Occult osteoid osteoma presenting as shoulder pain: a case report

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Abstract

Objective: The purpose of this case study is to describe the clinical course and treatment of a patient with recalcitrant shoulder pain and osteoid osteoma.

Clinical Features: A 28-year-old man had a 2-year history of progressively worsening shoulder and midscapular pain.

Intervention and Outcome: Before chiropractic consultation, he had been evaluated and treated by his family physician, an orthopedic surgeon, a neurologist, and a pain management specialist. The patient underwent arthroscopy with examination under anesthesia and debridement of a posterior labral tear and cervical spine epidural injections, but neither procedure relieved his symptoms. After seeking chiropractic care, presenting symptoms were reproducible during direct clinical examination; and an initial working diagnosis of secondary right glenohumeral impingement syndrome with coexisting scapulothoracic dyskinesia was made. After 2 weeks of chiropractic rehabilitation, therapy was stopped because of no change in symptoms. The patient was referred for orthopedic consultation. Another series of plain films were ordered, and follow-up magnetic resonance imaging revealed an osseous mass at the medial aspect of the proximal metadiaphyseal region of the right humerus, with a diagnosis of osteoid osteoma. The patient underwent radiofrequency thermoablation of the tumor nidus, which was unsuccessful and resulted in open surgical resection. Resolution of symptoms with minimal pain was reported 3 weeks after the surgery. Four years later, the patient's shoulder remains asymptomatic.

Conclusion: This case demonstrates that osteoid osteoma may present with clinical features that mimic common functional musculoskeletal conditions of the shoulder. Information from the patient history and diagnostic imaging are important for diagnosis and appropriate management. © 2012 National University of Health Sciences.

Introduction

Osteoid osteoma is a benign bone tumor of children and young adults between the ages of 10 and 25 with a 2:1 to 3:1 male to female ratio consisting of a well-

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demarcated core and a surrounding area of reactive bone formation.¹⁻⁶ There is a predilection of these tumors to arise in the femur and tibia, with a remaining equal distribution reported in the spine, hand, and foot.^{3,4} Atypical locations can occur, but are rare, as in this case, in the proximal metadiaphyseal region of the humerus^{1-4,7}; and osteoid osteoma is rarely reported in the medical literature.¹⁻⁴

As well, there is a paucity of osteoid osteoma cases in the chiropractic literature. There are only 2 cases of osteoid osteoma reported.^{8,9} No known cases in the chiropractic literature have been reported in the upper extremity.

As osteoid osteomas can mimic common musculoskeletal disorders, it is up to the clinician to gauge response to treatment and revisit the differential diagnosis when symptoms persist.

This case study describes the presentation, management, and outcome for a patient presenting with chronic, persistent shoulder pain and the pathway followed to arrive at the final diagnosis and treatment of osteoid osteoma.

Case report

A 28-year-old white man had experienced progressive right shoulder, arm, and midscapular pain for approximately 2 years. He reported that his symptoms began while performing overhead pull-down resistance training exercises that resulted in a mild onset of right axillary pain. He followed with his primary care physician, was prescribed anti-inflammatory and pain medication, and was given a referral to see an orthopedist.

Plain films of the right shoulder were read as negative. Results of follow-up magnetic resonance imaging (MRI) of the right shoulder were reported to be consistent with mild degeneration at the acromioclavicular joint, mild rotator cuff tendinopathy without tendon tear, and attenuation of the biceps tendon substance that may have represented some degree of partial tear/tendinopathy and some blunting of the inner margins of the labrum anteriorly and posteriorly. The patient underwent right shoulder arthroscopy with examination under anesthesia and debridement of a posterior labral tear. The surgery provided no relief for the shoulder pain. Follow-up plain film radiographs of the right shoulder were performed postsurgically and were read as negative (Fig 1).

