Phalanx osteoid osteoma. Five-case report and literature review

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ABSTRACT

Introduction: Osteoid osteomas are rarely found in the phalanges. The purpose of this study is to report 5 cases with histologic diagnosis of phalanx osteoid osteoma and highlight the most important aspects of its diagnosis and treatment. **Materials and Methods:** A descriptive, retrospective study was conducted. Evaluation included pain level using the visual analog scale (VAS), active range of motion, and QuickDASH score before surgery and at final follow-up (mean, 35.4 months). **Results:** The mean time from first consultation to diagnosis was 10.6 months (range, 5-16 months). The mean preoperative QuickDASH score was 22.72 (range, 6.8-40.9). At last follow-up (mean, 35.4 months; range, 17-63), all patients were satisfied with the outcome. The mean VAS score for pain was 0. The mean QuickDASH score was 0. **Conclusions:** Osteoid osteoma should be considered in the differential diagnosis when patients complain of subacute or chronic digital pain that improves with NSAIDs. Surgical treatment by simple curettage or en bloc resection shows satisfactory results. However, due to its low prevalence, osteoid osteoma is commonly underdiagnosed resulting in diagnosis and treatment delays.

Key words: Osteoid osteoma; phalanx; finger; chronic pain; swelling.
Level of Evidence: IV

Osteoma osteoide de falange. Reporte de cinco casos y revisión bibliográfica

RESUMEN

Introducción: El osteoma osteoide de falange es un cuadro infrecuente. El objetivo de este artículo es presentar cinco pacientes con diagnóstico histopatológico de osteoma osteoide de falange y resaltar los aspectos más relevantes del diagnóstico y el tratamiento. **Materiales y Métodos:** Estudio descriptivo, retrospectivo. Se evaluaron el dolor, según la escala analógica visual, la movilidad activa y el puntaje QuickDASH antes de la cirugía y en el control final a los 35.4 meses. **Resultados:** El tiempo medio desde la primera consulta hasta el diagnóstico fue de 10.6 meses (rango 5-16). El puntaje QuickDASH medio preoperatorio fue de 22,72 (rango 6,8-40,9). Luego del seguimiento medio de 35.4 meses (rango 17-63), todos los pacientes estaban satisfechos con el resultado. El puntaje medio de dolor fue 0. El puntaje QuickDASH medio fue 0. **Conclusiones:** El osteoma osteoide debe pensarse como diagnóstico diferencial ante un paciente que consulta por dolor digital subagudo o crónico que cede con antinflamatorios no esteroides. El tratamiento quirúrgico mediante el curetaje simple o la resección en bloque logra resultados satisfactorios. Sin embargo, como su prevalencia es baja, se suele pasar por alto y así se retrasan el diagnóstico y el tratamiento.

Palabras clave: Osteoma osteoide; falange; dedo; dolor crónico; tumefacción.
Nivel de Evidencia: IV

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INTRODUCTION

Osteoid osteoma was first described by Jaffe in 1935.[1] It consists of a benign osteoblastic tumor, histologically characterized by an osteoid-rich center with a variable degree of calcification (nidus), surrounded by a rim of reactive bone.[1,2] Osteoid osteoma accounts for 10% of all benign bone tumors.[3] It mainly occurs in children and young adults and predominantly affects males (male:female ratio, 2:1). Osteoid osteoma of the hand has an incidence of about 8% of all reported cases, metacarpal bones being the most affected. Despite their small size, osteoid osteomas may produce intense symptoms, probably due to the prostaglandin secretion by tumor cells.[5,6] Phalanx involvement is rare,[7-9] with most publications consisting of isolated cases, and given its unusual location and clinical features, it commonly results in diagnosis and treatment delays.[2,9-11]

The purpose of this study is to report 5 cases with histologic diagnosis of phalanx osteoid osteoma and highlight the most important aspects of its diagnosis and treatment.

MATERIALS AND METHODS

We conducted a retrospective cohort study. We report the cases of 5 patients with phalanx osteoid osteoma, confirmed by delayed histologic diagnosis, who underwent surgery between July 2015 and February 2018. Intraoperative localization of the lesions was performed using fluoroscopy before resection (Figure 1).

