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Case Report

Osteoid Osteoma in the Proximal Humerus in a 52-Year-Old - A Case Report

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Abstract

Introduction: Osteoid osteoma is a rare bone tumor with high prostaglandin expression that often presents in the second decade of life and is usually responsive to non-steroidal anti-inflammatory agents.

Aim: We report an osteoid osteoma of the proximal humerus in a 52-year-old, an unusual location and in a patient who is older than most who get these tumors.

Case Report: A 52-year-old man comes to the outpatient orthopedics office with R shoulder pain. After receiving a corticosteroid injection without relief of symptoms, further imaging studies revealed an osteoid osteoma in the proximal humerus which was unresponsive to non-steroidal anti-inflammatory therapy. He underwent radiofrequency ablation of the lesion and was symptom free after the procedure.

Conclusion: Osteoid osteoma can appear in a wide variety of locations and in different age groups. Treatment with radiofrequency ablation is safe and effective.

Keywords: Osteoid Osteoma; NSAIDs; Radiofrequency Ablation; Computed Tomography; Humerus

Abbreviations

OO: Osteoid Osteoma; NSAIDs: Non-Steroidal Anti-Inflammatory Drugs

Introduction

Osteoid osteoma (OO) is the third most common benign primary bone tumor, accounting for 10-14% of benign bone tumors and 2-3% of all primary bone tumors [1]. The tumor was initially described and characterized by Jaffe in 1935 with typical radiographic features of a radiolucent nidus surrounded by reactive osteosclerosis [1]. Peak incidence of OO is in the second decade with average age of diagnosis between 5 and 30. 13% of OO is diagnosed in patients over the age of 30 and males are more likely to get OO than females (2-3:1) [1]. OO is most commonly diagnosed in the appendicular skeleton with the lower extremities (femur and tibia) more commonly affected than the upper extremities (most commonly humerus) [1].

OO can be classified into the following categories: intracortical (75%), intramedullary (20%), subperiosteal (5%), and endosteal (5%). Intracortical lesions are most commonly located in the diaphysis followed by the metaphysis [1].

There is some controversy surrounding whether OO pathophysiology. On biopsy, OO is histologically similar to osteoblastomas with atypical cellular and trabecular structure, making the OO seem like a tumor [1]. However, due to the OO's self-limited nature, small size, and containment of viral particles, some researchers believe the OO is a sequalae of unusual healing of bone without apparent injury [1].

Prostaglandins (PGs) play a major role in the development of these lesions. Levels of PGs in OOs are extremely high and are thought to be the cause of pain [2]. PGs cause vasodilation which increases the blood flow to the lesions, thereby increasing the pressure inside the bone leading to pain [2]. Additionally, PGs amplify the bradykinin pathway (just like in soft tissue injury), leading to increased pain [1].

Clinical features include mild to moderate pain that increases at night. The pain is intermittent initially but increases in frequency and intensity over time [1]. The pain is relieved by non-steroidal anti-inflammatory drugs (NSAIDs) or salicylates (i.e., aspirin) [1]. If not diagnosed, the OO can cause bone widening and bone deformities.

The standard treatment for osteoid osteoma includes NSAID or salicylate therapy and radiofrequency ablation (RFA) for definitive therapy of the lesion. OOs tend to regress spontaneously so there is a role for conservative NSAID therapy trial before sending the patient for RFA. However, there is no way to predict in which patients the lesion will regress and in which patients the lesion needs RFA excision [3]. NSAIDs and salicylate will inhibit the COX-2 enzyme which will dramatically decrease the concentration of PGE_2, thus decreasing the pain associated with the lesion. Of note, Rofecoxib, a selective COX-2 antagonist, demonstrated superior pain relief during rest, exercise, and at night than conventional NSAIDs and aspirin in OOs [4]. There have also been studies analyzing OO responses to bisphosphonates with NSAIDs showed that addition of bisphosphonates (pamidronate 60mg or zoledronic acid 4mg IV monthly infusion until resolution of symptoms) increased response rates of conservative therapy to 74% at 12 months compared to 30-75% with aspirin alone [5,6].