The patient was referred for neurological evaluation 2 months after his shoulder surgery and given a primary diagnosis of moderate right C5-C6 radiculopathy and

Fig 1. Anteroposterior plain film right shoulder status postarthroscopy (May 2005) showing slight medial cortical expansion (black arrow).

mild to moderate bilateral cubital tunnel syndrome. An MRI of the cervical spine along with electrophysiological studies was ordered. The MRI of the cervical spine without contrast was interpreted as normal. Electrophysiological studies were performed, and it was determined that the patient exhibited signs and symptoms consistent with mild to moderate right and left C5-C6 radiculopathy and mild to moderate bilateral cubital tunnel syndrome. Cervical epidural steroid injections ($\times 2$) at the C5-C6 level followed. The epidural injections provided no relief of the shoulder pain. The patient then followed with his primary care medical physician and was subsequently referred to our chiropractic office for evaluation and treatment. Other than a 6-pack-year smoking history, the patient complained of nocturnal pain when sleeping on his right side and an overall pain rating of 10 on a 1 to 10 scale. He was 79 kg and 5'9" tall. He was currently taking naproxen and acetaminophen during the evening to help him sleep, and he took naproxen every 4 hours during the day.

Cervical spine examination revealed normal range of motion without pain at extremes and no evidence of tenderness or muscle spasm. Foraminal compression and distraction maneuvers were unremarkable. Inspection of the right shoulder revealed mild right-sided scapular winging and mild muscle wasting of the right deltoid musculature compared with the left. Acromioclavicular and sternoclavicular joints were comparable. Palpation revealed tenderness at the long head of the biceps tendon and teres minor. Active range of motion revealed full abduction, external rotation, and internal rotation bilaterally. There was notable dyskinesia of the right scapulothoracic articulation through repetitious abduction. Fatigue at 8 to 10 repetitions revealed a mild

increase in right scapular winging. Passive range of motion of the right shoulder revealed joint crepitus of the scapulothoracic articulation. Cross-arm abduction (acromioclavicular grind test) created mild right axillary pain, as did apprehension (external rotation and abduction). Neer impingement reproduced right anterior shoulder pain. Load and shift procedures of the right glenohumeral joint revealed laxity in the anterior to posterior and posterior to anterior vectors. Lift-off test reproduced anterior right shoulder pain. There was notable rapid scapular retraction after 3 to 4 active resisted push-ups. Muscle stretch reflex, sensation, and strength in the upper extremities were symmetric and without any focal deficits. Measurement of the extremities demonstrated that they were symmetric and without atrophy, with the exception of the right deltoid.

In summary, orthopedic examination of the cervical spine had a normal result and did not provoke shoulder pain. Shoulder evaluation revealed normal range of motion and neurovascular findings. There were some notable scapular dyskinesis, positive impingement signs, tenderness of the biceps tendon, and glenohumeral laxity with load and shift maneuvers. An initial working diagnosis of secondary right glenohumeral impingement syndrome with coexisting scapulothoracic dyskinesis was determined. Plain film radiographs were not ordered, as the examination findings did not support additional films and they were performed within the previous year and were read as normal.

Based upon the history and presenting symptoms that were reproducible during direct clinical examination, the patient underwent a 2-week period of rehabilitation

Fig 2. A, Anteroposterior plain film right shoulder (April 2006), showing marked medial cortical expansion (black arrow). B, Magnetic resonance imaging sagittal stir fast spin echo (FSE) (April 2006) demonstrating widespread bone marrow edema (arrows). C, Magnetic resonance imaging coronal stir FSE (April 2006) showing widespread bone marrow edema (arrows). D, Magnetic resonance imaging axial T2 FSE (April 2006) with evidence of hyperintense rim surrounding low signal (arrows).

Fig 3. Both images are pre-surgical post biopsy images. A, Magnetic resonance imaging oblique sagittal fast inversion recovery (FIR) (August 2006) revealing hyperintense signal change surrounding low signal focus (arrows). B, Magnetic resonance imaging oblique axial FIR (August 2006) showing oval hyperintense lesion (arrows). It is possible that the lesion could be at the biopsy site; however, the biopsy came back as “inconclusive but having inflammatory characteristics.”