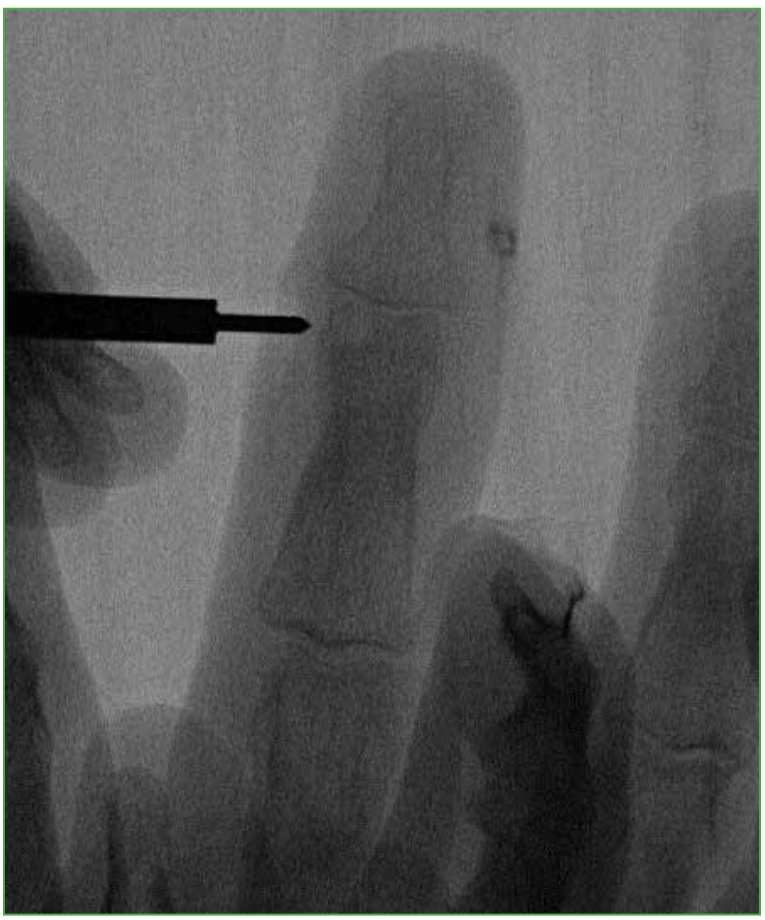

Figure 1. Case 2. Intraoperative localization of the lesion using fluoroscopy.

The surgical procedure was performed through a lateral approach in 3 patients (cases 1, 2, and 5) and through a volar Brunner-type approach in 2 patients (cases 3 and 4). The case 4 patient first surgery was performed through a dorsal approach and involved the simple curettage of the lesion. Two months later, with persistent symptoms, no clear histologic diagnosis, and nidus presence confirmation via computerized tomography (CT), the patient underwent another surgery involving the en bloc resection of the lesion through a volar Brunner-type approach. One patient underwent simple curettage of the lesion (case 1); another patient, curettage and filling with distal radius autograft (case 2); two patients, en bloc resection and prosthetic bone replacement (cases 3 and 5); and 1 patient, en bloc resection with no grafting (case 4). Patients were immobilized for 3 weeks with finger splints and then began their physical therapy program. The mean follow-up was 35.4 months (range, 17-63). Collected data included times from first consultation to diagnosis, signs and symptoms at presentation, and pain levels using the VAS score, with 0 indicating no pain and 10 indicating the worst pain imaginable. We also used the score from QuickDASH questionnaires, with 0 being the best score, and 100 the worst. Patients were contacted for a final clinical and radiographic evaluation in September 2019. Evaluation included goniometric assessment of the active range of motion, and pain levels according to the VAS score, and patients were required to complete the QuickDASH questionnaire.

RESULTS

Four of the 5 patients were male. Three cases involved the proximal phalanges with lesion in intracortical locations; 1 case, the middle phalanx in cancellous location; and 1 case, the distal phalanx in cancellous location (Figure 2).

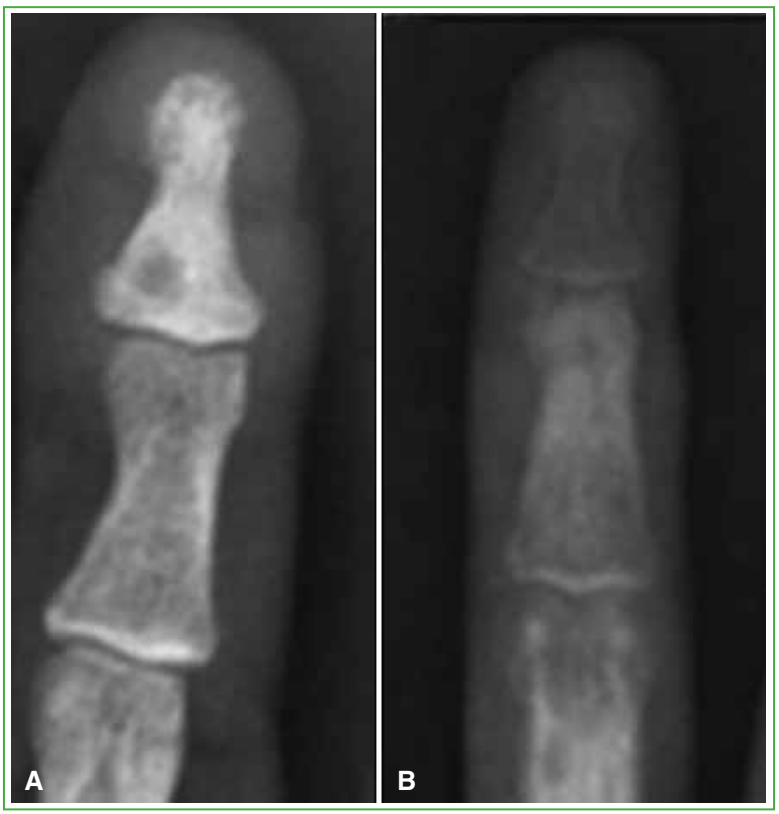

Figure 2. A. Case 1. Osteoid osteoma in cancellous location. Right little finger radiograph. Osteolytic image at the base level of the proximal phalanx. No evidence of sclerotic rim or central calcification. **A.** Case 2. Osteolytic image evidencing small central calcification at the middle phalanx radial condyle.

One patient underwent simple curettage of the lesion (Figure 3); 1 patient, curettage and bone graft filling (Figure 4); 2 patients, en bloc resection and bone grafting; and 1 patient, en bloc resection with no grafting.

