A Rare Cause of Elbow Pain, Osteoid Osteoma of the Coronoid Process of the Ulna: Report of a Case

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Abstract

Osteoid osteoma is a relatively common benign neoplasm of the bone. An intra-articular osteoid osteoma is a rare form of the tumor with subtle radiologic and clinical findings mimicking an inflammatory arthritis which usually leads to a delayed diagnosis. We reported a 22-year-old male patient with subacute pain and decreased range of motion in his right elbow. Paraclinical evaluations showed a cystic bony lesion of the coronoid process of the proximal ulna that went through the surgical excision. Histopathologic examination revealed an intra-articular osteoid osteoma. The patient had a favorable outcome.

Keywords: Coronoid Process; Elbow; Osteoid Osteoma

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Introduction

Being a relatively common bone lesion, Osteoid osteoma accounts for 10-12% of all benign bone neoplasms (1, 2). It primarily affects young people in their first two decades of life, with the majority of cases occurring between the ages of 5 and 30 (3). Men are 2-4 times more likely to be affected than women (4).

The tumor can develop in almost any bones in the human body. However, in more than 50% of cases, the long bones are the source of the tumor (3), with the tibia and femur being the most frequently affected sites (about 50-60% of cases) (4-5). The diaphysis or metaphysis of long bones are the most frequently diagnosed sites for osteoid osteomas (5). With a frequency of around 10-13%, epiphyseal and intra-articular locations are deemed rare, with the hip being the most common site (4-7). Other less involved areas include the knee, ankle, elbow, and wrist. A lesion is classified as intra-articular when it develops inside a synovial joint capsule at the extreme ends of long bones or in close proximity to it (3).

This condition's etiology remains mostly unclear and the incidence of a familial occurrence is exceptionally unlikely. Cytogenetic modifications such as the deletion of chromosome 22q have been identified in cases of osteoid osteoma, supplementary researches are needed to definitively relate this entity to a clonal mutation though (8, 9).

Identified by pain and a decreased range of motion, osteoid osteoma of the elbow remains a diagnostic challenge for the orthopedic surgeon due to its nonspecific clinical and subtle radiographic findings (10). Timely diagnosis and treatment are important keys to prevent subsequent osteoarthritis and possible irreversible cartilage loss. We herein reported a case of osteoid osteoma of the elbow.

Case Report

The patient was a 22-year-old gentleman referred to the outpatient clinic of Imam Hossein Medical Center, Tehran, Iran with a 2-month history of dull pain in his right elbow. He mentioned a trivial trauma to his elbow from a ground-level

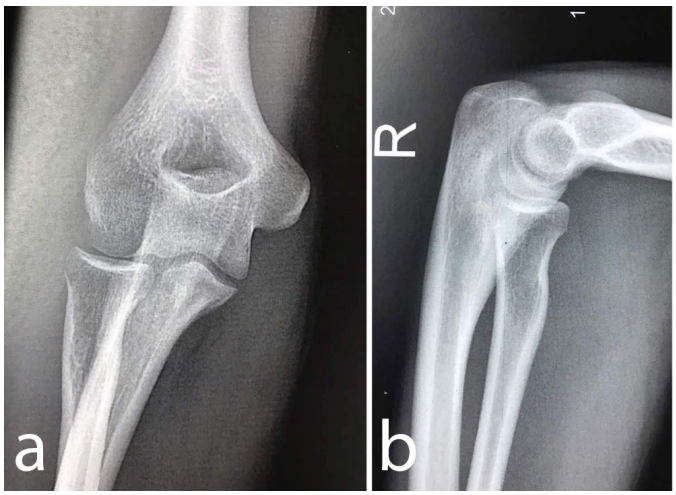

Figure 1. Anteroposterior (a) and lateral (b) X-rays of the affected elbow show no pathological findings