to strengthen rotator cuff and lower trapezius muscles. Therapy was stopped after 2 weeks secondary to no change in his symptom profile. The patient was referred for a second orthopedic consultation with a different shoulder specialist. Plain films were ordered and were remarkable for focal cortical thickening at the medial aspect of the proximal metadiaphyseal region of the right humerus; and, consequently, a follow-up MRI

revealed abnormal nonspecific bone marrow signal change in the same region as well as adjacent focal signal change and thickening of the medial cortex (Fig 2). These findings prompted another referral to an orthopedic oncologist wherein a bone biopsy was performed that was nondiagnostic but suggestive of an inflammatory process. A second opinion with another orthopedic oncologist led to repeated MRI and a whole-body bone scan. The second MRI findings were consistent with nonspecific abnormal bone marrow edema-like signal in the right proximal metadiaphyseal region along with a central 1-cm oval hypointense lesion in the marrow cavity surrounded by a rim of hyperintensity on postcontrast images. In addition, in the same region, there was an 8-mm oval hyperintense lesion in the medial cortex (Fig 3). The total body bone scan revealed a solitary, moderately large, moderately intense lesion visualized at the medial aspect of the proximal metadiaphysis of the right humerus (Fig 4). Noncontrast computed tomography (CT) of the right proximal humerus was performed and was remarkable for a lateral cortically based

Fig 4. Nuclear bone scan demonstrating focal radionuclide uptake in the proximal shaft of the right humerus on the medial side.

lucency measuring $1.3\text{ cm} \times 5\text{ mm}$ representative of the previous biopsy defect and a more medial 1-cm cortically based lucency surrounded by thick periosteal reaction most likely representative of osteoid osteoma (Fig 5). The patient underwent radiofrequency thermoablation by passing a Jamshidi needle through the previous biopsy hole in the lateral cortex of the humerus and then into the lesion medially. The probe was activated to 90°C for three 2-minute cycles. At 6 weeks postsurgical follow-up, there was no change in the patient's symptoms. It was determined at that time that there was some uncertainty of the etiology of the patient's pain, prompting retrospective review of previous imaging and repeated plain film radiography of the right humerus. The plain films demonstrated a

Fig 5. A, Computed tomograph coronal (August 2006) demonstrating cortically based lucency surrounded by thick periosteal reaction (arrows). B, Computed tomograph axial (August 2006) demonstrating a radiolucent lesion within the medial cortical margin (arrows).

Fig 6. Plain film anteroposterior shoulder (November 2006) demonstrating subtle depiction of an ovoid lucency (arrow).

1-cm ovoid lucency within the proximal shaft of the right humerus (Fig 6). There was uncertainty whether this lucency represented a residual defect from biopsy, residual osteoid osteoma, or residual tract from radiofrequency ablation. The final surgical procedure included curettage and grafting with allograft of the right proximal humeral lesion. By reduction and rotation, the surgeon was able to present an osseous prominence that was felt to overlie the lesion. The small osseous bump was osteotomized and revealed what looked like a nidus of tumor underneath. It was subsequently thoroughly curetted, and then a power burr was used to extend the curettage. After copious irrigation of the wound, approximately 2 mL of Allomatrix bone graft substitute (Wright Medical Technology Inc., Arlington, TN) was injected to consolidate the hole. Relative complete resolution of symptoms with minimal pain was reported 3 weeks after this procedure. Four years later, the patient remains asymptomatic.

Discussion

The term *osteoid osteoma* was introduced into the medical literature in 1935 by Jaffe¹⁰ who he described