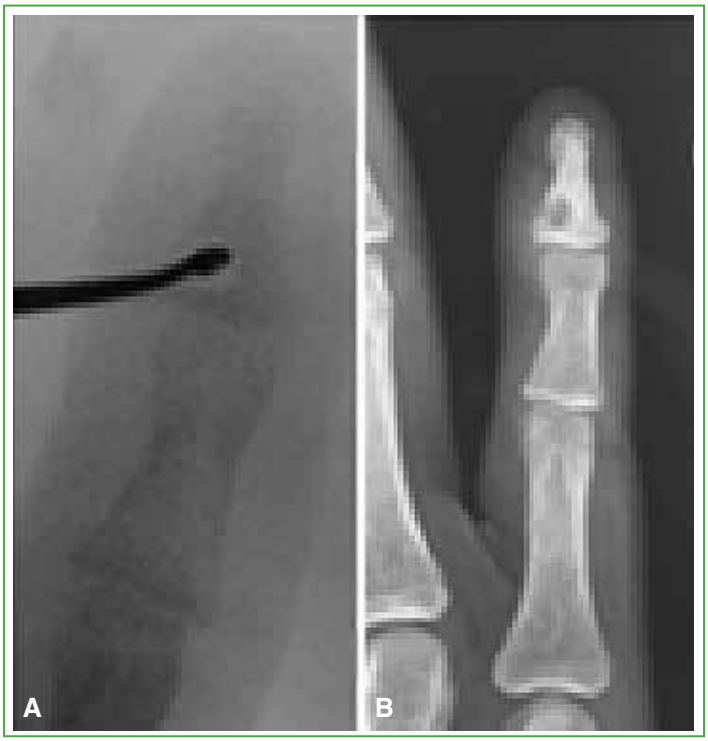

Figure 3. A. Case 1. Lesion curettage through a lateral approach.
B. Postoperative X-ray.

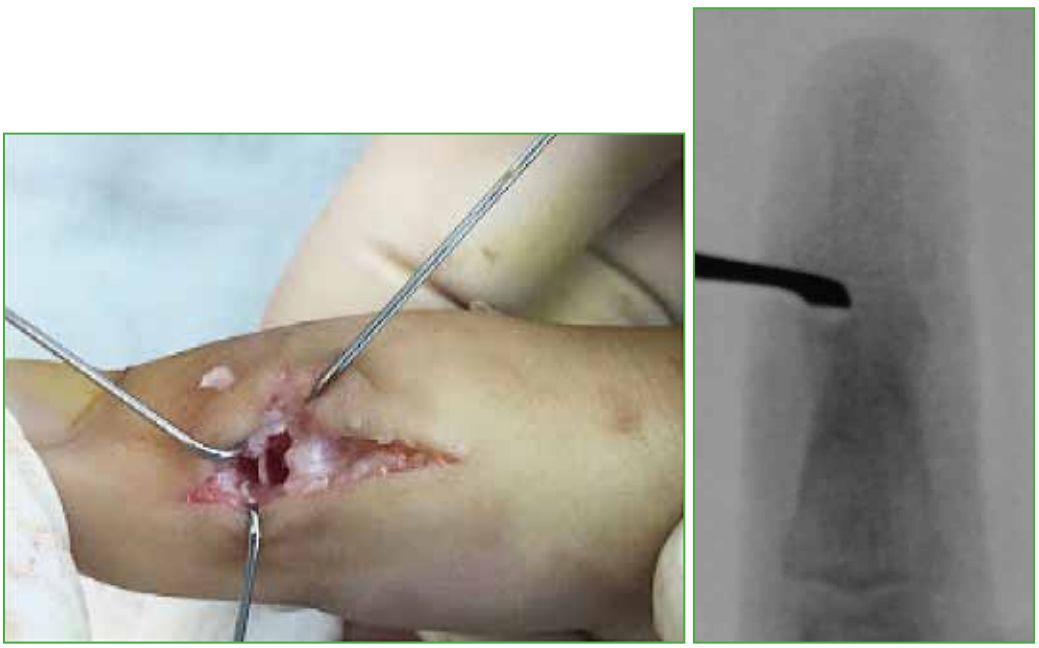

Figure 4. Case 2. Lesion curettage through a lateral approach. Articular cartilage was preserved. The defect was filled with cancellous graft from the distal radius.

One patient (case 4) required a second surgery due to incomplete resection. The mean size of lesions was 5.1cm in maximum diameter on CT (range, 2.9-8.14 mm) (Table).

Table. Cases, demographics, and outcomes

Case	Age	Sex	Diagnosis delay (months)	Symptomatology	Location	Size (maximum diameter)	Treatment	Preoperative pain (VAS)	Postoperative pain (VAS)	Preoperative Quick-DASH score	Postoperative Quick-DASH score	Complications
1	14	M	16	Night pain, tenderness, increased diameter, clubbing	Distal phalanx base, cancellous	6.1mm	Lateral approach Simple curettage	6/10	0	25	0	No
2	24	M	14	Continuous pain, tenderness, excessive sweating, distal interphalangeal arthritis	Middle phalanx condyle cancellous subchondral	3.3mm	Lateral approach Curettage and filling with distal radius autograft	8/10	0	22.7	0	No
3	16	F	12	Continuous pain, increased diameter, flexor tenosynovitis	Proximal phalanx, volar intracortical	2.9mm	Volar approach En bloc resection and prosthetic bone replacement	6/10	0	18.2	0	No
4	13	M	5	Pain, increased diameter, limited flexion	Proximal phalanx, volar intracortical	8.14mm	Volar approach En bloc resection (no grafting)	4/10	0	40.9	0	Patient required a second surgery due to incomplete resection
5	27	M	6	Pain, tenderness, increased diameter	Proximal phalanx, medial intracortical	N/A	Lateral approach Curettage and prosthetic bone filling	3	0	6.8	0	No

F = female; M = male; VAS = visual analog scale.