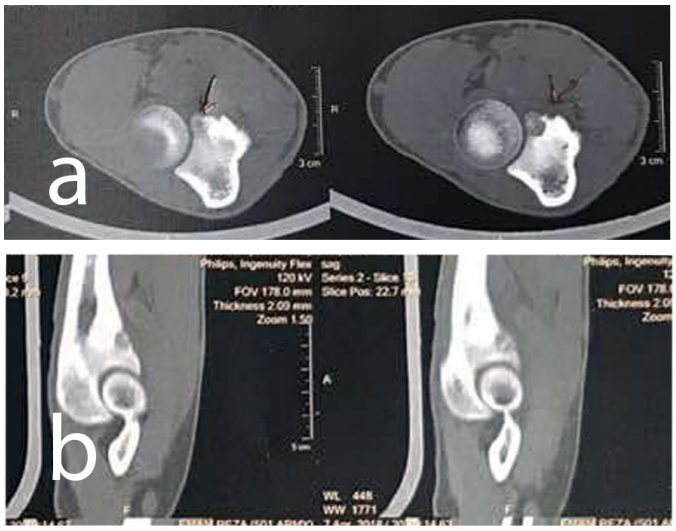

Figure 2. Axial (a) and sagittal (b) CT scans show the cystic bony lesion of the coronoid process.

fall two months earlier and the symptoms started afterward. Conservative treatments including oral non-steroidal anti-inflammatory drugs and elbow support brace temporarily reduced the symptoms but did not cure the condition. His previous medical and surgical history was unremarkable. Physical examination with the orthopedic goniometer placed on the lateral epicondyle of the distal humerus revealed a decreased range of motion in the final 30 degrees of elbow extension with no obvious bony tenderness around the joint. The neurovascular examination including distal arterial pulses and sensory and motor functions was unremarkable. Blood works inclusive of the inflammatory markers were all within normal limits. A plain X-ray of the elbow was obtained and showed no obvious pathological findings (Figure 1).

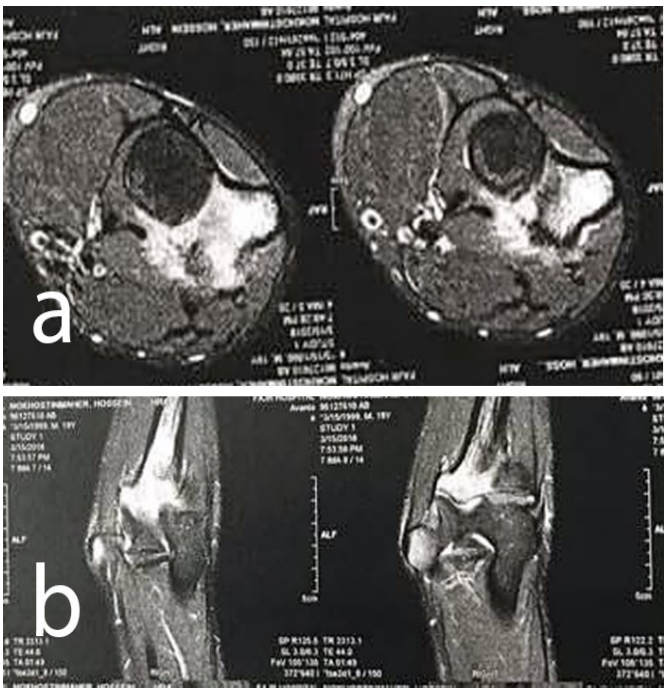

Figure 3. Axial (a) and coronal (b) MRI of the elbow show diffuse bone marrow edema of the proximal ulna

CT scan of the elbow revealed a small well-defined cystic lesion in the coronoid process of the proximal ulna with no cortical destruction (Figure 2). Magnetic resonance imaging (MRI) of the joint showed bone marrow edema in the proximal ulna along with the cystic lesion (Figure 3).

Tc99m-MDP bone scintigraphy revealed a localized hot point of uptake in the right elbow (Figure 4). A surgical excisional biopsy was planned and the lesion was approached via a midline anterior skin incision (Figure 5).

Histopathological examination shows tissue with a central nidus made of bony trabeculae encompassed by a ring of sclerotic bone separated by vascular fibrous connective tissue that correspond with osteoid osteoma

Upon the patient's request, a genetic evaluation was done which turned out to be positive for V617F mutation in the JAK2 tyrosine kinase gene. A rehabilitation program with physical therapy, ultrasound, laser therapy, and electromagnetic field therapy to regain range of motion, decrease pain/inflammation, and retard muscular atrophy was initiated. A 2-year follow-up of the patient was uneventful with a near-normal range of motion and no residual pain.

