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Case Report

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Metachronous osteoid osteoma in adjacent proximal phalanges: a case report and literature review

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ABSTRACT

Osteoid osteoma (OO) is a benign bone tumor commonly affecting long bones, and in rare cases, involves small bones such as hand phalanges. The presenting symptom is usually nocturnal inflammatory pain. OO is in the majority of cases a solitary lesion, with very rare reported cases of metachronous lesions in different locations. We present a case of a 31-year-old female patient who presented for severe pain and swelling over her fourth finger proximal interphalangeal joint (PIPJ) of the right hand. After thorough radiological investigations, a presumptive diagnosis of OO was made and the patient underwent surgical excision and curettage with resolution of her symptoms post-operatively. Six years after remission, the patient presented again to our clinics with the same clinical picture in the third finger PIPJ of the right hand. Clinical and radiological investigations revealed a metachronous OO in the adjacent phalanx of the previous surgical site. Similar management consisting of excision and curettage was performed, resulting in a symptom-free follow up. To our knowledge, this is the first case of metachronous OO in adjacent proximal phalanges of the hand. This case highlights the importance of considering metachronous OO in the differential diagnosis of patients previously diagnosed and treated for OO and presenting with a similar clinical and radiological picture.

Keywords: OO, Multifocal, Metachronous, Hand

INTRODUCTION

Osteoid osteoma (OO) is a common benign tumor associated with moderate to severe inflammatory pain, with unknown etiology, accounting for 3% of all primary bone tumors and 10-13% of all benign bone tumors, mainly affecting children and young adults.[1,2]

OO typically develops in the long tubular bones, particularly the femur and tibia (around 50%) with only 5% to 10% in the hand and wrist.[3,4] This lesion, termed a nidus,[5] is usually sub-centimetric and exhibits a round or oval shape with a central region of mineralization surrounded by a variable zone of sclerosis.[6] Commonly associated with severe localized pain, mainly intensified at night and relieved by nonsteroidal anti-inflammatory drugs especially salicylates, OO induces an inflammatory and vasomotor reaction, along with sclerosis in the adjacent bone due to prostaglandins produced by the tumor.[4,7,8,9] OO may be challenging to detect on plain radiographs, especially if small or obscured by reactive bone sclerosis, necessitating the use of thin cuts computed tomography (CT) for early identification and to guide RFA.[5] Magnetic resonance imaging (MRI) is deemed more sensitive than CT scans, particularly in the context of hand phalanx OO.[3] The imaging features of OO include a small, round and hyperemic tumor surrounded by sclerotic reactive bone. On a microscopic level, the nidus contains immature osteoid trabeculae with active osteoblastic rimming, along with osteoclasts, capillaries, and nerve

fibers.[11,12] In some instances, OO can be multiple, presenting as either multifocal lesions within the same skeletal segment or synchronous multi-centric lesions in different bones. Furthermore, a single lesion with more than one nidus is considered "OO with multicentric nidus.[5] Despite its benign nature, OO is traditionally treated in order to relieve pain and prevent growth disturbances in immature skeletons. While surgical procedures such as curettage or en-bloc resection are considered invasive, percutaneous radiofrequency ablation (RFA) is presently standard of care, depending on tumor location and size.[8,10]

Phalanx OO is rare, often leading to delayed diagnosis and treatment due to unusual symptoms and location, with only a minority of cases displaying evidence of recurrence.[4,13]

In this article, we present the case of a 31-year-old female patient with a diagnosis of OO located on the distal aspect of the proximal phalanx of her right fourth finger. Management involved surgical excision, with symptom resolution at follow up visits. Six years later, the patient returns with the same symptoms in the adjacent finger. Imaging studies suggested a new diagnosis of OO on the distal aspect of the proximal phalanx of her right third finger. Similar surgical treatment was performed with full symptom resolution on follow-up.

CASE REPORT

A 31-year-old previously healthy female patient presented with a five months history of spontaneous, intermittent, non-traumatic right fourth finger pain.

She had worsening of her symptoms at night, with no irradiation, fever, or other associated symptoms. Pain was gradually increasing, and eventually became unresponsive to non-steroidal anti-inflammatory drugs (NSAIDs), particularly salicylates. At first clinical examination, diffuse tenderness of the right fourth finger was noted, unrelated to movement, with palpable mild swelling observed. No other deformities were seen. Plain radiographs of the hand and finger showed no bony lesions or abnormalities (Figure 1).

Figure 1 (A and B): X-ray of the 4th right finger, lateral view and anteroposterior view.