The average age at presentation was 18.8 years (range, 14-27 years). The mean time from first consultation to diagnosis was 10.6 months (range, 5-16 months). All patients presented with pain, with a 5.4 mean-VAS score (range, 3-8). They all had swollen fingers (Figure 5). The mean preoperative QuickDASH score was 22.72 (range, 6.8-40.9). At last follow-up (mean, 35.4 months; range, 17-63), all patients were satisfied with the final outcome. Both VAS and QuickDASH questionnaire mean scores were 0. All patients achieved full active range of motion (Figures 6 and 7). One patient (case 4) required a second surgery due to incomplete resection (Table).

Figure 5. Case 4. Notice the swelling of the middle finger. Patient with middle phalanx osteoid osteoma, in intracortical location.

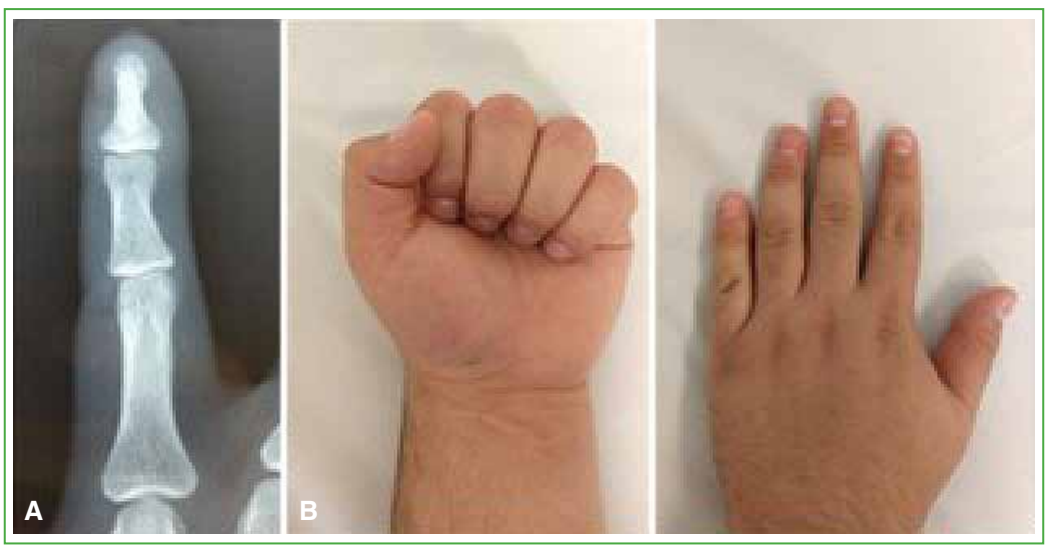

Figure 6. Case 1. **A.** Complete bone remodeling. **B.** Active range of motion at 17-month follow-up.

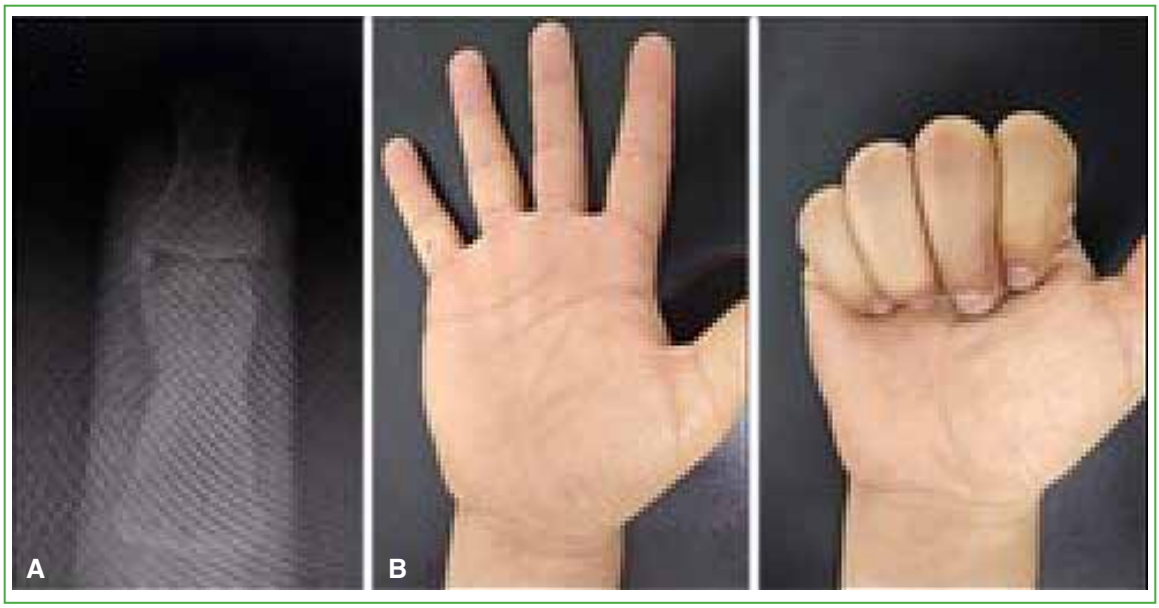

Figure 7. Case-2 patient, 2 months after surgery. **A.** Partial graft resorption. **B.** Final mobility.

