Osteoid Osteoma of the Talus Mimicking Pigmented Villonodular Synovitis in a 15-Year-Old Male: A Case Report

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Osteoid osteoma is a benign tumor of the bone which tends to occur in diaphysis or metaphysis of the long bones. The lesion is generally intraosseous with vague clinical symptoms, hence given the name “great mimicker”. When located subperiosteally and juxtaarticulary, atypical clinical presentation and radiological may lead to a delayed or missed diagnosis. Performing surgery with a misdiagnosis carries the risk of incomplete resection of the lesion and recurrence. We report the case of a 15-year-old male with a subperiosteal osteoid osteoma of the talus, who was misdiagnosed with pigmented villonodular synovitis and operated through
anterior ankle arthrotomy. A nodular lesion 1 cm in diameter with hard rubber consistency was removed from the dorsal aspect of the talar neck. The pathological specimens were consistent with subperiosteal osteoid osteoma. The patient’s symptoms resolved rapidly in the early postoperative period. The patient remained asymptomatic at the 20th-month follow-up and the control MRI revealed no signs of recurrence. Atypical radiological and clinical presentation of juxtaarticular subperiosteal osteoid osteomas cause misdiagnosis, delay in diagnosis, incomplete resection and recurrence. It is important to keep in mind "juxtaarticular subperiosteal osteoid osteoma" in the differential diagnosis of cases with suspected Pigmented Villonodular Synovitis.

Introduction

Osteoid osteoma (OO) is a relatively common benign tumor of the bone / bone-forming tumors with characteristic radiological features. Described by Jaffe in 1935, it accounts for 2-3% of all primary bone tumors and 10-14% of benign lesions. It is more common in adolescents and young adults with male predominance. The dysplastic osteoid core of the lesion is called nidus, with osteoid and osseous cellular components embedded in a fibrovascular stroma. Secretion of prostaglandin E2 from osteoblasts in this highly vascularized tissue is responsible for the characteristic pain alleviated by cyclooxygenase inhibitors. The appearance of the nidus as a...
radiolucent region inside a reactive sclerotic rim is nearly pathognomonic for osteoid osteoma.

Localization of the lesion with respect to the cortex and the anatomical region has profound effects on clinical and radiological findings. In 1966, Edeiken described 3 types of osteoid osteoma with distinctive roentgenographic features: Cortical, cancellous, and subperiosteal. In most of the cases, the lesions are intraosseous and tend to occur in the diaphysis or metaphysis of the long bones of the lower extremity 3. Subperiosteal osteoid osteoma is the least common type and the literature on this variant consists of either case reports or case series with a limited number of patients 4. It usually lacks the characteristic roentgenographic features, which are otherwise nearly pathognomonic for cortical lesions. When the tumor is juxtaarticular, joint-related symptoms may predominate and mimic arthritis. Atypical radiological and clinical presentation of juxtaarticular subperiosteal osteoid osteomas cause misdiagnosis, delay in diagnosis, incomplete resection, and recurrence 5–7. We report the case of a 15 year-old-male with subperiosteal osteoid osteoma of the talus, who was misdiagnosed with pigmented villonodular synovitis and operated through anterior ankle arthrotomy.

Case

A 15-year-old male presented to our clinic with relapsing ankle pain. The symptoms had begun about one year ago and had been gradually worsening. Lately, the ankle had become diffusely swollen. On physical examination, there was marked effusion in the ankle and the patient
elicited global tenderness to palpation. Ankle range of motion (ROM) was limited. The patient’s initial MRI 2 months ago was reported as diffuse edema in the talus and minimal synovial hypertrophy, with no evident joint effusion (Figure 1). The patient was presented at our institution’s Sarcoma Council, and a new MRI with contrast (Gadolinium enhancement) was planned. Findings of diffuse synovial hypertrophy with areas of hemosiderin deposits and marked joint effusion in his new magnetic resonance imaging (MRI) were deemed suggestive of pigmented villonodular synovitis, and the patient was scheduled for open synovectomy (Figure 2)

The ankle joint was approached through an anterior midline arthrotomy. Following resection of the marked hypertrophic synovium inside the capsule, anterior of the tibia plafond, talar dome, and talar neck were exposed. A nodular brown-red lesion 1 cm in diameter with hard rubber consistency was noted on the dorsal aspect of the talar neck (Figure 3). The lesion was excised with a sharp osteotome, and the adjacent cortex of the talar neck was curetted. A subtotal synovectomy was performed using the same incision, and the layers were closed in a regular fashion.

The pathological specimens were consistent with subperiosteal osteoid osteoma and chronic synovitis with myxoid degeneration (Figure 4). The patient’s symptoms resolved rapidly in the early postoperative period. His control MRI revealed no sign of recurrence, complete resolution
of talar edema and occurrence of disuse osteopenia in other tarsal bones (Figure 5). The patient remained asymptomatic and there was no abnormal osseous image on computed tomography at the 20th 66-month follow-up (Figure 6).

Discussion

Majority of the osteoid osteomas occur in the diaphysis or metaphysis of the long bones of the lower extremity. When present in the foot region, the lesion is generally in the talar neck and most of the time subperiosteal. Surprisingly, the first case to be ever defined as osteoid osteoma by Jaffe in 1935 was an osteoid osteoma of the talus, which was operated on the assumption that it was a sequestrum associated with osteomyelitis. Although the subperiosteal variant is the rarest, most of the lesions reported in the talar region are subperiosteal. In Capanna et al.'s report on 33 cases with talar OO, 32 lesions were in the neck region and 25 of them were subperiosteal. Because of their mostly subperiosteal histopathology and the proximity of the lesion to the ankle joint, lesions in the talar region tend to have an atypical radiographic and clinical presentation, which may complicate the diagnosis.

