

Case Report

Intraosseous uric acid tumor without joint involvement: case report

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Abstract

We report an unusual case of extra-articular gouty tophus in the left medial malleolus. A 33-year-old man with a previous diagnosis of chronic gout presented with mild, non-disabling ankle pain associated with gout attacks. Imaging and histopathologic findings were inconclusive. The tumor was surgically resected, and the cavity was filled with methyl methacrylate. Histopathology confirmed the diagnosis in a sample collected intraoperatively. Giant cell tumor and bone cyst were ruled out. The patient had a good postoperative outcome.

Level of Evidence V; Therapeutic Study; Expert Opinion.

Keywords: Uric acid; Giant cell tumors; Bone cysts; Case reports; Diagnosis.

Introduction

The incidence of hyperuricemia leading to the formation of gouty tophi has increased in the last decades, given that risk factors associated with its etiology have become increasingly common in everyday life. The formation of gouty tophi results from chronic gout, which not always presents characteristic signs and symptoms and may vary according to the anatomical site. The classic manifestation of gout is an acute attack with inflammatory reaction, resulting in redness, burning sensation, increased sensitivity, swelling, and loss of function of the affected joints, among other systemic symptoms such as fever⁽¹⁾. Therefore, patients with chronic gout are predisposed to the deposition of monosodium urate (MSU) crystals in tissues, especially inside and around joints, forming tophi. In addition to tophus formation, chronic gout can manifest by chronic synovitis, bone erosions, and cartilage damage. This disease often affects joints, and immune mechanisms lead to the formation of MSU crystals. The accumulation of crystals results from decreased vascularity and susceptibility of the synovial membrane to pass the crystals. For this reason, gout tends to affect mainly peripheral joints⁽²⁾.

If gout is not controlled, attacks may occur leading to the development of tophaceous deposits, which, as mentioned above, mainly affect joints. However, they may also affect the subcutaneous tissue, renal tissue, tendons adjacent to the joint, and other less common sites, thus resulting in deformities characteristic of this disease. Rarely, tophaceous deposits can occur in intraosseous regions, with only a few cases reported in the literature⁽³⁾. In this context, we report a case of an unusual presentation of an extra-articular, intraosseous gouty tophus located on the medial malleolus of the left ankle.

Case Description

This rare case was reported after obtaining approval from the Research Ethics Committee and written consent from the patient.

A 33-year-old man, an agronomist, social drinker and non-smoker, presented with pain. He denied fever, use of medications, and underlying diseases, but reported a previous diagnosis of chronic gout. He reported a mild pain in the region of the medial malleolus of the left ankle for the

Study performed at the The Good Samaritan Charity Association, Maringá, PR, Brazil.

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past 6 months, which worsened during gout attacks. Physical examination showed mild hyperemia on the medial malleolus, with moderate edema, pain on palpation of the region, mild local bulging, and full range of motion of the ankle, with no joint locking and no signs of ankle ligament instability. The patient had no history of trauma in the affected area. He reported that the area began to bulge, growing slowly over the last 6 months, leading to non-disabling pain.

A radiograph of the ankle showed the presence of a nonspecific mass - a pseudotumor on the medial malleolus (Figure 1). Computed tomography (CT) and magnetic resonance imaging (MRI) showed a heterogeneous, expansile, infiltrative mass in the distal tibia with disruption of the medial cortex (Figures 2 and 3). The results of intraosseous needle aspiration and histological analysis were inconclusive. Therefore, we surgically resected the tumor and sent the resected specimen for pathological analysis.

During the procedure, we found a bone bulge in the metaphyseal region of the distal tibia, covered only by a thin capsule without medial cortical support. After resection and curettage of the cavity, we observed no communication with the talocrural joint (Figure 4). After debridement of the canal, we filled the cavity with methyl methacrylate. Histopathology revealed fibroconnective tissue with MSU deposits, clearly showing amorphous eosinophilic material, which confirmed the diagnosis of intraosseous gouty tophus.

The patient had no complications. No weight-bearing was advised immediately after surgery. Postoperative radiographs are shown in Figures 5 and 6. We removed the stitches

on day 15, and the patient started physical therapy. After 3 weeks, the load was progressively increased. Over the course of the month, he no longer had limitations or residual pain. After all procedures, the patient was also followed up at the rheumatology clinic.



Figure 1. Preoperative anteroposterior radiograph of the ankle showing an eccentric lytic lesion, with thinning and disruption of the medial cortex of the medial malleolus, without invading the articular cortex.



Figure 2. Coronal T1-weighted magnetic resonance imaging of the ankle showing expansion of the lesion to the medial cortex.

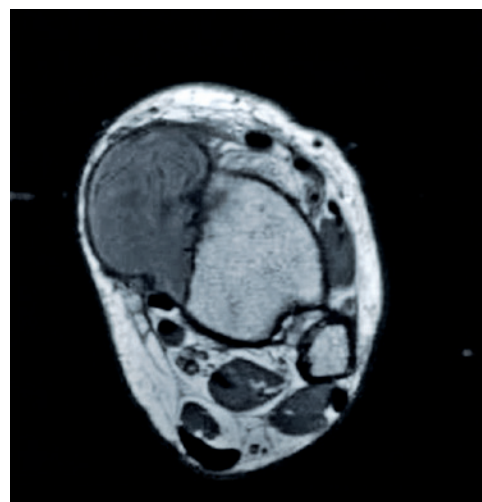


Figure 3. Axial T1-weighted magnetic resonance imaging of the ankle showing expansion of the lesion beyond the medial cortex.

