

UNCOMMON PRESENTATION OF OSTEOID OSTEOMA IN THE ELBOW: CASE REPORT

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Abstract: Osteoid osteoma is the third most common benign bone neoplasm. The femur and tibia are the most affected sites. In this case, unusually, we report its occurrence in the elbow with intra-articular presentation, which many times, as in this patient, can delay the definitive diagnosis. Open surgery, available at the case service, was chosen as the treatment and used for complete removal of the tumor, which had histopathological analysis confirmed for the diagnosis of osteoid osteoma.

Keywords: Bone Neoplasms; Elbow; osteoid osteoma.

INTRODUCTION

Osteoid osteoma (OO) is a non-progressive osteoblastic benign lesion, first mentioned by Heine¹ (1927), consisting of hypervascularized immature osteoid tissue surrounded by reactive sclerotic bone. Henry Lewis Jaffe², in 1935, classified this bone lesion as a clinical pathological entity, distinguishing it from other bone neoplasms.

OOs are the third most common benign bone neoplasm, representing about 14% of cases.³ It is more prevalent in males and preferentially affects the age group from five to 20 years.⁴ Typically, they affect the shafts of the long bones of the limbs lower limbs, more commonly in the medial region of the proximal femur.¹ OO in the upper limb is less frequent, with only 3% of cases occurring in the elbow.⁴ In epiphyses, with intra-articular presentation, it occurs less frequently⁴, with an incidence of up to 13% of cases, with the hip being the most affected region.⁵

The most characteristic clinical manifestation of OO is intermittent pain, mainly at night, which improves after the administration of non-steroidal anti-inflammatory drugs.⁴ Furthermore, it may present with erythema and local edema, with limitation of movement. Muscle atrophies

are often present and can lead to mistaken diagnoses of neurological pathologies.³ The typical radiographic image of OO is represented by the radiolucent nidus, normally up to 1.5 cm in diameter, surrounded by a variable amount of sclerotic bone or cortical thickening.⁶

The intra-articular presentation is less frequent and its diagnosis is usually time consuming and difficult. This occurs because the originating symptoms can mimic inflammatory or infectious arthropathies, coursing with joint effusion, decreased range of motion and synovitis. The diagnostic difficulty is also greater due to the radiographic image, since the radiolucent nidus surrounded by sclerotic bone, mentioned earlier, is less exuberant in intra-articular cases, which can, in these cases, lead to a delay in the definitive diagnosis of OO.⁵

The purpose of this report is to pay attention to cases of OO with an infrequent location, based on a case with intra-articular involvement of the elbow, which was resolved with open surgery.

CASE REPORT

The entire procedure was carried out in accordance with the ethical standards determined by the Research Ethics Committee for research on human beings, and by the 1964 Declaration of Helsinki. Informed consent was obtained from the patient by signing a specific term.

A 25-year-old male, denied a history of alcoholism and smoking, presented with pain in the posterior region of the left elbow, which started two years ago, worsening at night, and without a history of trauma. He denied morning stiffness or polyarthralgia. With a clinical diagnosis of synovitis, he was previously submitted to pharmacological treatment with NSAIDs, without improvement. On physical examination, he

had neurovascular preservation, slight edema in the posterior region of the elbow, without limitation of range of motion. A simple radiographic examination of the elbow showed no alterations (Figure 1). Through Computed Tomography (CT) and Magnetic Resonance Imaging (MRI), the following alterations in the distal humerus can be observed: 1.0 cm nodulation, location of the nidus (calcified central portion of 0.6 cm), bone sclerosis, without cortical lesion and subperiosteal location, and small joint effusion. Laboratory tests were performed, which included blood count and inflammatory markers within normal limits, which helped to rule out inflammatory or infectious causes. Imaging tests were important to rule out traumatic causes and the findings led to the diagnostic hypothesis of OO (Figure 2).



Figure 1: Radiographs in anteroposterior (“a”) and lateral (“b”) views of the elbow, without visualization of the nidus.