the osteoblastic tumors as being composed of osteoid and atypical bone. Osteoid osteoma can occur in any bone; however, there is a predilection for the lower extremities, with 50% or more occurring in the femur and tibia.^{1,3,7,11,12} Most these tumors occur in the metadiaphyseal region of long bones; the remaining 30% are equally distributed among the spine, hand, and foot.^{1,3,7} Atypical locations include the skull, ribs, ischium, mandible, patella, and proximal humerus.^{1,3,4} Osteoid osteomas are classified as cortical (most prevalent), cancellous, and subperiosteal, with the latter two typically arising in an intraarticular or juxtaarticular location.³ Extraarticular cortical tumors demonstrate a radiologic appearance of a representative centralized radiolucent “nidus,” rarely greater than 1.5 cm, surrounded by a sclerotic rim in the shaft of a long bone.^{1,3-5,10} Unusual locations and equivocal early radiological findings are common and can result in diagnostic difficulties, particularly those that are intraarticular.^{3,4,7,13} Patients typically have mild initial pain that is progressive over time and nocturnal pain that is relieved with anti-inflammatory medication.^{3-5,7,13,14} The radiolucent nidus contains nerve fibers, vascular elements, and prostaglandins that may be responsible for chronic reactive change in the surrounding bone resulting in periosteal sclerosis and synovitis.^{3,15} The prostaglandin concentration is thought to be responsible for the constant and severe pain because the vasodilatory effect increases intracortical pressure within the nidus.^{15,16} The expression of cyclooxygenases (COX-1 and COX-2) is thought to be the source of high levels of prostaglandin E₂, COX-2 being one of the main mediators of pain and inflammation effects on the surrounding tissues.^{3,15,16} Nonsteroidal anti-inflammatory drugs and salicylates may decrease pain symptoms secondary to the inhibition of prostaglandin formation. The differential diagnosis of osteoid osteoma includes osteoblastoma, Brodie abscess, stress fracture, bone island, eosinophilic granuloma, and malignant lesions such as Ewing sarcoma and osteosarcoma.^{2,3,10,17} Treatment classically involves open surgical resection, but there are relatively newer surgical techniques such as radiofrequency thermal ablation that are also used.^{1,8,11,12}

Osteoid osteomas occurring in the proximal humerus are rare.^{1,3,4} More commonly, osteoid osteomas occur in the cortex of long bones, particularly the femur and tibia.^{1,3,7,11,12} Extraarticular cortical tumors demonstrate a radiological appearance of a nidus surrounded by a sclerotic rim.^{1,3,5,6,8} Cancellous and subperiosteal osteoid osteomas are found in an intraarticular or juxtaarticular location wherein sclerosis may be absent or mild to moderate and may be distant from the lesion,

unlike the classic cortical tumor.^{3,12} This case represented a cortical osteoid osteoma residing in the proximal metadiaphyseal region of the humerus with bone marrow edema extending through both extraarticular and intracapsular regions. This may be important for 2 reasons. First, the location of the tumor does not neatly fit into the extraarticular/intracapsular classification. Rather, it borders the 2 regions, therefore giving it characteristics of both an intracapsular and extraarticular lesion. Moreover, a rare location combined with extensive bone marrow edema allows the possibility of second hidden nidus, one cortical and one cancellous. Second, this possibility might partially explain why the thermal radiofrequency ablation procedure was unsuccessful and deeper curettage was successful in excision of the tumor nidus/nidi.

Initial plain film radiographs can be equivocal when working up an osteoid osteoma, and MRI is inferior to CT regarding localization and surrounding morphology of the tumor nidus.^{3,4,7,10,12,13,15,18} This being said, with initial MRI findings consistent with proximal humeral bone marrow edema, recognizing this and following up with a CT scan may have led to a significant decrease in morbidity and a clear path to diagnosis and treatment. Bone scintigraphy is valuable in assessing osteoid osteoma where initial radiographs are negative or symptoms are atypical.^{3,19}

Treatment of osteoid osteoma is excision or ablation of the tumor nidus. Percutaneous radiofrequency thermal ablation is a relatively newer surgical technique that is thought to carry less surgical morbidity than open surgical excision; however, surgical excision is still the preferred treatment method.^{1,3,12,15,18} Return of symptoms can occur if removal of the tumor nidus is incomplete. In this case, the patient underwent unsuccessful radiofrequency ablation that may be due in part to incomplete nidus ablation or, as mentioned above, a multicentric presentation of tumor nidi, the latter being more amenable to deeper surgical resection.

Osteoid osteomas have been reported in young, healthy individuals and athletes with the initial presentation of pain secondary to exercise and/or minor trauma.^{3-5,7,10} Moreover, initial symptoms and clinical features of some bone tumors have been reported to become symptomatic from routine physical activity and/or exercise, commensurate with this case.^{11,20} What makes this case unique is that the presenting symptoms were reproducible during direct clinical examination, thus dually supporting and masquerading as a common musculoskeletal disorder of rotator cuff impingement syndrome.