Discussion

With explicit clinical, radiological, and pathological features, osteoid osteoma is a relatively common benign bone tumor

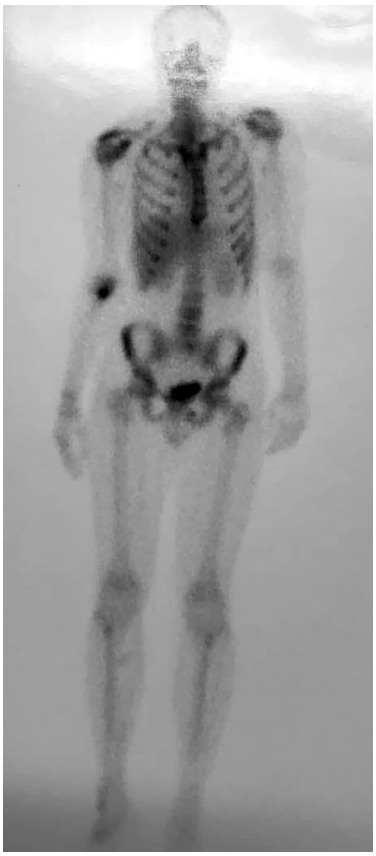

Figure 4. Tc99m-MDP bone scintigraphy shows the increased uptake in the right elbow

that most often affects men under the age of 30 (3, 4, 10, 11). Clinical symptoms of intra-articular activity are nonspecific, and radiographic results may be attenuated, thereby making diagnosis difficult. Osteoid osteoma of the elbow is characterized by discomfort, recurrent synovitis, joint effusion, decreased range of motion, and flexion contracture, which mimic inflammatory arthropathy (12). Moreover, reactive sclerosis can be minimal or missing on radiography, causing a substantial pause in diagnosis or even misdiagnosis (13, 14).

While Bergstrand (15) was the first to describe it, the term of osteoid osteoma was first used in the medical literature in 1935 by Jaffe who defined the disorder as a distinct pathological entity based on a series of five cases (16). In a study by Kalil *et al.* (9), the osteoid osteoma was identified in two brothers both in the proximal femoral cortices who were the same age when their symptoms began. These data may suggest a genetic predisposition. Our patient in the present case report had JAK2 V617F mutation, but this might be an accidental finding and the clinical relevance and significance are currently unclear.

Osteoid osteoma of the coronoid process is exceedingly rare. To our knowledge, less than 10 cases have ever been

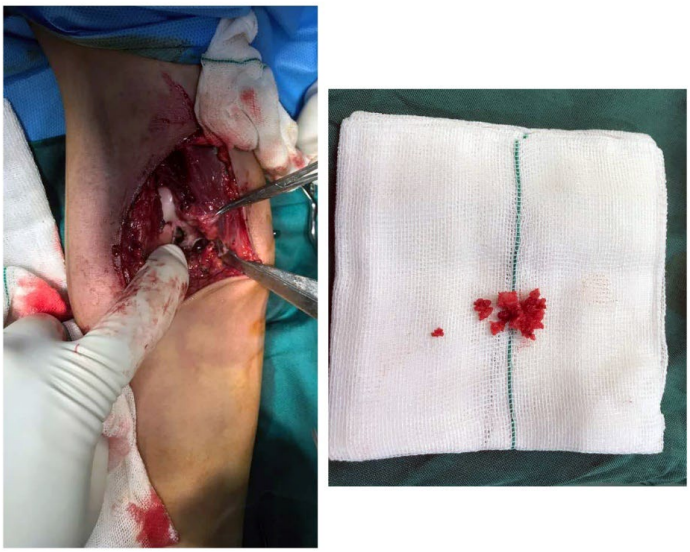

Figure 5. Intraoperative picture of the coronoid process lesion

reported in the English medical literature. The first report was by Masquelet *et al.* (17) that that unlike our case, a posterior approach was used for surgical excision. Akman *et al.* (18) surgically removed a coronoid process osteoid osteoma in a 23-year-old patient with flexion contracture of the elbow through the posterolateral (Kocher) incision. Becker *et al.* (19) reported a pronation loss and Decramer *et al.* (20) reported a supination loss after the surgical removal of the coronoid process osteoid osteoma lesion. We did not experience such a complication with our case.

According to immunohistochemical test results, the osteoid osteomas have nerve fibers in the reactive region, and their innervation pattern is different compared to other bone neoplasms (17). The clinical manifestation of acute pain in the troubled region, which exacerbates overnight and improves 30 minutes after salicylates, is present in 75 percent of instances and indicates the diagnosis. High E2 prostaglandins produced by the tumor appear to mediate pain and are hindered by the usage of anti-inflammatory medication. The inordinate discomfort of one patient could be linked to the presence of distinguished focal nerve fibers in the area of neighboring soft tissue reactions. The use of non-steroidal anti-inflammatory medications (NSAIDs) for pain control might be less successful in the intra-articular forms of the tumor than in the extra-articular presentation (22), a finding that is consistent with our case.