Ultrasound of the fourth right finger was done showing no soft tissue abnormalities around proximal interphalangeal (PIP) joint. A complementary CT scan of the right hand showed a 2 mm cortical erosion with central micro-calcification, with net borders, suspicious of OO, at the ulnar head of the fourth proximal phalanx head (Figure 2).

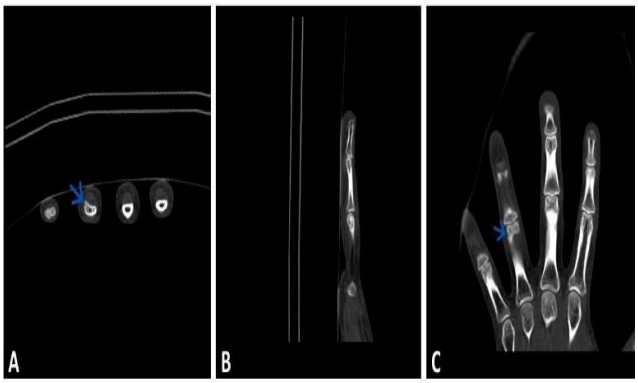

Figure 2 (A-C): Axial, sagittal, and coronal views from the CT scan of the right hand showing a 2 mm cortical erosion with central micro-calcification with net borders at the ulnar head of the fourth proximal phalanx head (Blue arrow).

In addition, scintigraphy of the right hand showed a net hyper-metabolic inflammatory focus of the fourth PIP joint, further confirming the diagnosis (Figure 3).

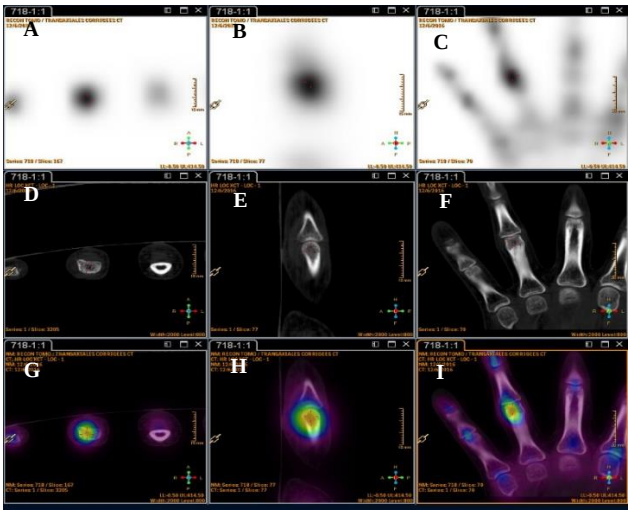

Figure 3 (A-I): Bone scintigraphy of the right hand showing an increased uptake at level of 4th proximal phalanx.

The patient's symptoms were affecting her daily living and seeing that her symptoms are no longer responsive to NSAIDs, a decision was made for surgical management. The intervention was done under general anesthesia, and the lesion was located at the ulnar border of the distal aspect of the fourth proximal phalanx of the right hand using the different CT scan thin cuts and under fluoroscopic intra-operative guidance. Curettage with safe

margins was done, the articulation was stable and the medial collateral ligament insertion was spared, with some bundles detached for exposure, which were re-sutured to the distal proximal aspect of the joint capsule. At one- and three-month's follow-up, patient had full resolution of her symptoms with regain of painless range of motion of PIPJ.

Six years later, the patient returns to the clinic with the same previous symptoms, namely inflammatory type of pain at the level of the 3rd finger PIPJ of the right hand. Patient had a similar physical examination, and reports alleviation of her symptoms with NSAIDs. She also reports epigastric pain since starting the course of NSAIDs. Plain radiographs were done showing no bony abnormalities at the level of the third PIPJ (Figure 4).

Figure 4 (A-C): Xray of right hand, anteroposterior oblique, and lateral views of site of previous excision at level of the fourth proximal phalanx (Blue arrow).