Source: Authors compilation

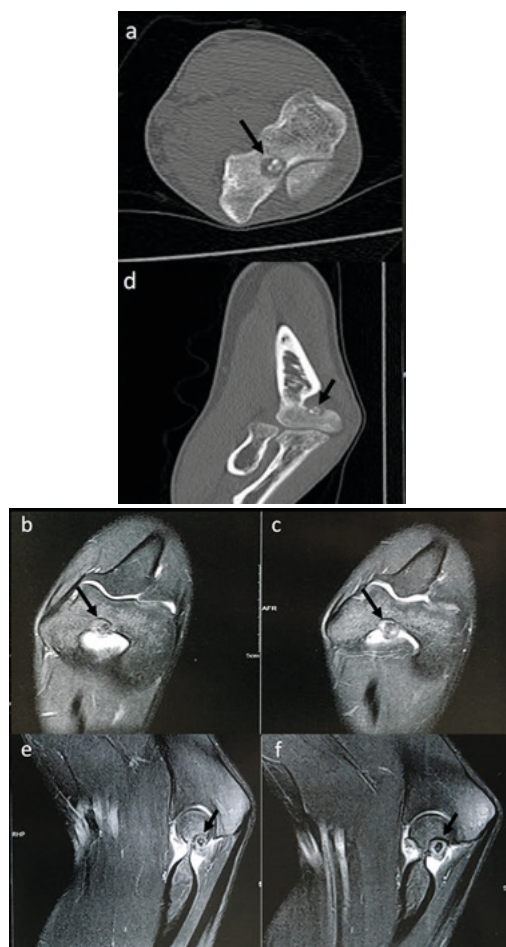


Figure 2: Tomographic images in the axial (“2.a”) and coronal (“2.b”) planes of the same patient demonstrate the lesion (arrows). Magnetic resonance coronal section (2.c and d) and sagittal section (“2.e” and “f”); where the subperiosteal osteoid osteoma in the olecranon fossa is visualized.

Source: Authors compilation

We chose to perform tumor resection by open surgery due to the unavailability of materials to perform other techniques in the service where the procedure was performed. In surgical management, tumor resection with intralesional margin was performed through posterior access to the left elbow via transtricipital route (Figure 3). The anatomopathological analysis confirmed the diagnosis of OO (Figure 4). After the procedure, the patient presented complete resolution of the symptoms, without their

recurrence for the period of one year in which he was followed up by the Traumatology service.

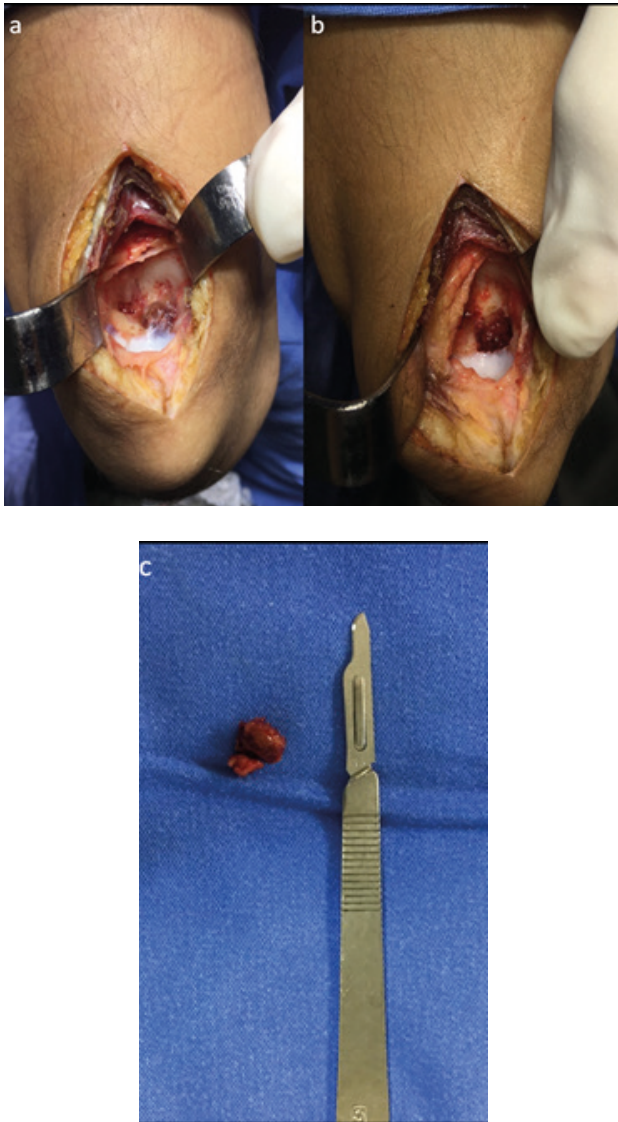


Figure 3: (“3.a”) Identification of the bone lesion in the intraoperative period through the open route in the posterior region of the elbow; (“3.b”) – Marginal resection of the nidus in the olecranon fossa; (“3.c”) - Surgical specimen - resected osteoid osteoma.