DISCUSSION

Phalanx osteoid osteoma is a rare condition.[7,8] The most common clinical manifestation is progressive pain that worsens at night and may be continuous.[8,11]Pain relief is usually achieved with salicylate or other NSAIDs therapy.[2,4-6,8,11]

The etiology of the pain associated with osteoid osteoma may be multifactorial. Prostaglandins found in the nidus are at levels 100 to 1000 times that of normal tissue, resulting in vasodilation and increased capillary permeability in the tissues surrounding the lesion.[11] Nakatsuchi *et al*. found a bundle of nerve fibers in and around the lesion adjacent to blood vessels or as independent fibers, suggesting that the pain may be mediated by the nervous system via this nerve fibers. Pain may also reflect changes in vessel pressure or irritation of the nerve fibers located in and around the nidus.[13]The etiology of the soft-tissue hypertrophy remains unclear, the accelerated metabolism of the tumor being a possible cause.[14,15]

Both premature growth plate closure and overgrowth have been found in cases involving the distal phalanx.[11,15-17] Phalanx osteoid osteomas involve such atypical symptoms that include patients with painless presentations. Synovitis and reduced range of motion are common in patients with juxta-articular locations of the osteoma.[7,11,18]

Osteoid osteomas may be classified according to location into cortical, cancellous, and subperiosteal.[19] The typical cortical radiograph image comprises a small radiolucent area of less than 1cm in diameter, subcortically placed, and associated with dense adjacent bone sclerosis and periosteal reaction. A nidus is commonly detected centrally within the radiolucency. The cancellous variety also presents with a radiolucent area, but with little or no adjacent sclerosis. The least common variety is the subperiosteal osteoid osteoma, which radiographic image shows an area of 0.5-1cm in diameter, immediately adjacent to bone. These cases may involve cortical evidence of pressure defect or irregular bone resorption.[19]

Although the osteoid osteoma classification based on location applies to phalangeal locations, it should be noted that the size of the aforementioned lesions corresponds to other locations and that phalangeal lesions are significantly smaller. Our series lesions ranged from 2.9mm to 8.1mm in maximum diameter on CT imaging.

The cortical variety of osteoid osteoma is the most common overall, but the cancellous and subperiosteal varieties are likely to be more common in the small bones of the hand and foot.[9,18]

Radiographs of the phalanges in osteoid osteoma patients are commonly atypical, may only evidence osteolysis,[11,15,16,20] or even be negative.[14] In such cases, CT, magnetic resonance imaging (MRI), and in some cases bone scintigraphy[9,18] play a key role in establishing the presumptive diagnosis.[5,6]

CT scanning is useful when the nidus is not detected on plain radiographs and has high specificity to detect and differentiate the lesion (Figures 8 y 9).[21] We suggest performing a high-resolution helical CT scan of the phalanges, with sections as thin as possible (commonly 1mm) because osteoid osteomas may not be evidenced due to their small size.

Typical CT findings include:

- a low-density round or oval area, evidencing the nidus,
- a high-density area within the nidus, of varying size according to the extent of calcification,
- A rim of peripheral reactive sclerosis or periosteal reaction (rare in phalanges).

MRI is a more sensitive imaging study than CT in detecting phalanx osteoid osteomas.[21,22] Besides, MRI imaging may also provide additional details regarding adjacent soft-tissue inflammatory reactions. The closest the lesion is to the bone marrow, the more useful MRI becomes in detecting the nidus in comparison with CT. MRI shows different signals for bone marrow, nidus, reactive osteosclerosis, and soft tissues. The nidus appears as an area of hypointensity or isointensity on T1-weighted images, and variable intensity on T2-weighted images, according to the degree of mineralization. Peripheral reactive sclerosis evidences as low signal intensity on T1- and T2-weighted images. Marrow edema and surrounding soft-tissue edema are commonly found (Figure 10).[23,24] A partially mineralized nidus may be evidenced by a target-shaped image. The administration of gadolinium enhances the imaging of the nidus as a result of the nidus hypervascularity.[22]

In cases where reactive peripheral bone is not evidenced, the nidus may prove hard to detect. Bone scintigraphy becomes especially useful in these cases as it has close to a 100% sensitivity in detecting these lesions, showing increased uptake in its three phases (Figure 11).[25]

Osteoid osteomas have are well vascularized and more osteoblasts than normal tissue, which results in the tumor great absorption of technetium 99 that binds to hydroxyapatite crystals. The double density sign (also known as the hotter spot within hot area sign) is diagnostic of osteoid osteoma and is especially useful to differentiate the condition from osteomyelitis.[26]