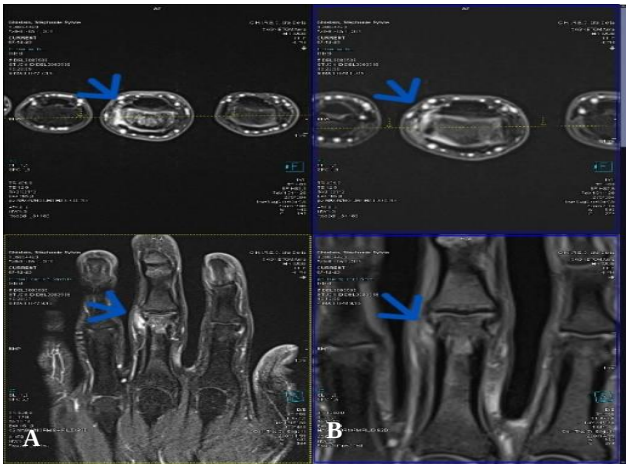

Figure 5 (A and B): MRI of the right hand showing moderate capsular and ulnar collateral ligament swelling at level of 3rd PIPJ with soft tissue edema (Blue Arrow).

This was complemented by an MRI, showing moderate capsular and ulnar collateral ligament swelling at the level of the PIPJ, associated with soft tissue edema. There were no signs of mass, rupture or disinsertion (Figure 5). Additionally, a bone scintigraphy was performed, showing an intense focus of hyper-fixation of the distal end of the proximal phalanx of the third right finger, suspicious of OO (Figure 6). CT scan with thin cuts showed a 3 mm lesion with a calcified center surrounded by reactive bone sclerosis, in favor of OO (Figure 7).

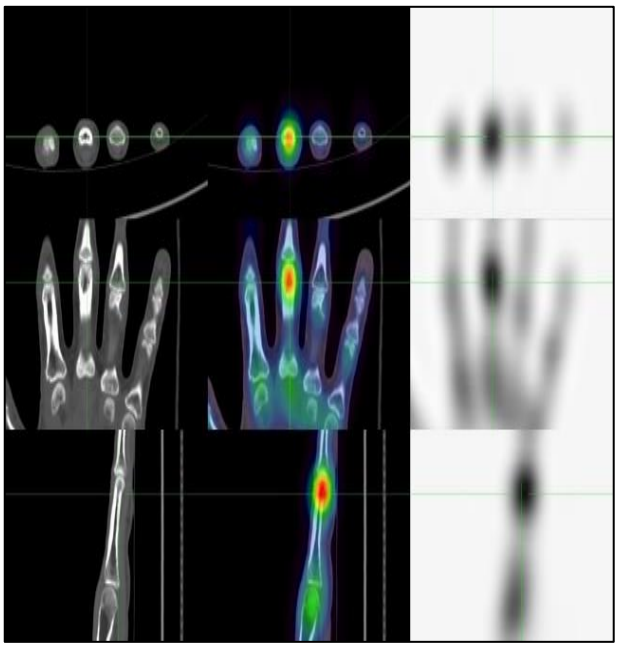

Figure 6: Bone scintigraphy of the right hand showing an increased uptake at the level of the third proximal phalanx.

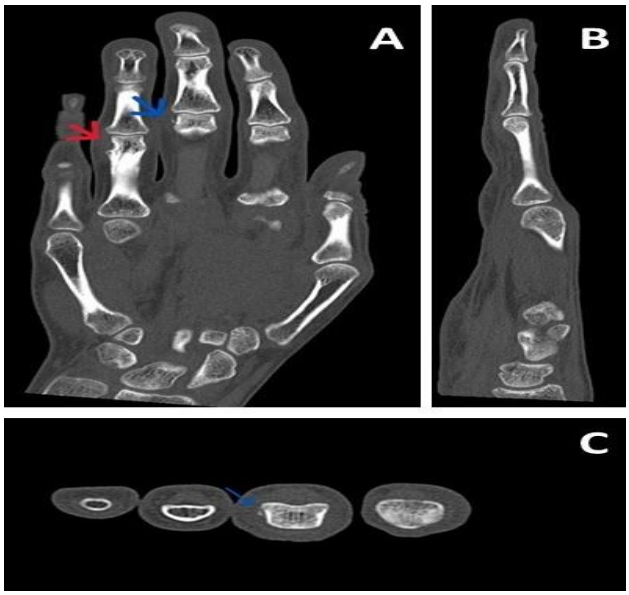

Figure 7 (A-C): Coronal sagittal, and axial cuts from the CT scan of the right hand showing a 3 mm lesion with a calcified center surrounded by reactive bone sclerosis highly suspicious of OO (Blue arrow). Site of previous excision at the level of the fourth proximal phalanx is also seen (Red arrow).

After discussing the possible therapeutic options with the patient, it was decided to opt for a surgical treatment by

excision and curettage, under fluoroscopic guidance, with an uneventful post-operative recovery (Figure 9 and 10). At one- and three-month's clinical follow-ups, the patient reports resolution of her symptoms, regaining painless full PIPJ range of motion.

