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Gouty Tophi in Sinus Tarsi of Bilateral Feet Mimicking Synovial Sarcoma: A Case Report

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ABSTRACT

Chronic gout is defined as accumulation of monosodium urate crystals in joints, cartilage, tendons, bursae, bone, and soft tissue. The foot is the most common location for acute gout flares, with the first metatarsophalangeal joint being the most frequent site of tophus formation. However, few studies have reported gouty tophus formation in the subtalar joint. Gout has been termed the “great mimicker” because of its tendency to mimic other pathologic conditions, such as pigmented villonodular synovitis and synovial sarcoma. Herein, we present a rare case of chronic tophaceous gout in the sinus tarsi in both feet in a 23-year-old healthy male, with extensive bony erosions mimicking pigmented villonodular synovitis and synovial sarcoma. We discuss the clinical presentation, distinguishing radiologic characteristics, surgical procedures, and outcome regarding this unique presentation.

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Gout is a form of inflammatory arthritis defined by the inability to metabolize purines (1,2). It is characterized by deposition of monosodium urate (MSU) crystals in joints and periarticular soft tissue (3). Epidemiologic studies have shown a higher prevalence of gout among US males at 5.9% (6.1 million) compared with females at 2.2% (2.2 million) (4). Clinically, acute gout flares present with joint swelling and pain, versus chronic gout, which is defined as accumulation of MSU crystals in joints, cartilage, tendons, bursae, bone, and soft tissue (5). Commonly, the first episode of gouty arthritis in males appears after age 40 years (6), and the foot is the most common location for gout (7), with the first metatarsophalangeal joint being a frequent site of tophus formation (8). Few studies have reported gouty tophus formation in the subtalar joint (9,10). However, gouty tophus formation, specifically in the sinus tarsi of bilateral feet in a relatively young healthy patient, has not been reported in the literature to the best of our knowledge.

Typical imaging features of tophaceous gout have been well described in the literature (11). Tophaceous deposits have the tendency to resemble other pathologic conditions, such as pigmented villonodular synovitis (PVNS) and synovial sarcoma (12). However, there are studies lacking in the literature, in which malignant neoplasm is a differential diagnosis in more challenging cases of tophaceous gout. The mentioned pathologies share a number of features with gout or with

tophaceous deposits, which makes definitive diagnosis challenging based on imaging alone (13). Herein, we present a rare case of chronic tophaceous gout of the sinus tarsi in bilateral feet in a relatively young healthy individual, with extensive bony erosions mimicking PVNS and synovial sarcoma.

Case Report

The patient was a 23-year-old male of Asian descent who presented to our outpatient clinic in January 2017 with an intermittently painful and slow-growing soft tissue mass at the level of the sinus tarsi of both feet (Fig. 1). Masses in both feet appeared > 7 years ago after the patient sustained a right ankle sprain while playing basketball. On the Visual Analog Scale (VAS), he related 2 out of 10 aching, cramping, and diffuse pain once or twice a year in both masses. He has a medical history significant for gout to the first metatarsophalangeal joint and the metacarpophalangeal joint of bilateral upper and lower extremities, and hyperlipidemia. The patient stated that the pain he experienced in the masses of both feet was not the same as his gout flares. He has been taking allopurinol 100 mg and fenofibrate 54 mg daily. He was a former smoker and previously smoked 2 to 3 cigarettes every 3 to 4 months. His most recent uric acid level was 8.5 mg/dL. On initial examination, the masses were protruded and nonmobile and did not transilluminate. There was no pain on direct palpation to the masses, and Tinel and Valleix signs were negative on percussion of all pedal nerves bilaterally. No sensory deficit was noted when tested with Semmes-Weinstein 5.07-g monofilament. Muscle strength was 5 out of 5 to all lower extremity muscle groups, and there was no pain with passive or active

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Fig. 1. Palpable mass to the sinus tarsi of the left foot measuring 5 × 6 cm, outlined before surgery.

range of motion (ROM) to joints of the foot and ankle bilaterally. Subtalar joint ROM measured 22° inversion and 8° eversion bilaterally.

Plain film radiographs of the right foot showed erosions to the lateral aspect of the navicular and medial aspect of the cuboid, best visualized in the oblique projection. Plain film radiograph of the left foot showed extensive multifocal lytic destructive changes at the level of the sinus tarsi, involving the navicular, cuboid, talus, calcaneus, and lateral cuneiform bones (Fig. 2). Magnetic resonance imaging (MRI) with and without contrast of both ankles was obtained to further assess the masses. A T1 hypointense and T2 hyperintense mass measuring 2.4 × 2.4 × 1.9 cm in the right foot and 2.6 × 4.7 × 3.1 cm mass in the left foot were noted. Both masses were arising from the anterior lateral sinus tarsi and extending dorsally displacing the extensor digitorum brevis (EDB) muscle belly superolaterally. Both masses demonstrated adjacent bony remodeling without substantial bone marrow or soft tissue edema (Fig. 3). Given imaging characteristics and bilateral occurrence, PVNS or synovial sarcoma were the tentative differential diagnosis. Chronic gouty tophus formation was possible but was low on our list of differential diagnoses.