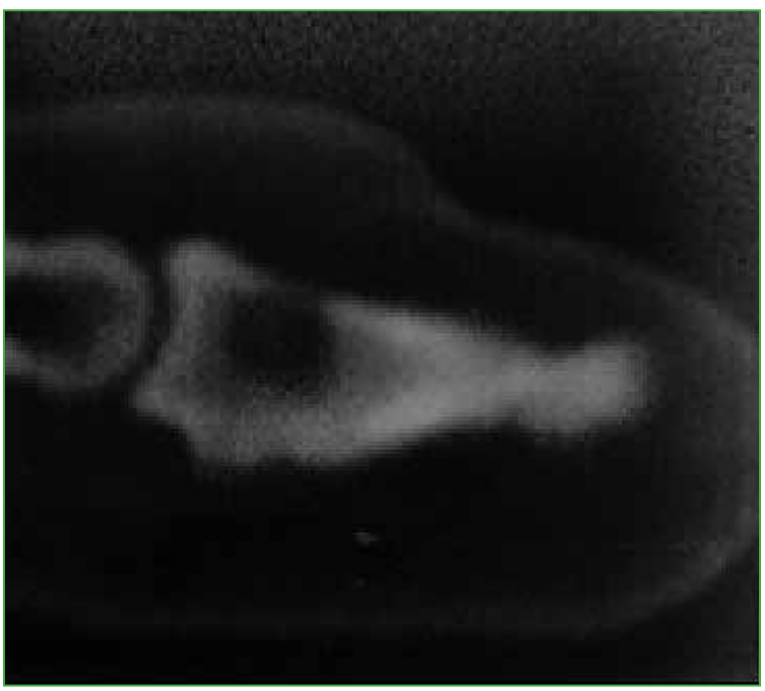

Figure 8. Case 1. CT sagittal section. Osteolytic image evidencing dorsal cortical involvement. Notice the lack of central calcification and reactive sclerosis.

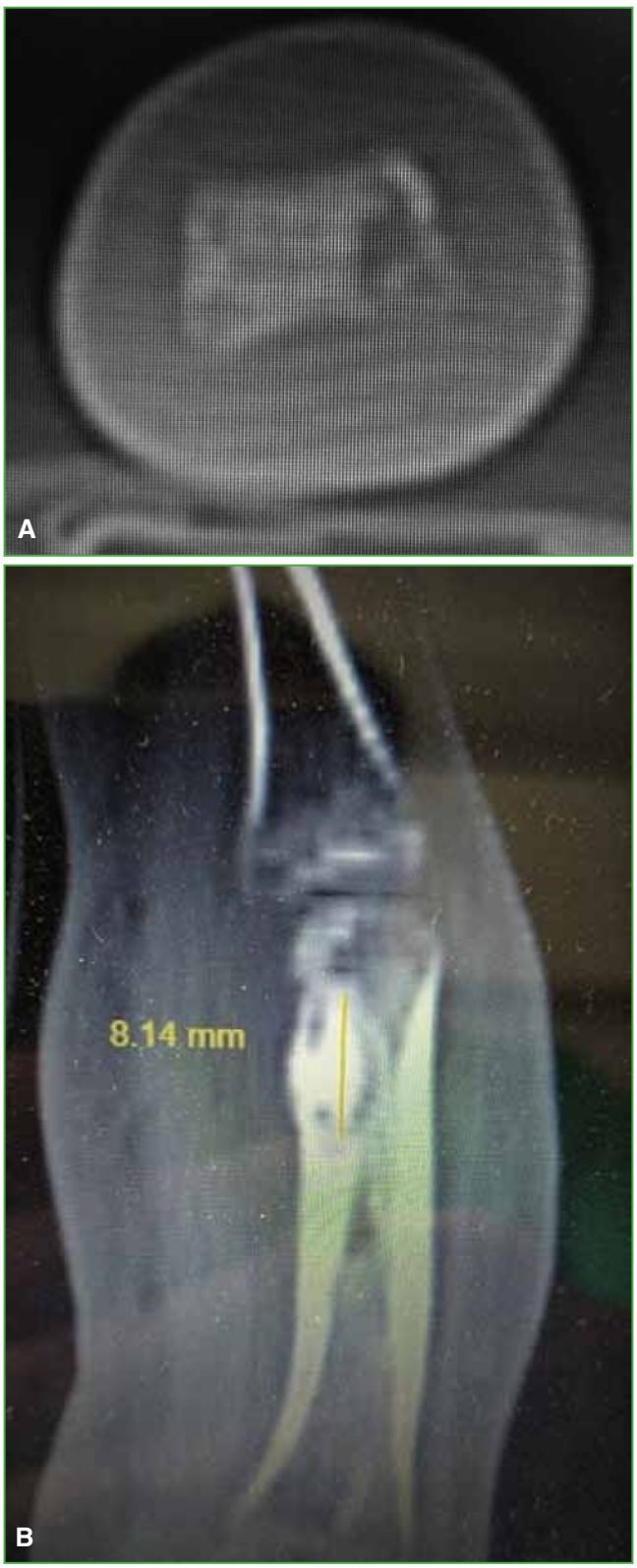

Figure 9. **A.** Case 2. Computed tomography. Notice the osteolytic image with central calcification associated with the nidus of the middle phalanx radial condyle. **B.** Case 4. Measuring of the lesion maximum diameter.

Based on our literature review and our limited experience, in patients with suspected phalanx osteoid osteoma, we suggest first ordering plain radiographs and later adding CT scans. In most cases, we consider that the imaging findings will suffice to administer treatment. In cases with no mineralized nidus (as this series case 1), radiographs and CT may prove inadequate to confirm a tentative diagnosis, and MRI or scintigraphy studies should be considered.

An important factor to consider when using MRI is that it commonly "magnifies" the lesion, showing bone edema involving the entire phalanx and surrounding soft tissues, which may mislead the doctor into suspecting infection or cancer. Both case-1 patient and case-4 patients were referred to other health-care centers with suspected cancer based on the osteolytic images of radiographs and the MRI findings that included no evidence of nidus and significant bone and soft-tissue edema (Figure 10).

