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CASE REPORT

Persistent wrist monoarthritis: down to the bone

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SUMMARY

A minority of osteoid osteomas are found to be juxta-articular and within the small bones of the wrist. We present a 30-year-old man diagnosed with an osteoid osteoma of the lunate bone, presenting with 3 years of left wrist pain, swelling and reduced range of motion. Given the patient's background and laboratory testing, consideration was given to both inflammatory and infectious causes and the diagnosis was delayed, requiring repeat interval imaging and assisted by multiple imaging modalities. Management by surgical excision led to resolution of pain and swelling. In cases of a prolonged isolated monoarthritis, juxta-articular osteoid osteoma should be considered in the differential.

BACKGROUND

Osteoid osteoma, a benign tumour of bone, generally occurs in the diaphysis or metaphysis of long bones.¹ However, it has also been described in areas closer to a joint, termed juxta-articular osteoid osteoma. This particular case occurred in the lunate, a site less commonly reported, diagnosed only after serial and multimodality imaging and presented as a wrist monoarthritis, for which the differential tends to focus on more common causes of an inflammatory arthritis, the management of which is radically different. This case highlights the need for ongoing assessment and reassessment when there is uncertainty over the correct diagnosis and potential reasons for delayed diagnosis in this case are explored.

CASE PRESENTATION

A 30-year-old man presented to the Rheumatology clinic with a 3-year history of left wrist pain, swelling and reduced range of motion. It had started while lifting weights at the gym and due to persistence of the pain over a number of weeks; he sought medical attention and was referred to Orthopaedics at an outside institution. Initial imaging with MRI revealed possible avascular necrosis of the lunate (Kienbock's disease) with management consisting of splinting and physical therapy to no avail. Given the non-response, he was seen at a second Orthopaedic practice where he proceeded to wrist arthroscopy, the report commenting on synovitis of the wrist joint and tenosynovitis of multiple tendons. Samples were not submitted for pathological review. The patient remained symptomatic and therefore sought the advice of a third Orthopaedic surgeon where he underwent an intra-articular steroid injection to

the left wrist without benefit. Laboratory testing consisting of erythrocyte sedimentation rate (ESR), C reactive protein (CRP), Lyme disease antibodies, antinuclear antibody and rheumatoid factor were sent and all within the reference range or negative. Plain radiograph of the left wrist was unremarkable for any acute or chronic pathology. At that point, he was referred to the Rheumatology clinic for further assistance with management.

On initial assessment, he reported ongoing pain centred over the dorsum of the left wrist that was present consistently throughout the day. He had tried various non-steroidal anti-inflammatory drugs (NSAIDs) and found naproxen 500 mg two times a day, alongside cold compresses and rest most effective, although this regimen did not completely resolve his pain. Symptoms were not worse in the morning and there was no stiffness. No other joints were involved and there were no systemic symptoms such as fever, chills, night sweats, rash suggestive of psoriasis or changes in bowel habit, such as diarrhoea. Of note, the patient is originally from Nepal and reported that approximately 7 years ago, a roommate had been diagnosed with tuberculosis (TB) and that at the time he was negative on testing. Examination was unremarkable save for the left wrist which revealed well healed arthroscopy scars, tenderness, fullness and warmth over the dorsum of the wrist, with range of motion restricted to 20° flexion and 40° extension.

Differential diagnosis at that time included a systemic inflammatory arthritis such as rheumatoid arthritis or a seronegative spondyloarthropathy, chronic infection such as TB and less likely, crystal disease given the longevity of symptoms. Further laboratory testing again revealed ESR and CRP within the reference range. Additionally, antibody to cyclic citrullinated peptide was negative; however, interferon-gamma release assay (IGRA) for TB returned positive with chest X-ray negative. Assessment of the synovial fluid was negative for crystals or growth of bacteria, fungi or acid-fast bacilli (AFB).