Because of high suspicion of a malignant neoplasm, we recommended surgical excision and biopsy of the mass to the right foot first because the left foot was asymptomatic at that time. Goals of surgery involved (1) complete excision of the mass and the specimen sent to the pathology department as a frozen section during surgery, and (2) bone biopsy with curettage and insertion of crushed cancellous bone chips into the bony defects.

The patient was placed on the operating table in the supine position. A thigh tourniquet was applied to the right lower extremity and inflated to 325 mm Hg. During surgery, the EDB muscle was identified and reflected. A large mass consisting of a chalky white tophaceous formation was identified in the sinus tarsi and floor of calcaneus, which was consistent with tophaceous deposits (Fig. 4.) The white tophaceous mass was completely excised from the sinus tarsi and sent to the pathology department (Fig. 5). Intraoperative frozen section was deferred because of findings consistent with tophaceous gout. Debridement of bone from the lateral aspect of the navicular and medial aspect of the cuboid was also performed with a curette. The area was flushed with copious antibiotic solution using a pulse lavage. The bony defect was filled with allogenic crushed cancellous bone chips. The EDB muscle belly was reattached, and closure was completed in layers. The incision site was then dressed with a sterile compressive dressing.

After surgery, the patient was allowed to bear weight as tolerated to the right foot in a postoperative shoe. His incision site healed successfully within 3 weeks with no complications. He then underwent a similar procedure on the left foot 1 month later. The incision site to the left foot healed successfully as well, with no postoperative complications. The patient did not relate any pain to joints of bilateral feet with active and passive ROM. His subtalar joint ROM to both feet remains unchanged.

Histopathologic examination confirmed the diagnosis of tophaceous gout to both feet (Fig. 6). After 12 months of follow-up, the patient reported 0 out of 10 pain on visual analogue scale to bilateral feet and not experience any limitation in performing daily activities. Radiographs of both feet showed progressive consolidation of the osseous defect in the tarsal bones (Fig. 7). The patient's primary care physician made no changes in medical management for hyperuricemia after the surgery. He continues to take allopurinol 100 mg daily. Moreover, the patient's hyperlipidemia was being managed with fenofibrate 54 mg daily, which also has the ability to lower serum urate by increasing renal uric acid clearance (14).

Discussion

Tophaceous gout can be challenging to diagnose by imaging because it can mimic several entities. Perhaps the most important findings suggesting gout include juxta-articular osseous erosions with the Martel sign, which describes the overhanging margin of new bone along the edge of the erosion, along with dense gouty tophi buildup, both of which can vary in size. Most commonly, gout involves the first metatarsal phalangeal joint. However, it can involve other joints as well, with a predilection for smaller joints in the lower extremity and joints with a history of previous injury. It can also involve multiple joints simultaneously, usually in an oligoarticular pattern and usually asymmetrically. Radiographs are usually sufficient for diagnosis, but MRI may also be useful in less clear cases, because gout may be radiographically occult for up to 10 years (15).

Radiographically, overall bone mineralization is maintained, and as mentioned earlier, gout usually presents with characteristic punched-out bony erosions with an overhanging edge, along with calcification and dense tophi, which appear cloudy and occasionally with calcifications. The typical overhanging edge is seen in approximately only 40% of cases of tophaceous gout (11). In severe cases or in late stages, intraosseous calcifications may occur, and the extent of the erosions and tophi deposition may mimic entities such as hyperparathyroidism, PVNS, amyloid deposition, synovial osteochondromatosis, and rarely neoplasms such as synovial sarcoma. It has been suggested that osteosarcoma can produce a punched-out erosion (16). Computed tomography, especially dual-energy computed tomography, may be helpful in evaluating the extent of disease and for monitoring response to therapy, as is ultrasound, but play a lesser role in diagnosis (17).



Fig. 2. Anteroposterior and oblique radiographs of both feet. Note the cloudy, dense tophus (hollow arrows) and osseous erosions (solid arrows). The latter are more readily apparent radiographically in the left foot. Overall mineralization is maintained, and joint spaces are relatively preserved.