Surgical indications include intolerance of NSAID or salicylate therapy, continued pain after period of conservative management, and no willingness to activity limitations [1]. En bloc resection and burr-down technique were the gold standard of OO surgical removal for many years [3]. These procedures would ensure complete removal of the tumor; however, in larger lesions, prophylactic internal fixation was required after resection [7]. Furthermore, difficulty in tumor visualization during the procedure and identification of tumor margins could be difficult in these lesions [8]. Therefore, newer minimally invasive methods were developed to enhance tumor visualization and decrease surgical risk and prolonged recovery. Percutaneous RFA, where thermal energy is passed through an electrode inducing coagulation necrosis, has a successful OO excision rate of 95% [9]. CT-guided radiofrequency ablation (CT-RFA) is the gold standard in treatment for osteoid osteomas [10]. CT-RFA has a low rate of primary treatment failure and secondary treatment failure, 8.3% and 3.1% respectively [10]. The most common complications of CT-RFA are skin burns (0.7%) and infection (0.5%) [10]. Other minimally invasive methods of OO removal have shown promise as well. Cryoablation uses argon gas or carbon dioxide to freeze cells to -40 degrees Celsius inducing cell death without destruction to intracellular components [9]. This theoretically creates an abscopal effect, where the immune system can interact with the dead tumor cell contents and create an immunologic response to tumor cells outside the ablation zone [11]. Response rates in the short term are 96% at three months and 90.5% at 12 months [12]. The OO response rates for RFA and cryotherapy were statistically similar in efficacy [9]. Microwave ablation uses microwaves to ablate tumors thermally [9]. These waves can propagate further into tissues and are less dependent on tissue impedance or type than RFA [13].

Case Presentation

Our patient is a 52-year-old man with a history of type 2 diabetes, hypercholesterolemia, and factor V Leiden presented to the outpatient orthopedics office with insidious right shoulder pain that began nine months prior and had gotten considerably worse. The pain radiated into the subdeltoid region and the pain was 8/10 in severity at its worst. He had tried ice, rest, and Advil without much relief although he said the pain was mild except at night when he would awaken due to pain. There was no specific injury to the shoulder that instigated the pain even during work as a delivery driver who lifts heavy car parts on a regular basis. Physical exam showed impingement signs but otherwise he had normal inspection, palpation, range of motion, sensation, and reflexes. Right shoulder x-rays (3-views) were obtained and demonstrated calcific tendonitis. He was counselled to perform home exercises for 4-6 weeks and continue with ice/NSAIDs as needed. He also received a subacromial cortisone injection in the right shoulder. He was told to follow up if the pain did not improve in 6 weeks.

Eight weeks later, the patient called the office and said that he was having increasing pain and he had a weak grip and was dropping objects he was carrying. A magnetic-resonance imaging (MRI) study of the right shoulder was ordered (Figure 1).

Figure 1: MRI of right shoulder showing an anterior sclerotic lesion with a hypointense central nidus suggestive of osteoid osteoma. CT was recommended as follow for further characterization of bone abnormality.

Four weeks after the telephone call, the patient came back to the office to review the MRI results. He said he got a few months of pain relief from the subacromial cortisone injection but now the pain was worse than before. He restated that the pain at night was

the worst. He also stated that when he stopped taking his omega-3 fatty acid supplement, his pain improved. He was not able to take NSAIDs since the prior visit because he has Factor V Leiden for which he was put on Xarelto. On physical exam, he had impingement signs, and the empty can test was positive. Otherwise, the rest of the physical exam was benign. The MRI showed supraspinatus and infraspinatus tendinosis with partial thickness articular surface tear of the infraspinatus footplate, calcific tendinitis of the supraspinatus, and most importantly a nonspecific lesion in the anterior humerus measuring 1.5 x 1.4 cm that was not seen on the prior x-ray. There was a hypointense central nidus which would suggest the possibility of an osteoid osteoma. Further evaluation was recommended with a computed tomography (CT) scan (Figure 2).