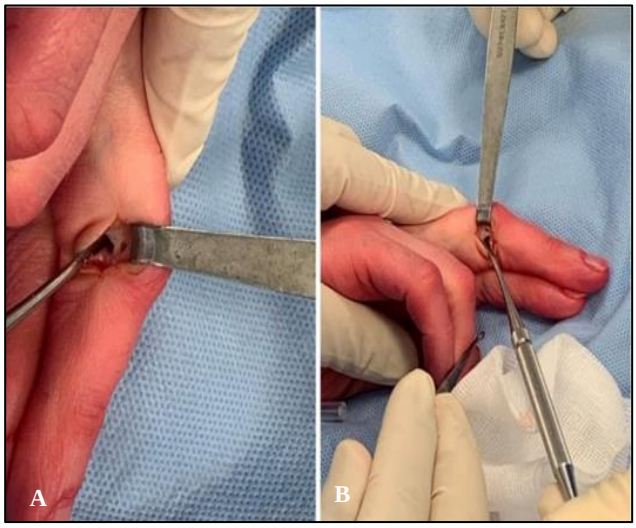

Figure 8 (A and B): Intra-operative images.

Figure 9 (A and B): Intra-operative fluoroscopic control.

DISCUSSION

Although the mechanism for OO development in the hand remains unclear, history of trauma is reported in 20-50% of patients with wrist and hand OOs.[9,14,15] In a retrospective review done by Simon et al on 37 cases of hand OO, almost 60% involved the phalanges, 24% affecting the metacarpals and 16% at the level of the carpal bones.[16] The most common location of OO is around the hand's midline axis, which passes through the scaphoid, capitate and the long finger ray, with OO incidence decreasing with increasing distance from the latter axis.[17]

OO affecting the wrist and hand most commonly occurs in young adults with the age between 23 and 35 years.[9,14,18] The average time to diagnosis ranges between a year to 20 months, with distinctive features allowing earlier diagnosis including nocturnal inflammatory pain relieved by NSAIDs, particularly salicylates.[9,14,18,19] When it comes to radiological diagnosis, as discussed above, high resolution thin cuts CT scan of the phalanges remains the gold standard, since plain radiographs are rarely sufficient, especially in hand OOs. MRI is a more sensitive imaging tool, providing additional surrounding soft tissue findings and inflammatory reactions, especially with lesions close to the intramedullary canal. However, this modality remains limited by availability and cost.

The treatment of OO of the hand is mostly surgical, with conservative treatment dying out of favor, consisting of chronic NSAID use for up to 3 years of symptomatology, not to mention the mental burden and fatigue, as well as its cost on healthcare systems.[20] While radiofrequency ablation (RFA) is a cornerstone in the management of OO, predominantly in long tubular bones, it presents a high risk of tissue injury at the level of the hand and phalanges, due to the close proximity of tissues with little to no frank subcutaneous layer. Some studies showed that RFA use in phalanx lesions could result in chronic pain and persistent swelling, mostly due to thermal damage. Surgical treatment consists of curettage excision or en bloc resection.[21,22] Curettage may be beneficial in cancellous bone lesions, with a higher rate of recurrence in lesions with cortical involvement.[3] En-bloc resection seems to result in better outcomes, especially in terms of recurrence rate. Multiple surgical techniques have emerged, adding or modifying certain surgical gestures: For instance, the use of a high-speed burr around the margins decreases the risk of incomplete resections, through mechanical and thermal effects.[3] The recurrence rate of surgically treated OO is around 12%.[9,15,18] Bone grafting is generally considered in cases of recurrence, treated with repeat surgical excision. The latter leads to wider margins of excision, making bone grafting essential to prevent the increased risk of postoperative fractures.[19]

OO are solitary in most cases, with very rare (<1%) occurrence of multiple tumors in the same patient.[23] The above case describes a patient with metachronous OO in adjacent bones of the same hand, occurring six years apart, adding it to the short list of rare reported cases with such presentation. The aim of this report is to highlight the importance of considering metachronous OO in the differential diagnosis of patients previously treated for OO and presenting with a similar clinical and radiological picture.

CONCLUSION

Furthermore, this case demonstrates that the challenging diagnosis of OO relies on the combination of typical clinical presentation, and the use of different imaging modalities, especially when dealing with smaller bones such as the phalanges. Last but not least, features of the lesion, notably its location and size, are used to guide the choice of the optimal treatment modality.

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