After consultation with colleagues in Infectious Disease, repeat interval MRI of the left wrist was obtained revealing a well-defined intraosseous focus of abnormal signal in the dorsal lunate with adjacent productive bone and oedema, suggestive of an osteoid osteoma (figure 1). This was further characterised on CT scan which confirmed the presence of a 6 mm articular defect in the dorsal lunate, with a 4 mm density within it representing the calcified nidus



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Figure 1 Coronal proton density sequence MRI of the left wrist revealing a well-circumscribed focus of abnormal signal in the dorsal lunare.

and mild surrounding sclerosis, all in keeping with an osteoid osteoma (figures 2 and 3).

TREATMENT

Once the diagnosis of osteoid osteoma was suspected by imaging, the patient underwent surgical excision by our Orthopaedic colleagues.

OUTCOME AND FOLLOW-UP

Pathology of the excised tissue revealed a relatively circumscribed bone-forming lesion demonstrating interlacing trabeculae, sheets of woven bone with partial mineralization in a moderately cellular fibrovascular stroma with conspicuous osteoblastic rimming and scattered osteoclasts, all consistent with osteoid osteoma (figure 4). Assessment of the sample for infection including AFB was negative. At his initial postoperative check at 1 week, the patient reported that his pain had completely resolved and that he was not using any analgesia of any kind. He started a programme of occupational therapy in order to regain function. At a further clinic visit 4 weeks after excision, he remained pain-free.

DISCUSSION

Osteoid osteoma is a benign bone tumour, originally described in 1935,² comprising approximately 12% of benign skeletal



Figure 2 Coronal CT of the left wrist demonstrating the articular defect in the dorsal lunare, with a calcified nidus within it.

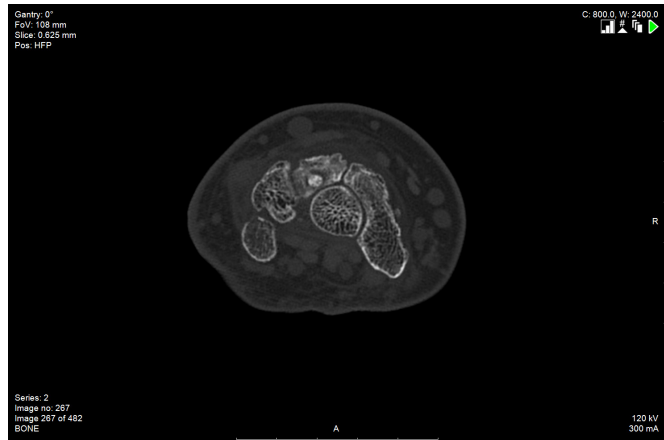


Figure 3 Axial CT of the left wrist demonstrating the articular defect in the dorsal lunare, with a calcified nidus within it.

tumours with a predilection for younger males. Pain is the most common presenting symptom, usually worst at night and often extremely responsive to NSAIDs.^{1,3} The majority are extra-articular, found in the diaphysis or metaphysis of long bones especially of the lower extremity (femur and tibia) and localised within the bone cortex. Juxta-articular or intra-articular osteoid osteomas are estimated to comprise 5.2%–10% of all osteoid osteomas, can be subchondral, intracortical, intramedullary or subsynovial and have been described in a number of locations including the small bones of the feet, ankle, knee and hip joints.^{4–6} The first definitive descriptions in the hand and wrist were reported in the 1950s.^{7,8} These original descriptions did not include lesions of the lunare but these have also subsequently been reported.^{9–15} Management consists of either surgical excision or percutaneous intervention using a variety of methods such as radiofrequency-ablation or cryo-ablation.¹

Diagnosis of osteoid osteoma, including those of the hand and wrist as well as other juxta-articular or intra-articular locations, can be difficult due to atypical presentations, infrequent occurrence or symptoms, signs and laboratory testing that may lead to initial misdiagnosis. In this case, there were some atypical features in the history such as pain that was not worse at night and not significantly improved with NSAIDs. Additionally, there