The management of this case raises several interesting clinical and diagnostic issues. It is understandable that the initial plain films could have been read as normal secondary to the subtle changes in the medial metadiaphyseal cortex; however, this cannot be said about the initial MR images. Retrospective review of the initial MR images revealed bone marrow edema in the metadiaphyseal region of the right proximal humerus. Although the tumor nidus is not visualized, bone marrow edema was not reported and, subsequently, may have prolonged the diagnosis. Moreover, osteoid osteomas often mimic other etiologies, as in this case. Complicating diagnostic matters were the rare location for this lesion, the complex anatomy of the shoulder, and the patient relating the symptoms to trauma.

His initial neurologic and orthopedic findings in our clinic suggested glenohumeral joint laxity determined by load and shift maneuvers as well as positive impingement signs, painful isometric rotator cuff strength testing, and scapular dyskinesis. Because of his clinical findings and extensive past medical workup, we took the liberty of not irradiating the patient, as he had previous plain films within the previous year that were read as normal. Treatment was geared toward strengthening of the rotator cuff and muscles used for scapular stabilization after determining an initial working diagnosis of secondary impingement syndrome. *Impingement* can be defined as pressure on the rotator cuff or overlying bursa by subacromial spur formation or secondary to abnormal glenohumeral joint laxity as was determined in this case. Aberrant mechanical movement patterns of the glenohumeral joint secondary to capsular laxity can perpetuate chronic impingement of the rotator cuff during normal purposeful movement of the upper extremity, thus driving chronic shoulder pain. Rotator cuff attenuation can also produce nocturnal pain, particularly when sleeping on the affected side. These findings culminated in what appeared to be commensurate with the working musculoskeletal diagnosis. However, there was no response to treatment after 2 weeks of shoulder rehabilitation.

This information highlights several points about patient management. First, treatment should cease if the patient is not responding to conservative treatment in a timely fashion. This point is dependent upon the clinician's experience relating to usual and customary response to common musculoskeletal disorders and therefore the doctor's heightened index of clinical suspicion.

Second, glenohumeral capsular laxity, confirmed by clinical examination, may not necessarily result in

pain generation from subacromial cuff impingement as was illustrated in this case. This could be due in part to the relatively young age of the patient and early-stage nonsymptomatic cuff attenuation. Moreover, a major portion of the shoulder examination involves active, passive, and resisted movement. This patient experienced pain with impingement tests and resisted movements (Neer, Hawkins, empty can, isometric muscle testing of the rotator cuff). Passive movements of the glenohumeral joint to end range and isometric resistance maneuvers of the rotator cuff impart tensile and rotational forces through the muscles, tendons, and upon the periosteum at the tendinous insertions. It is possible that the examination procedures affected edematous bone with an expansile cortical lesion, but identified the rotator cuff as the primary pain generator masquerading as secondary impingement syndrome. Furthermore, after open resection of the tumor, there was near-complete resolution of symptoms, reflecting equivocal contribution from the cuff. This illustrates the need for close scrutiny of the patient's subjective complaints and appreciation of differential diagnosis when examination findings support a common musculoskeletal diagnosis not responding to care.

In summary, osteoid osteoma is a benign skeletal tumor in which the clinical and radiological picture may be unclear, thus delaying diagnosis. Patient complaints alone are often diagnostic: night pain predominant, of increasing severity, and relieved by nonsteroidal anti-inflammatory drugs. If initial radiographs are equivocal, bone scintigraphy is recommended. Although nonspecific, scintigraphy locates the lesion allowing for follow-up conventional CT for precise location of the nidus. Computed tomography is not only used for nidus location but may be used to provide surgical guidance.

Limitations

There are several limitations to this case report. Initial plain films were not available for this case, limiting complete characterization of tumor growth over time. In addition, it is not common for these tumors to reside in the proximal metadiaphyseal region of the humerus, which limits discussion relating to usual and customary behavior of osteoid osteomas. Furthermore, it is not certain that the cause of shoulder pain was generated from the osteoid osteoma; however, based upon the patient's response to surgery, we feel that it was likely.

Conclusion

This case demonstrates that osteoid osteoma may present with clinical features that mimic common functional musculoskeletal conditions of the shoulder. Information from the patient history and diagnostic imaging are important for diagnosis and appropriate management.

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