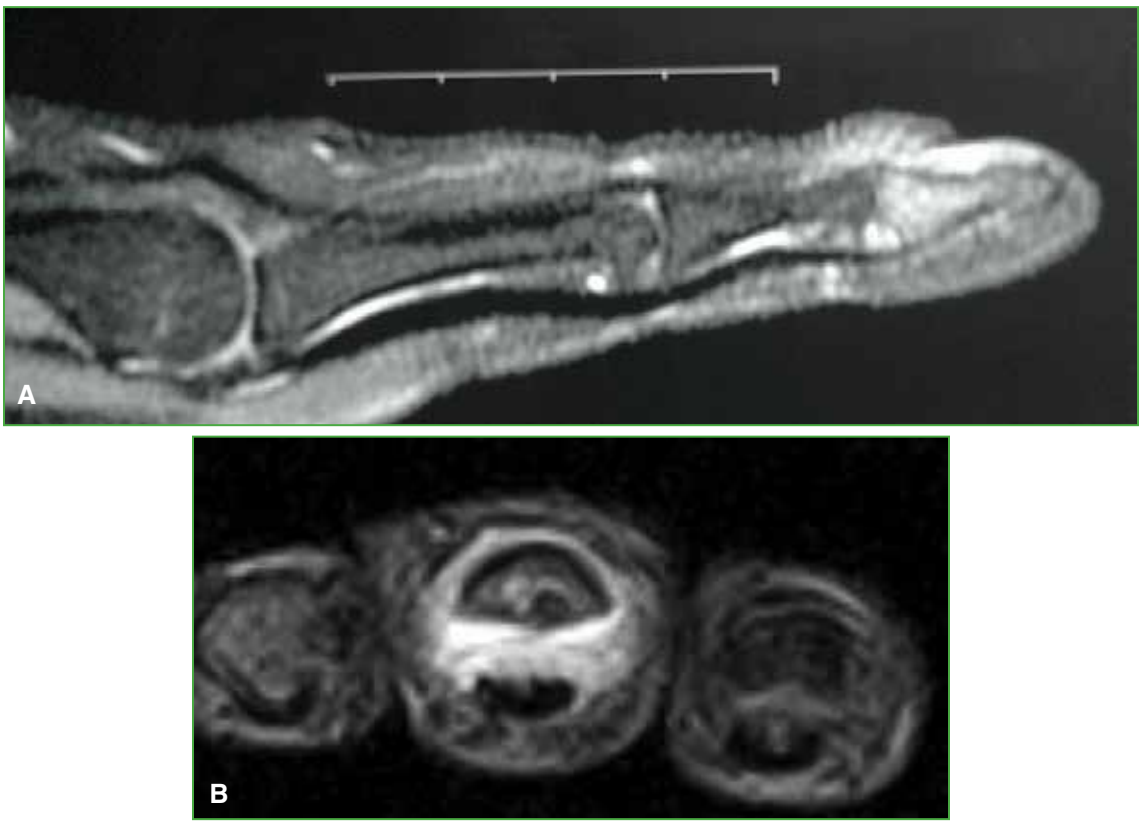

Figure 10. A. Case 1. MRI sagittal section. Hyperintense image (T2-weighted) of the distal phalanx and soft-tissue edema. **B.** Case 4. MRI-axial section. Notice the significant soft-tissue edema. Nidus may be evidenced in the volar cortical.

Doctors examining patients with subacute or chronic finger pain and no trauma history should consider the following manifestations as indicative of osteoid osteoma:

- Night or continuous pain that improves with NSAIDs,[6,8,11,27,28]
- tenderness,[18]
- local swelling,[8,11,15,16,18]
- enlargement of the nail plate (distal phalanx involvement)[8,11,15-18]
- synovitis (epiphyseal involvement),[7,11]
- excessive sweating.[8,15,18]

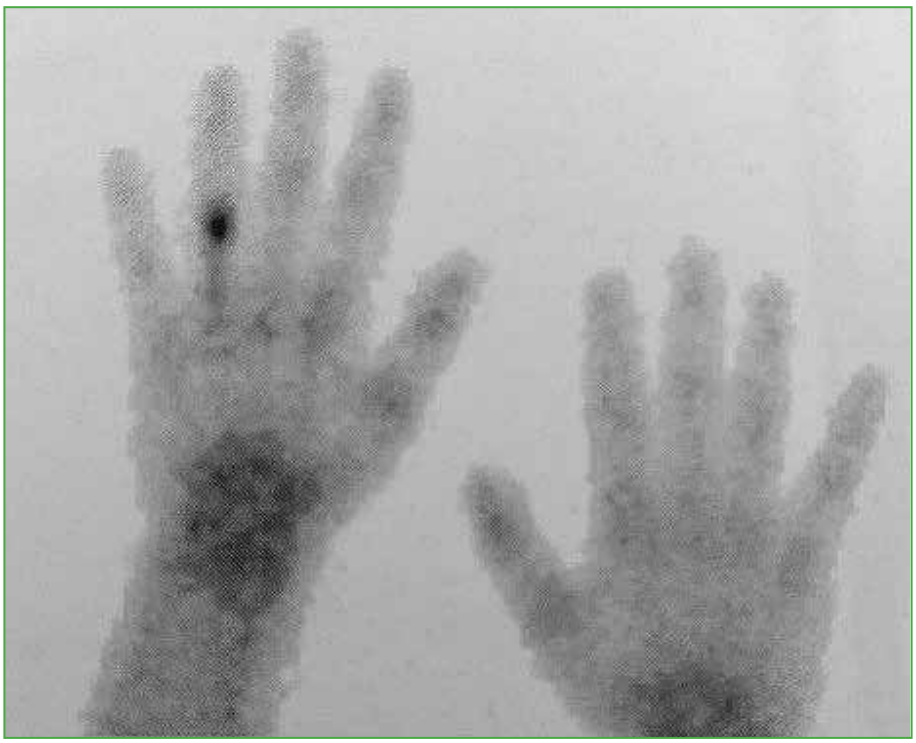

Figure 11. Case 4. Three-phase bone scintigraphy showing increased uptake in the proximal phalanx.

