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LETTER

Osteoid osteoma mimicking refractory juvenile arthritis in a pediatric patient

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A 9-year-old patient, complaining of pain and swelling of his left elbow for 3 months, was referred to the paediatric rheumatology consultation. On clinical examination, there was a local swelling with limited joint mobility of the left elbow. The other joints were normal. There were no associated skin lesions, weight loss, or fever. His personal and family medical history were not relevant. A blood test for inflammatory markers, complete auto-immune serology, and Borrelia serology were all normal.

On the basis of these elements, we suspected juvenile idiopathic arthritis (JIA) and he was treated with local corticosteroid infiltration and oral non-steroidal anti-inflammatory drugs (NSAIDs). The patient was seen 1 month later, with poor benefit from the infiltration. A new ultrasound was performed, which revealed an intra-articular effusion without a positive Doppler signal, indicating inactivity of the rheumatological disease. Physiotherapy was then proposed, as well as continuation of NSAIDs.

However, given the unusual evolution and persistence of pain, swelling, and increasingly disabling extension defect, a joint lavage was performed. Bacterial cultures were sterile and a slight chronic lymphoplasmocytic synovitis was shown by anatomopathological analysis. These analyses remained compatible with our first diagnosis of JIA and a second line treatment with methotrexate was started.

Because of the absence of improvement after 12 weeks, a new X-ray was carried out, showing the presence of periosteal appositions on a distal humeral metaphysis, which is unusual in JIA (Figure 1). Therefore, magnetic resonance imaging (MRI) was performed. The results revealed a small, heterogeneous, well-defined, 5 mm lacunar lesion located in the lateral part of the left humeral metaphysis, and the radiologists concluded that it was chronic osteomyelitis (Figure 2).

Histology of the resected lesion by open surgery revealed an osteoid osteoma. After the surgery, the pain quickly resolved and the persistent joint limitation was treated by intensive physiotherapy to recover normal joint mobility, with good evolution. Osteoid osteoma is a benign bone tumour, affecting patients between 10 and 20 years of age, the majority of whom are male (1–4). Different localizations are possible and were described by Kayser et al in 1998, the most frequent localization being intracortical (about 75% of lesions) (4). These so-called ‘classical’ lesions generally

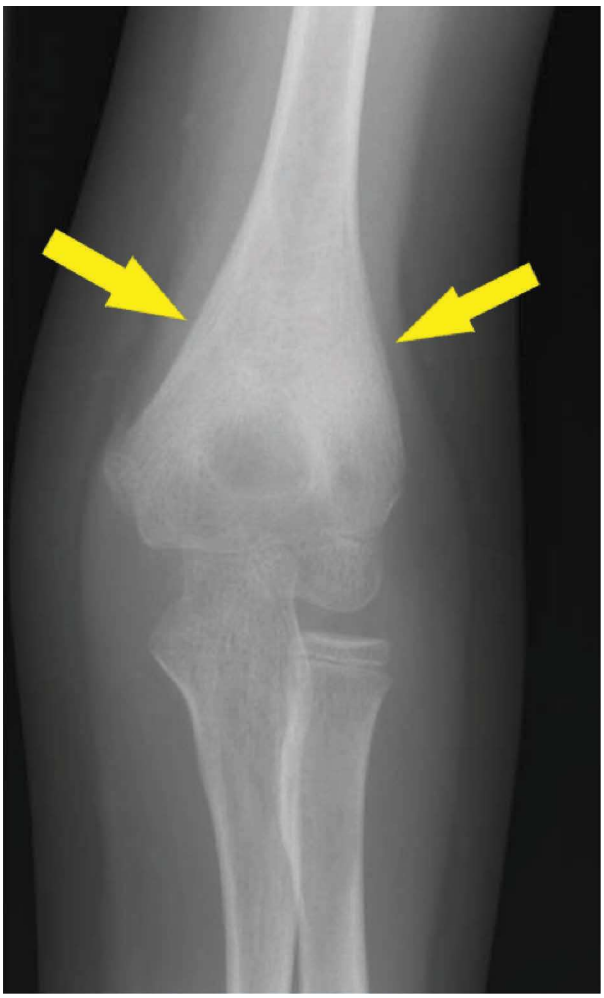

Figure 1. X-ray of the right elbow. The arrows show periosteal apposition on a distal humeral metaphysis.

