic double density sign is usually absent in isotope scan in intra-articular osteoid osteoma, it plays an important role in localization of the nidus. We report a case with an intraepiphyseal osteoid osteoma without any signs of synovitis or growth disturbances. During the en bloc resection of nidus, we noticed no communication between the nidus and joint or physis in our cases. It seems that communication of nidus with joint space or growth plate is essential to allow mediators of inflammation pass to joint or growth plate and produce synovitis or growth disturbances, respectively.

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INTRODUCTION

Osteoid osteoma is a well-known benign osteoblastic tumor that usually occurs in the second and third decades of life (1-3). Nocturnal pain, response to salicylates and radiolucent nidus with surrounding sclerosis are its characteristic features that generally lead to easy diagnosis (2,3). However, sometimes the clinical and radiographic features are atypical, especially when it is located in atypical locations such as intra-epiphyseal, intra-articular or juxta-articular localization. This could lead to difficulties in diagnosis, attempting unnecessary procedures and delayed treatment (1-6).

We report a case of osteoid osteoma in the proximal tibial epiphysis juxta-articular to the knee

joint in a 9-year-old girl in whom diagnosis was made after 1 year delay. In spite of intra-articular localization, no signs of synovitis were observed. Osteoid osteoma should be considered in the differential diagnosis of patients with atypical knee pain. The sophisticated imaging techniques, such as repeated X-ray, isotope bone scan, and computed tomography (CT) scan facilitate the diagnosis.

CASE REPORT

A 9-year old girl was referred to our ward with a 12-month history of left knee pain. History of trauma was not forthcoming. Initially, her pain was mainly nocturnal and awakened her at night, but during the past few months she began to complain of pain both during day and night. Aspirin had been prescribed and it helped to relieve pain for almost four hours.

On examination there was no swelling and effusion around the knee. Active and passive range of motion in her left knee was not limited and there was

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Intraepiphyseal osteoid osteoma

no knee instability. The pain was aggravated by digital pressure over lateral joint line anteriorly. There was an obvious atrophy of left calf and thigh. Routine and inflammatory blood tests carried out were within reference range. Review of standard X-rays that had been taken at the beginning of her disease revealed no abnormality. Subsequently the patient underwent another conventional radiography which revealed soft tissue atrophy and diffuse osteoporosis along with a lytic lesion with sclerotic rim in anterolateral aspect of proximal tibial epiphysis (Fig. 1).

An elevated uptake was seen on bone scintigraphy. CT-Scan showed a lytic lesion at proximal tibial epiphysis that was interpreted as subacute osteomyelitis by radiologist (Fig. 2).

The lesion was operated on by an anterolateral approach over the lesion. The growth plate was determined. Localization of lesion was done under image intensifier control. Osteoid osteoma was completely excised with great care not to damage physis or articular cartilage (Fig. 3).

Successive histological examination confirmed the diagnosis (Fig. 4). The preoperative pain disappeared the same night after the operation.

At the latest follow-up 4 years after surgery, she complained of no pain or growth disturbance in her left lower extremity (Fig. 5). Muscle atrophy of her calf and thigh was resolved completely.



Fig. 1. Preoperative X-ray revealing soft tissue atrophy and diffuse osteoporosis along with a lytic lesion with sclerotic rim



Fig. 2. Preoperative CT scan showing a lytic lesion at proximal tibial epiphysis.



Fig. 3. Postoperative X-ray showing complete excision of osteoid osteoma.

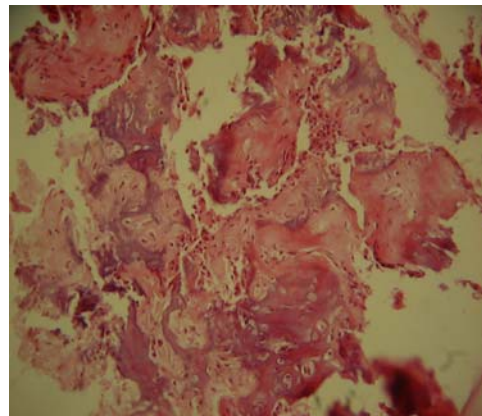


Fig. 4. Histological examination confirmed the diagnosis of osteoid osteoma.



Fig. 5. Latest follow-up X-ray, 4 years after surgery.

DISCUSSION

Osteoid osteoma is a common bone tumor, representing approximately 10% of all benign skeletal lesions (1-5). It is most frequently seen in the 10-25 year age group, although cases between the ages of 5 and 53 years of age have been documented (1). Males are affected three times more than females. Our case was a 9-year-old girl at the time of diagnosis.

The typical location is the subperiosteal region of the diaphysis of long bones, most frequently the femur followed by the tibia. Approximately 13% of osteoid osteomas arise within a joint, the commonest site being the hip. The ankle, elbow, wrist and knee are affected less commonly (6-9). The intraepiphyseal location of an osteoid osteoma in a skeletally immature individual is an extremely rare occurrence, with only few cases being reported (6, 10-14).

Clinical presentation of intra-articular lesions typically consists of variable articular pain indistinguishable from other arthropathies. Unlike the more classically located osteoid osteoma, night pain is not typical. Also, the pain from intra-articular lesion is less responsive to non-steroidal anti-inflammatory drugs than the extra-articular variety (1-3). Joint tenderness, soft tissue swelling, synovitis

and effusion are typical, like any mono-articular inflammatory arthritis (7,15).

It has been shown that the level of prostaglandin E2 (PGE2) and prostacyclin (PGI2) can reach levels ten times their production in normal tissue. In intra-articular osteoid osteoma, these mediators of inflammation may play an important role in the pathogenesis of the concomitant synovitis (16). It seems that an intra-articular perforation or communication of nidus should occur in intra-articular osteoid osteoma with concomitant synovitis (9, 17). In our case, we did not find any communication between nidus and joint and this could explain why she had no sign of concomitant synovitis in her knee.

The mean time to diagnosis is 2-3.5 year and it is made by a combination of clinical, radiographic, CT and scintigraphic features (4, 5, 19, 20). Clinical features of osteoid osteoma may present 6 to 8 months before onset of any characteristic radiographic findings of the lesion and the radiolucent nidus is often overlooked in initial radiographs. Initial radiographs are not diagnostic in most cases (2). Also the nidus is not reliably detectable if below 3 mm in size. In our case, initial radiographs could not demonstrate the lesion, but later radiographs did. This shows that repeated radiographs in uncertain cases may be helpful. Regional periarticular osteopenia is a prominent feature in the early stages. With lesions located in the hands and feet, appearance can be indistinguishable from reflex sympathetic dystrophy.

Osteoid osteomas have typical scintigraphic feature, when studied with technetium 99m, designated as double density sign. However, when the lesion is intra articular, this sign is typically absent and the activity is often generalized within the joint due to associated synovitis, osteoporosis and hyperemia. Focal accumulation of the radioisotope is commonly seen, corresponding to location of the nidus (2).

CT scan is the investigation of choice for identifying the nidus (1). In intra articular osteoid osteoma; however detection of the lesion is highly dependent on technical factors. Axial or coronal imaging with no more than 2-3 mm slice thickness, reconstructed with a bony algorithm and imaging on

Intraepiphyseal osteoid osteoma

bone windows is ideal. In this case, CT scan determined the exact location of the lesion and was helpful in planning surgery.

In conclusion, an intraepiphyseal osteoid osteoma in a skeletally immature individual may be present without joint related symptoms. Characteristic clinical features of osteoid osteoma are often absent. So, only high index of suspicion along with logical use of radiologic and nuclear medicine findings can lead the surgeon to the right diagnosis.

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