Differential diagnosis is based on clinical and imaging findings and includes the following conditions:
- glomus tumor,[16,18,21]
- osteomyelitis,[7,9,11,14,16,18]
- tenosynovitis,[7,11]
- epidermoid inclusion cyst,[9,14,16,18]
- reactive periostitis,[9]
- arteriovenous fistula,[14,16]
- benign or malignant tumor.[7,14,23,29]

Consistent with other reported cases,[3-5,9,11,18,20,22,23,30] these study patients had previously consulted for their conditions several times, were misdiagnosed on many occasions, and received unnecessary treatments that often resulted in diagnosis delays.

Standard osteoid osteoma treatment involves en bloc resection or curettage of the lesion, with or without bone graft. Over the past years, CT-guided percutaneous treatment techniques have become a popular treatment choice. Both radiofrequency ablation and laser photocoagulation are currently considered first-line treatments in reference centers, with success rates over 90%.[31,32] However, there is still a small number of reports on the use of these techniques for phalanx osteoid osteomas and they are limited to isolated cases.[33] Recent reports[34,35] warned that they may produce chronic pain and persistent swelling when performed on phalanx lesions. Pain may result from thermal damage to the digital neurovascular bundle, considering its close proximity to the treated area.

Lesion resection constitutes the definitive treatment. Recurrence is uncommon, and recurrence cases may be associated with incomplete resections. Curettage treatment may prove sufficient in lesions localized in cancellous bone, but have a higher recurrence rate in cases with cortical involvement, as it was the case of our case-4 patient. Therefore, we suggest en bloc resection of the affected cortices. An alternative to en bloc resection is simple resection followed by extension of the margins using a high-speed power burr to prevent incomplete resections, which constitute the most common cause of treatment failure.

Patients with distal phalanx involvement experience significant pain and swelling improvement after the osteoid osteoma resection, but the nail bed and plate remain enlarged. (Figure 12).[16,36]

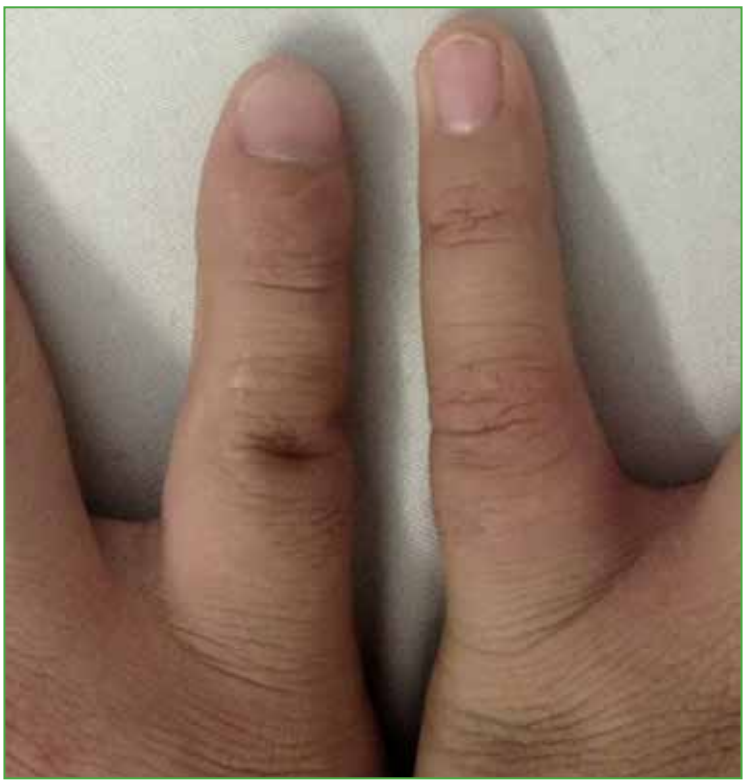

Figure 12. Case 1. Notice the increased diameter of the finger and the nail clubbing.

CONCLUSIONS

Phalanx osteoid osteoma is a rare presentation that involves different clinical and radiological manifestations from osteoid osteomas in other locations. Osteoid osteoma should be considered in the differential diagnosis when patients complain of subacute or chronic digital pain that improves with NSAIDs. Patients may commonly present with increased diameter of the finger or enlarged nail plate, tenderness, excessive sweating, and synovitis, on which a tentative diagnosis may be built. Plain radiography and CT scanning are the first-line diagnostic studies. Surgical treatment by simple curettage or en bloc resection shows satisfactory clinical and radiological outcomes. However, due to its low prevalence, osteoid osteoma is commonly underdiagnosed, resulting in diagnosis and treatment delays.

Conflict of interests: Authors claim they do not have any conflict of interest.

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