As is the case with other subperiosteal lesions, radiographic findings are usually non-specific and subtle when the subperiosteal tumor is in the talar neck. Reactive sclerosis of the cortex, the hallmark of cortical osteoid osteoma, is generally absent and the majority of the cases barely demonstrate any cortical or periosteal changes on conventional radiographs.
absence of cambium from the periosteal layer in the juxta-articular region may account for the absent or minimal reactive cortical thickening in these lesions. Plain roentgenograms of our patient also did not demonstrate cortical sclerosis. There was a nodular lesion with minimal calcification barely visible on plain radiographs, and the underlying cortex was eroded. A bone spur was present close to the talonavicular joint, which could have been mistaken for degenerative arthritis if the patient was not an adolescent. There are reports of OO appearing as a spur in the talar neck, mimicking long standing arthritis.

When appearing close to joints, it is not uncommon for these lesions to cause synovitis, joint effusion and bone erosion of the adjacent cortex. The overlying skin may be warm and tender to palpation and the soft tissue swelling may be the predominant finding. As symptoms of capsular distention and synovitis predominate, the lesion may be mixed with inflammatory arthropathy or other joint related pathologies. This may lead to preference for MRI over CT for differential diagnostics, as was in our case. However, the accuracy of MRI in diagnosing osteoid osteoma is very low compared to CT scans, and may lead to misdiagnosis in up to 97% of cases if interpreted without clinical correlation. Nevertheless, the lesion may still be missed if CT is not thin sliced. With MRI, changes in the joint and surrounding soft tissue may be much more striking than the lesion itself, and the radiological features of the nidus may be obscured by extensive bone edema. In our case, effusion and excessive synovitis were the main findings,
which led to the misdiagnosis of PVNS despite consulting with the Sarcoma Council.

Treatment of symptomatic lesions refractory to conservative treatment is surgical resection or ablation of the nidus. With well-defined lesions, histopathological analysis is not mandatory, and treatment modalities such as RF ablation, which preclude the possibility of taking an intraoperative sample, may be utilized. In cases with atypical locations and presentations, obtaining a pathology specimen may be preferred. There are mixed opinions about the ideal surgical technique for resection. He Xu et al. propose that the lesion can be obscured by diffuse hypertrophic tissue during arthroscopy⁵, whereas others promote arthroscopy for the ability to perform a thorough synovectomy thru small portals in an outpatient setting. Results of arthroscopic resection are encouraging, with very few incidences of recurrence reported. Preferably, motorized instruments should be utilized only after the margins of the nidus are delineated and the lesion is removed in one piece with a grasper⁴,¹⁴–¹⁶. Damage to the neurovascular and tendinous structures in the anterior aspect of the ankle and joint stiffness are noted as concerns for anterior arthrotomy. However, there is no incidence of vascular injury reported during open resection of OO in the talus. We were able to perform an en-bloc resection of the lesion thru open surgery, and the patient’s ankle ROM was identical to his non-operated foot 20 months after surgery (Figure 7). The patient is able to attend recreational sports activities without any pain or limitation.
Conclusion

Atypical presentation of subperiosteal lesions on the talar neck may lead to a delayed or missed diagnosis. This report suggests the importance of keeping in mind "juxtaarticular subperiosteal osteoid osteoma" in the differential diagnosis of cases with suspected Pigmented Villonodular Synovitis. Performing surgery with a misdiagnosis carries the risk of incomplete resection of the lesion and recurrence. When performed properly, it is possible to obtain en-block resection of the subperiosteal lesion without any complications thru anterior arthrotomy.
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Figure 1. Preoperative radiograph and MRI without contrast. Diffuse edema in the talus and minimal synovial hypertrophy, with no evident joint effusion. Plain roentgenograms do not demonstrate cortical sclerosis. A nodular lesion with minimal calcification is barely visible on plain radiographs, and the underlying cortex is eroded.
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**Figure 2. Preoperative MRI with contrast.** Findings of diffuse synovial hypertrophy with areas of hemosiderin deposits and marked joint effusion in his new MRI were deemed suggestive of pigmented villonodular synovitis
Figure 3. Intraoperative findings. Brown-red lesion 1 cm in diameter with stiff rubber consistency, on the dorsal aspect of the talar neck.
Figure 4. Microscopic photographs of the excised tumor tissue. a. Nidus structure adjacent to the articular cartilage (HEX25), b. Irregularly shaped immature bone trabeculae in the fibrous stroma and reactive sclerosis in the periphery (HEX25), c. Immature bone trabeculae in varying size, anastomosing with each other (HEX100), d. Lymphoplasmocytic inflammation, with fingerlike projections in the synovium and local aggregates in the subsynovial area (HEX100).
Figure 5. Control MRI at 12 months. No sign of recurrence, complete resolution of talar edema and occurrence of disuse osteopenia in other tarsal bones.
Figure 6. Computerized Tomography at 20 months after surgery. No signs of bone defect or abnormal ossification
Figure 7. Last follow-up at 20 months. Ankle ROM is full as same as the non-operated foot.