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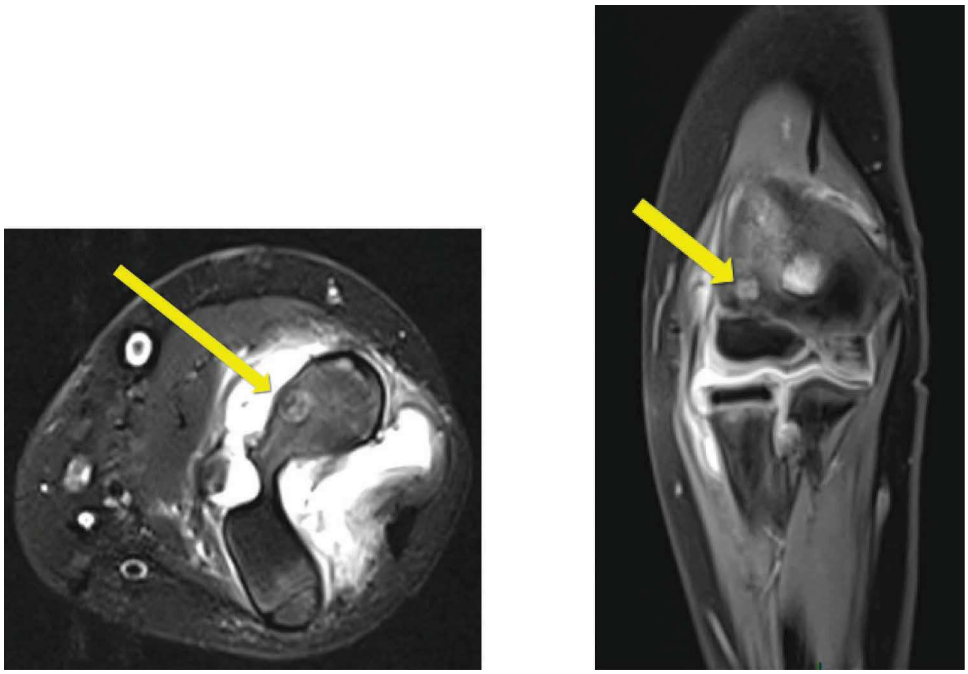

Figure 2. Magnetic resonance imaging of the osteoid osteoma. The arrows show a small, heterogeneous, well-defined, 5 mm lacunar lesion located in the lateral part of the left humeral metaphysis.

involve the diaphysis or metaphysis of the long tubular bones (the tibia and femur being the most frequent) and consist of a radiotransparent 'nest' located in the centre of a fusiform cortical thickening (1).

A migration of the lesion is possible which, from subperiosteal, changes position to become intracortical, endo-osseous or even intramedullary, causing continuous remodelling of the bone (1).

Several cases of osteoid osteoma mimicking arthritis have been described in the literature: if the benign tumour is localized intra-articularly, it can reach the cartilage and cause an intra-articular effusion mimicking arthritis (1–4).

Osteoid osteomas and rheumatic diseases produce excessive prostaglandins at the site concerned, which explains why NSAIDs provide clear pain relief (5). If management is conservative, diagnosis can take years, especially for intra-articular lesions, as shown by Szendroi et al in a small series, with an average time to diagnosis of 26.6 months for these lesions compared to 8.5 months for lesions in other locations (2, 5–7). The mean time for the diagnosis is between 4 months and 5 years (6).

Standard radiography, although still indispensable to exclude other diagnoses (5), may miss the osteoid osteoma and is largely surpassed by the computed tomography (CT) scan in thin sections, which remains the technique of choice, according to the literature (2, 5, 6). MRI does not seem to be as good for diagnosis, with up to 35% misdiagnosis (1, 8), although these statistics improve slightly after gadolinium injection, given the hypervascularization of the osteoid osteoma (9).