Source: Authors compilation

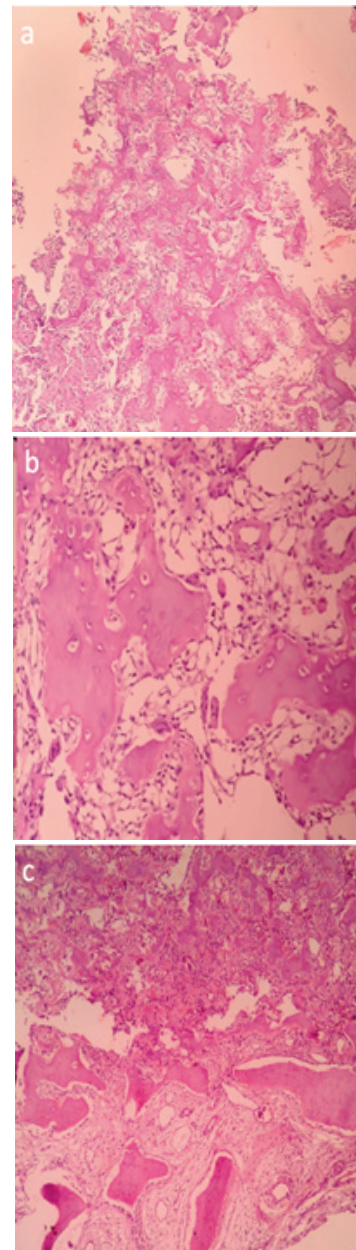


Figure 4: (“4.a”) Microphotography of the osteoid osteoma with newly formed bone in the middle of a fibrovascularized stroma with occasional osteoclasts (HE). (“4.b”) Image enlargement “a” - young bone trabeculae and angiectasia. (“4.c”) Bordering area of the resection margin with native trabecular bone.

Source: Authors compilation

DISCUSSION

As mentioned above, OOs preferentially affect young male patients, and their most typical location is in the diaphysis of the long bones of the lower limbs, mainly in the medial region of the proximal femur. However, in the case reported above, despite the epidemiology being consistent with the literature, the intra-articular location of the tumor, more specifically in the distal segment of the humerus, is unusual, causing a diagnostic challenge for cases like this.

Regarding the symptoms and atypically, as is the case of the reported intra-articular presentation, pain relief with the use of NSAIDs is often minimal or absent.⁷ In addition, the characteristic pain may also be absent, diverging from of literature. Such fewer specific manifestations can mimic inflammatory arthropathies, being significant impediments to a more agile diagnosis.

In a retrospective study, in two Brazilian hospitals and among all cases confirmed histologically from bone biopsy, a prevalence of 2.8% of OO located in the distal segment of the humerus was found, which reflects the relative infrequency of this presentation. As a result, the patients in the study, who had chronic pain and movement limitation, with a diagnosis of inflammatory arthropathies, underwent non-surgical therapies, with no improvement in their condition. The average found was 21 months between the onset of symptoms and the definitive diagnosis.⁸ The patient in the reported case had a similar clinical picture, with a previous diagnosis of synovitis only, with pain for a period of 24 months until the definitive diagnosis. The lack of knowledge about the details involved in the diagnosis of a not so common presentation can lead to a delay of almost two years in the resolution of cases.

Imaging exams represent an important diagnostic tool in intra-articular OO and, the

delay in requesting them can lead to mistaken clinical diagnoses and trigger prolonged arthritic symptoms, as in the reported case. The typical radiographic image is the central radiolucent nidus, also seen in the patient. They may or may not be surrounded by reactive sclerotic bone or cortical thickening, however, this usual presentation is present in only 47% of cases.⁶ In intra-articular lesions, the presence of reactive bone sclerosis seen in the patient may be absent or minimal, making the diagnosis even more difficult. In addition, there may be regional periarticular osteopenia in early stages.