The symptoms of pain and discomfort usually subside over a 2- to 5-year period while the tumor goes through histologic maturation. Osteoid osteomas tend to regress spontaneously, but the underlying histopathologic procedure is currently not well known.

The imaging technique used for the initial assessment is plain radiography. The classic radiographic finding in cases of osteoid osteoma is the so-called nidus, which is around or ovoid lytic lesion usually less than 2.0 cm in diameter surrounded by a focal region of sclerotic bone and/or reactive thickening of the cortex (18). These findings may differ depending on the affected anatomical region, location of the lesion, duration of symptoms, and age of the patient at the onset of symptoms. Besides, in some cases, the nidus could be consistently radiolucent or have varying degrees of calcification. As the lesion lasts for a longer period of time, signs of bone maturation become more noticeable in imaging studies.

When the conventional clinical and radiographic results are evident, the diagnosis is not complicated. However, in cases of atypical tumor locations such as intra-articular lesions, the usual clinical manifestation might not be present which compromise the diagnosis.

Osteoid osteomas could occur in the cortex, medullary canal, or subperiosteal region. Medullary or subperiosteal lesions might not show the typical sclerotic rim around the nidus. Intra-articular tumors are often medullary or subperiosteal, hence rendering radiographic diagnosis becomes even more challenging.

In such conditions when radiography is occult, the use of computed tomography with three-dimensional reconstructions are the preference methods to confirm the presence of nidus and surrounding sclerosis. CT scan is also very helpful in situations where there is a suspicion of a residual lesion after the treatment.

The use of MRI in the diagnosis of osteoid osteoma is debatable. The variable presence of the lesion as well as widespread reactive changes in the bone marrow and surrounding soft tissues may suggest a more active or distinct type of lesion. Adding the contrast medium to the magnetic resonance imaging combined with high-resolution images is a technique to improve diagnostic sensitivity and specificity based on hypervascularization of the nidus, particularly in cases where the lesion is placed in an unusual location and patients show no typical clinical symptoms (19-21).

Technetium-99m bone scintigraphy has a high sensitivity in the diagnosis of osteoid osteomas. The tumor demonstrates the expected pattern of double-density. This symptom is described by extreme hyper uptake in the nidus area and milder deposition of the tracer at the lesion's peripheral area, which includes reactive sclerotic bone and perilesional edema. However, in instances with intra-articular lesions, such a sign is often missing, since a generalized surge in movement occurs inside the joint owing to the occurrence of diffuse synovial uptake and hyperemia.

Pain, soft-tissue edema, and joint effusion are also symptoms of an intra-articular lesion. Synovitis may be severe, resulting in particular rigidity in the patient. Osteopenia, joint space narrowing, and hypertrophic changes can be noticed (5).

The disorder can mimic clinically and radiographically inflammatory arthritis, leading to a delay in diagnosis and treatment for months or in some cases even years. Intra-articular osteomas can result in osteoarthritis as a result of chronic synovitis and secondary changes such as synovial proliferation and subsequent cartilage loss. Another late complication is muscular atrophy due to limb disuse, especially if the tumor is long-term and painful.

The differential diagnoses of osteoid osteoma of the elbow include but are not limited to different types of inflammatory synovitis, periarticular tendinitis, osteochondritis dissecans (OCD), infectious processes such as osteomyelitis, occult fractures, chondroblastoma, and osteoblastoma. Osteoblastoma shares the same clinical and histopathological features with osteoid osteoma; nevertheless, osteoblastoma is greater than 2.0 cm in size, is more violent, and rarely regresses spontaneously.

The current study's findings, which are consistent with previous studies, indicates that diagnosis of intra-articular osteoid osteoma of the elbow may be difficult based on the history and radiographic findings of the patient.

Conclusion

Although prevalent and well-known, osteoid osteomas could bear diagnostic challenges when present in unusual anatomic locations such as near or within a synovial joint. The typical clinical and radiographic findings might not be present in cases of intra-articular lesions. Osteoid osteoma should be in mind for young patients suffering from subacute or chronic periarticular pain and unexplained limited range of motion, particularly when the symptoms are refractory to conservative treatments. Since the final consequence of intra-articular lesions could be permanent cartilage loss, timely diagnosis is an important key to a favorable outcome.

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