Bone scintigraphy can also be useful, with excellent sensitivity for the detection of osteoid osteoma (1), but in a study by Sharma et al conducted on 31 patients, single-photon emission computed tomography (SPECT)/CT had the best sensitivity and specificity for the detection of these lesions (10). It is also the technique that provides the most accurate information not only for the identification of the lesion and therefore the diagnosis, but also on its location for optimal therapeutic management (1, 10).

Removal of the lesion resolves the symptomatology without recurrence if the resection is complete (1, 11). The lesion may also involute spontaneously (1). Various surgical techniques have been developed, including CT-assisted radiofrequency ablation. The results of CT-assisted radiofrequency ablation are similar to open surgery (1, 8). This technique can be useful for locations that are difficult to envisage in open surgery. Other techniques have also proven their usefulness, including ablation using microwaves and high-intensity focused ultrasound under MRI control (1).

While osteoid osteoma is a challenge for the rheumatologist, paediatrician, orthopaedist, and radiologist, the pathologist usually has the last word. The histological image shows a nest, 1.5–2 cm in size, consisting of a network of trabeculae of osteoid matrix with varying degrees of calcification surrounded by osteoblasts, and in which a richly vascularized stroma is observed (2, 3).

In conclusion, osteoid osteoma should be considered in any patient with persistent and local pain, especially at night, which is well relieved by NSAIDs, and should

be included in the differential diagnosis of monoarthritis refractory to conventional treatments.

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References

1. Bhure U, Roos JE, Strobel K. Osteoid osteoma: multimodality imaging with focus on hybrid imaging. Eur J Nucl Med Mol Imaging 2019;46:1019–36.
2. Torrent J, Bailez A, Asuncion J. Recurrence of an intra-articular osteoid osteoma of the great toe: a case report and review of the literature. J Surg Case Rep 2017;1:1–4.
3. Lee EH, Shafi M, Hui JH. Osteoid osteoma: a current review. J Pediatr Orthop 2006;26:695–700.
4. Kayser F, Resnick D, Haghighi P, Pereira Edo R, Greenway G, Schweitzer M, Kindynis P. Evidence of the subperiosteal origin of osteoid osteomas in tubular bones: analysis by CT and MR imaging. AJR Am J Roentgenol. 1998 Mar;170:609–14.
5. Sagtas E, Gokkus K, Aydin AT. Intra-articular osteoid osteoma as a cause of anteromedial knee pain. Case Rep Orthop 2017;2017:5846368.
6. Eggel Y, Theumann N, Luthi F. Intra-articular osteoid osteoma of the knee: clinical and therapeutical particularities. Joint Bone Spine 2007;74:379–81.
7. Szendroi M, Köllo K, Antal I, Lakatos J, Szoke G. Intraarticular osteoid osteoma: clinical features, imaging results, and comparison with extraarticular localization, J Rheumatol. 2004 May;31:957–64.
8. Lindner NJ, Ozaki T, Roedl R, Gosheger G, Winkelmann W, Wortler K. Percutaneous radiofrequency ablation in osteoid osteoma. J Bone Joint Surg Br 2001;83:391–6.
9. Liu PT, Chivers FS, Roberts CC, Schultz CJ, Beauchamp CP. Imaging of osteoid osteoma with dynamic gadolinium-enhanced MR imaging. Radiology 2003;227:691–700.
10. Sharma P, Mukherjee A, Karunanithi S, Nadarajah J, Gamanagatti S, Khan SA, Bal C, Kumar R. 99mTc-methylene diphosphonate SPECT/CT as the one-stop imaging modality for the diagnosis of osteoid osteoma. Nucl Med Commun 2014;35:876–83.
11. Muller PY, Carlioz H. Récidive ou persistance d'un ostéome ostéoïde: une observation. Rev Chir Orthop 1999;85:69–74.

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