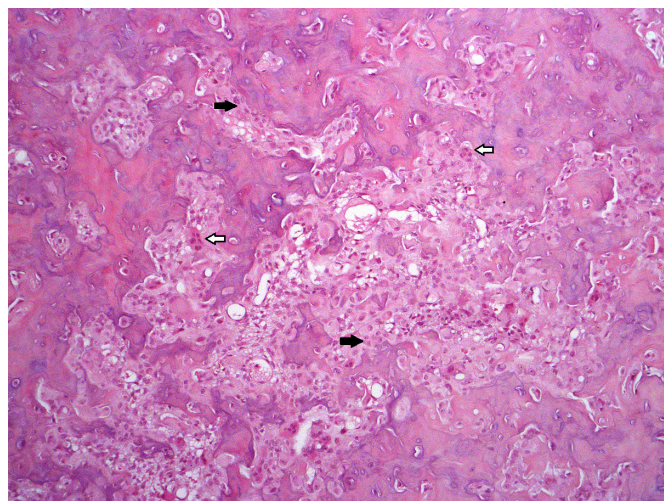


Figure 4 Pathology slide demonstrating interlacing trabeculae, sheets of woven bone with partial mineralisation, osteoblastic rimming (black arrows) and scattered osteoclasts (white arrows).

was initial misdiagnosis with concern for Kienbock's disease and treatment initiated for that. One other case in the literature was also initially diagnosed as Kienbock's disease.¹⁵ Given the swelling and synovitis/tenosynovitis noted on wrist arthroscopy, consideration was also given to this as a first presentation of a systemic inflammatory arthritis; however, there were no associated features suggestive of a systemic inflammatory arthritis and it is well known that juxta-articular or intra-articular osteoid osteomas can present with synovitis and effusion, more so than extra-articular osteoid osteomas.^{4 16–18} Finally, the patient also tested positive on IGRA, giving consideration to TB of the wrist. TB often presents as a monoarthritis, but more usually of the weight-bearing joints such as the hip and knee.¹⁹ In this case, the synovial fluid analysis and operative samples did not support the diagnosis.

Difficulty in diagnosis can also be due to choice of imaging modality used, with plain radiographs, CT, scintigraphy, single photon emission CT, ultrasound and MRI all being used. In this patient, initial plain radiograph was unrevealing and the next imaging modality was MRI, in order to better image the surrounding soft tissues rather than the bone itself. However, it has been shown that for diagnosing osteoid osteoma, this is inferior to CT scanning. With MRI, it has been noted that the appearances can be variable with surrounding bone marrow and soft tissue oedema or periostitis that can mimic an infectious or malignant process.¹ Furthermore, a retrospective study estimated the rate of missed diagnosis of osteoid osteoma on MRI compared with other imaging modalities (plain radiograph, CT or scintigraphy) at 35%.²⁰ In our case, only when the repeat MRI was suggestive of an osteoid osteoma that both a repeated plain radiograph (now reporting a dorsal irregularity in the lunate) and CT were performed, which confirmed the suspicion raised by MRI. Some authors have argued that for cases of juxta-articular osteoid osteoma, CT imaging should be the gold standard.¹

Overall, this is a case of a well-defined pathology occurring in a lesser recognised location with a number of distractors (examination with synovitis, initial imaging findings, history of exposure to TB) which led to other pathological processes being considered (Kienbock's disease, systemic inflammatory arthritis, infection) before the final diagnosis was eventually made. This highlights the point that when more common diagnoses cannot

completely account for the symptoms and signs presented, the diagnostic process should continue.

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Learning points

- ▶ Osteoid osteoma is a benign bone tumour that less commonly affects the bones of the hand and wrist.
- ▶ When situated close to a joint, in addition to pain, osteoid osteomas can present with symptoms mimicking an inflammatory monoarthritis and therefore should be considered in the differential once more common causes have been excluded.
- ▶ The choice of imaging modality directly impacts the ability to correctly diagnose osteoid osteoma with CT being the gold standard.

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