This benign neoplasm presents with a rare familial character and its etiology is still unknown. There are few studies in the literature that attribute the role of genetics in the development of OO. The main alteration found, based on the cytogenetic analysis of two patients with the histological diagnosis of OO, was deletions in chromosome 22q, suggesting its important role in the proliferation processes of this neoplasm. Changes in this same chromosome, whether numeric or structural, have already been described in patients with schwannomas, meningiomas, osteosarcomas, fibrosarcoma and malignant fibrous histiocytoma.⁹

OO has as differential diagnoses lesions of an inflammatory and infectious nature, or neoplasms, as well as chondroblastoma and osteoblastoma. high activity of the nidus and low activity of the surrounding reactive sclerotic zone although the final diagnosis is determined by histopathological examination.¹²

The curative intervention for OO is surgical resection. Currently, the surgical treatment of choice is minimally invasive techniques, such as arthroscopic resection or CT-guided radiofrequency thermoablation.¹³ These are efficient methods, with low failure rates and no recurrences. In addition, they have

the advantage of causing less morbidity and a faster return of patients to their activities. Traditional open resection surgery is reserved for cases in which the surgical planning requires it or when resources for minimally invasive procedures are not available, as in the case reported above, where open surgery was chosen due to limited resources and materials available at the Traumatology service in question.

CONCLUSION

Intra-articular OO is an infrequent source of elbow symptoms. The clinical and radiological presentation is not typical, which makes the diagnosis complex. It is important to take into consideration, the suspicion of OO in young people with complaints of pain, reduced mobility and signs of monoarthritis in the elbow, with no apparent cause or inadequate response to conservative management and unrelated to other pathologies. Plain radiography is the resource used in the initial evaluation, but tomography is the gold standard for recognizing the nidus. Prior diagnostic confirmation provides the best surgical approach, reducing symptoms.

REFERENCES

1. HENRY, L. JAFFE, M. D. Osteoid osteoma. **A benign osteoblastic tumor composed of osteoid and atypical bone.** Arch Surg. 1935; 31 (5), p. 709-728.
2. MACLELLAN, D.I. WILSON, F.C.: **Osteoid osteoma of the spine. A review of the literature of the literature and report of six new cases.** J Bone Joint Surg [Am]. 1967; 49, p. 111-121.
3. BOSCAINOS, P.J. COUSINS, G.R. KULSHRESHTHA, R. OLIVER, T.B. PAPAGELOPOULOS, P. J. **Osteoid Osteoma.** Orthopedics. 2013; 36(10), p. 792-800.
4. Bergstrand, H. Über eine eigenartige, wahrscheinlich bisher nicht beschriebene osteoblastische Krankheit in den langen Knochen der Hand und des Fusses. Acta Radiol. Novembro, 1930; 11 (6), p. 596-613.
5. KRANSDORF, M.J. STULL, M.A. GILKEY, F.W. MOSER JR, R.P. **Osteoid osteoma.** Radiographics. 1991; 11 (4), p. 671-696.
6. GREENSPAN, A. **Benign bone-forming lesions: osteoma, osteoid osteoma, and osteoblastoma.** Skeletal Radiol. 1993; 22 (7), p. 485-500.
7. CIFTDEMIR, M. TUNCEL, S. A. USTA, U. **Atypical osteoid osteomas.** European Journal of Orthopaedic Surgery & Traumatology, 2013. 25 (1), p. 17-27.
8. COTTA, A. N. et al. **Dificuldades diagnósticas no osteoma osteoide do cotovelo: estudo clínico, radiológico e histopatológico.** Colégio Brasileiro de Radiologia. Publicação científica oficial. Volume 45, Nº 1 -Janeiro/Fevereiro de 2012.
9. BARUFFI, M. R. VOLPON, J. B. BARBIERI, N. J. et al. **Osteoid osteomas with chromosome alterations involving 22q.** Cancer Genet Cytogenet. 2001; 124, p.127-131.
10. RESNICK, D. KYRIAKOS, M. GREENWAY, G.D. **Tumors and tumor-like lesions of bone: imaging and pathology of specific lesions.** Bone and joint imaging. Philadelphia, PA: Elsevier/Saunders; 2005. p. 1120-1126.
11. SZENDROI, M. KOLLO, K. ANTAL, I. et al. **Intraarticular osteoid osteoma: clinical features, imaging results, and comparison with extraarticular localization.** J Rheumatol. 2004; 31. p. 957-964.
12. ALLEN, S. D. SAIFUDDIN, A. **Imaging of intra-articular osteoid osteoma.** Clin Radiol. 2003; 58. p. 845-852.
13. KNEZEVIC, I. BOJANIC, I. **Comparison of arthroscopy versus percutaneous radiofrequency thermal ablation for the management of intra- and juxta-articular elbow osteoid osteoma: case series and a literature review.** Distúrbio musculoesquelético BMC. Mar 2022. 25;23(1):287.