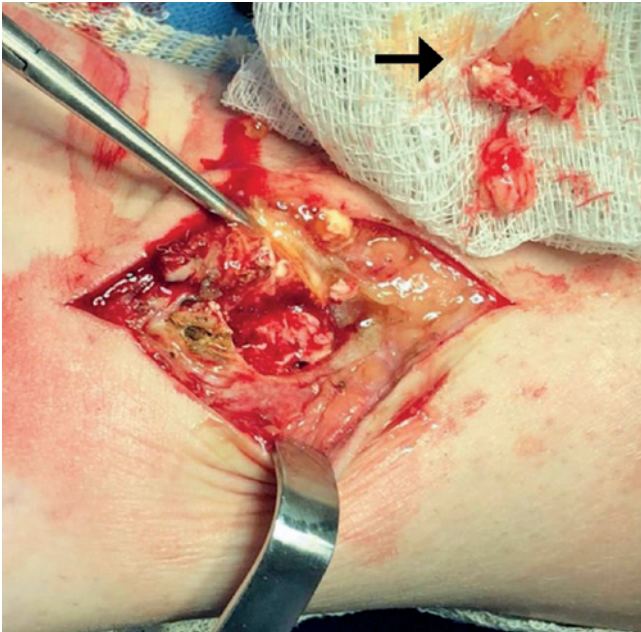


Figure 4. Intraoperative image - aspects of the tumor mass. Direct medial access to the ankle, dissection by planes, and identification of friable bone tissue, which was excised (arrow). There was no invasion of the articular surface.

Discussion

Tophi are formed by chronic MSU crystal deposition. Rarely, these depositions can occur in the bone itself, thus forming intraosseous gouty tophus, which is a rare condition with few reports in the literature⁽⁴⁾.

Pathophysiologically, MSU crystals deposit first into the Haversian system, slowly enlarging the cavity, which will be filled with a large amount of MSU crystals. The tophus enlarges in a way that adjacent structures may be invaded, such as the metaphysis of the bone, thus destroying the interior of the bone as the bone expands due to tophus growth⁽⁴⁾. Radiographic findings of bone erosion, resulting from tophus growth, are punched-out, round, or oval lesions with sclerotic rims. Regarding MRI, studies have shown limited specificity for the diagnosis of local disease⁽⁵⁾. Plain radiographs are often used for preliminary diagnosis, with CT and/or MRI being used to establish the differential diagnosis⁽⁶⁾.

It is necessary to differentiate intraosseous cystic gout lesions from other pathologies, such as giant cell tumor (GCT) and bone cysts. GCT is a neoplasm characterized by the proliferation of osteoclast-like cells, known as multinucleated giant cells⁽⁷⁾. Although usually benign, this proliferation can cause substantial changes in the local bone architecture, causing problems if located periarticularly. Typically, GCT is not visible in the mineralized matrix and lacks well-defined



Figure 5. Postoperative anteroposterior radiograph of the ankle showing lesion site filled with methyl methacrylate, supported by intact subchondral bone in the lower tibia, without extravasation of bone cement into the joint due to the integrity of the articular rhyme.



Figure 6. Postoperative lateral radiograph of the ankle showing lesion site filled with methyl methacrylate, without extravasation of bone cement into the joint due to the integrity of the articular rhyme.

sclerotic borders. The lesions are usually located in the epiphyseal region and tend to extend to the subchondral bone. CT and MRI are used to confirm lesion characteristics and differentiate them from other types of tumors, which may be confused with GCT⁽¹⁾.


Simple bone cysts are benign intraosseous lesions that appear as cavities filled with fluid and scattered giant cells⁽⁸⁾. Characteristically, cysts will contain yellowish viscous fluid if there is no associated fracture causing bleeding. Because these lesions are usually asymptomatic, diagnosis is made by routine imaging, such as ultrasound and radiographs. On imaging, cysts appear as radiolucent, well-defined, unilocular lesions. In addition, the cyst wall, composed of fibrovascular stroma, is thin and has an irregular or jagged appearance, with or without sclerotic rims⁽⁹⁾. In most cases, histopathology defines the diagnosis. However, the differential diagnosis with osteosarcoma, GCT, and other types of tumors requires a

correlation between clinical, radiographic, and histopathologic features to be determined, given the similarities they share⁽⁸⁾.

Based on the case reported here and on the analysis of the literature, we highlight the rarity of the case and the importance of distinguishing between differential diagnoses, such as GCT and simple bone cysts. In view of the features presented, the procedures were performed with the purpose of excluding differential diagnoses, improving quality of life, and reducing the patient's limitations.

Conclusion

Intraosseous gouty tophus is a rare, benign, poorly diagnosed condition in an uncommon site. In this case report, we showed approaches and challenges for diagnosis and treatment, since this type of tumor is rare and the postoperative recurrence rate is still unknown.

Authors' contributions: Each author contributed individually and significantly to the development of this article: EVJ *(<https://orcid.org/0000-0002-1385-2224>) Conceived and planned the activity that led to the study, wrote the article, participated in the review process; VP *(<https://orcid.org/0000-0002-9024-8071>) Conceived and planned the activities that led to the study, interpreted the results of the study; VMG *(<https://orcid.org/0000-0002-3574-833X>) Wrote the article, participated in the review process; VDP *(<https://orcid.org/0000-0003-2056-8253>) Wrote the article, participated in the review process.; MOA *(<https://orcid.org/0000-0002-3811-5810>) Wrote the article, participated in the review process. All authors read and approved the final manuscript. *ORCID (Open Researcher and Contributor ID) .

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