Figure 2: CT scan of right shoulder which shows a cortical lesion with a sclerotic rim measuring 11 by 13 mm with a lucent central nidus of 6mm which favored a diagnosis of osteoid osteoma.

Surgical History includes appendectomy, IVC filter placement, and low back surgery

Social History includes smoking history with 15 pack years. He denies alcohol use and works as a delivery driver.

Family history is noncontributory.

Outcome and follow up

The CT-scan showed a cortical lesion with a sclerotic rim measuring 11 by 13 mm with a lucent central nidus of 6mm. There was also soft tissue extension anteriorly. The overall pattern favored the diagnosis of osteoid osteoma with soft tissue infection and rotator cuff disease being less likely diagnoses. He also had incidental calcific tendinitis. A follow up bone scan was ordered to characterize the lesion and it showed moderate focal increased activity in the right humeral head with suggestion of double density sign favoring the diagnosis of an osteoid osteoma (Figure 3).

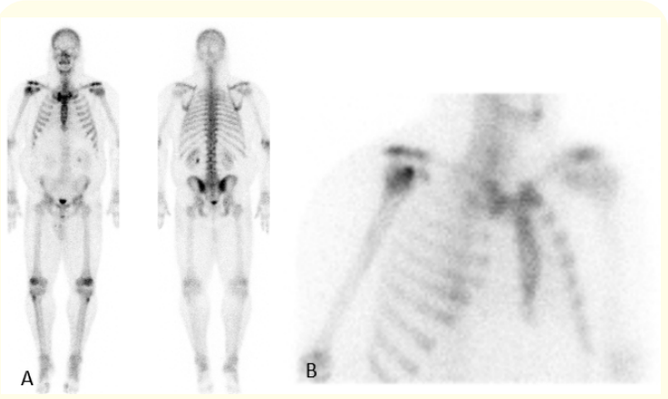

Figure 3: A. Full body bone scan with increased Technetium-99 uptake in the anterior right shoulder compared to the left side. B. Oblique view of the chest which shows increased technetium-99 uptake in the anterior right shoulder indicating increased metabolic activity in that area from the osteoid osteoma.

The imaging studies were reviewed with interventional radiology, and he was deemed a candidate for CT guided radiofrequency ablation treatment. He underwent radiofrequency ablation and on follow up was pain-free, not taking any NSAIDs, and was back to his normal activity level.

Discussion and Conclusion

While osteoid osteomas are a common type of primary bone, only 13% of osteoid osteoma patients were over the age of 30 [1]. The oldest patient in the literature with an osteoid osteoma was 77 but this lesion was in the hand [14]. Given the location of the osteoid osteoma in the humeral metaphysis and his age greater than 50 years old, this is an unusual presentation of osteoid osteoma.

One interesting aspect of the case was that the pain from the lesions improved with stoppage of his omega-3 fatty acid supplement. It was postulated that addition of polyunsaturated fatty acids, specifically eicosapentaenoic acid and docosahexaenoic acid would act synergistically with NSAIDs to prevent human cancer development and cardiovascular events in osteoid osteoma patients [15]. There have been no studies exploring the relationship between fatty acid supplementation and symptom response in OO patients. Given the conflicting theoretical benefits of polyunsaturated fatty acids (PUFAs) on OO and the patient's negative response to them, further investigation elucidating the risk and benefits of PUFAs is warranted.

Our patient's factor V Leiden is another interesting aspect of the case. There have been no other reports in the literature of osteoid osteoma patients having factor V Leiden as a co-morbidity. Factor V Leiden is usually an autosomal recessive condition where factor Va in the coagulation cascade develops a resistance to protein C,

an anticoagulant. It is unclear what impact this may have had on the pathogenesis of the tumor and the subsequent response to RFA treatment. This is an area where further investigation is warranted.

The RFA treatment is extremely well studied for treatment of OO and our patient underwent a successful RFA treatment without the need for repeat therapy.

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