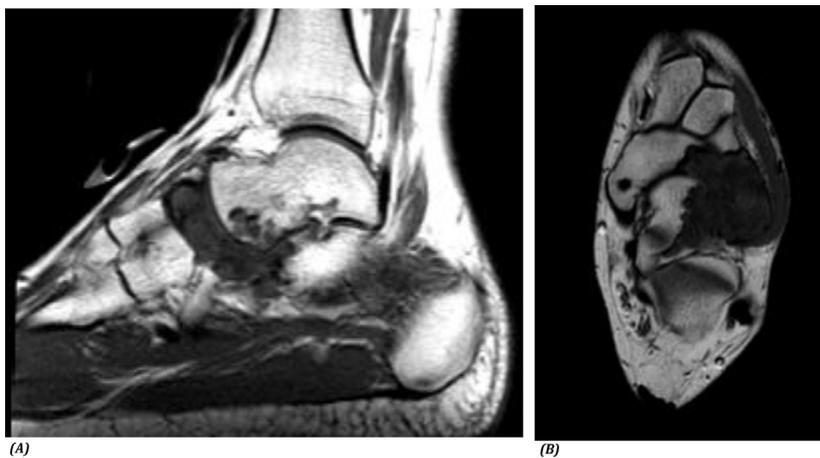


Fig. 3. Magnetic resonance scan of the left foot with sagittal (A) and axial (B) views. Soft tissue representing gouty tophus extends from the anterior/middle subtalar joint dorsolaterally. Calcaneocuboid, calcaneonavicular, and talonavicular joints were also involved, as well as the sinus tarsi. Note the osseous erosions best seen on the T1-weighted image (A) with avid enhancement on the T1 after contrast (B). In addition, note the relatively homogenous low signal on the T1-weighted image (A).

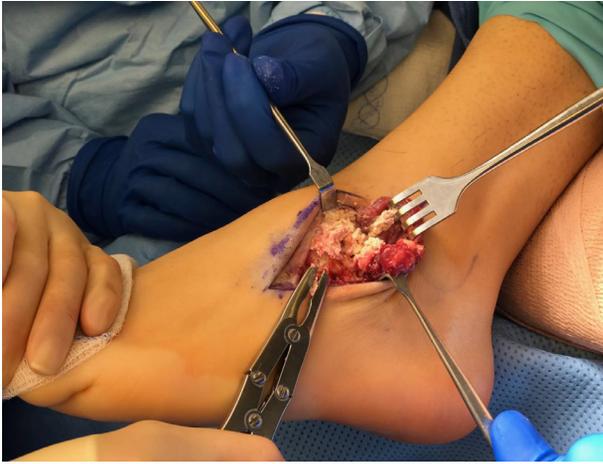


Fig 4. A linear 5-cm incision is made over the mass at the sinus tarsi of left foot. The extensor digitorum brevis muscle belly is reflected and retracted. Gouty tophi are visible in the sinus tarsi and debrided using a rongeur.



Fig 5. Multiple collections of gouty tophi are resected from the sinus tarsi and sent to the pathology department.

Synovial sarcoma commonly presents in young adults between ages 20 and 40 years, with most (80% to 95%) (18) tumors occurring primarily in extremities, with two thirds being located specifically in the lower limbs, and arise in the para-articular region of large joints (19). It is more closely related to tendons, tendon sheaths, bursa, and synovial sheath, which are some of the common locations for uric acid

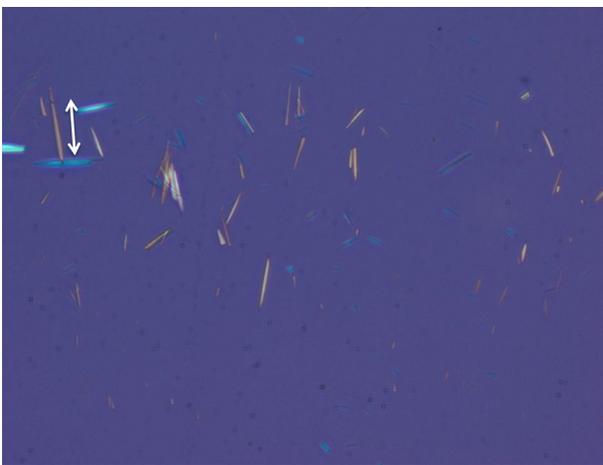


Fig 6. A needle-shaped negatively bi-refringent urate crystal appeared yellow when aligned parallel to the direction of polarization and blue when perpendicular, which is consistent with gout (magnification $\times 20$). The white arrow represents the direction of polarized light.



Fig 7. Radiographs show progressive incorporation of allograft to the osseous defect in tarsal bones.

deposition. MRI may be particularly helpful in these cases. In particular, gouty tophus usually does not demonstrate blooming artifact on gradient-echo MRI images caused by hemosiderin deposition seen in PVNS. Gouty tophus most commonly displaces tendons and muscles rather than invades them (may be seen with synovial sarcoma). Upadhyay et al (13) reviewed MRI features of 27 consecutive cases of biopsy-proven gout referred for suspected soft tissue sarcoma. All T1-weighted images demonstrated masses with low-to-intermediate signal. Fluid-sensitive imaging such as T2, short tau inversion recovery, or fat-saturated proton density-weighted images demonstrated a heterogeneous signal. In 17 cases (56%), a mild bone marrow edema-like signal was demonstrated (13), and these findings were consistent with MRI findings of our case as demonstrated in Fig. 3. However, the presence of an edema-like signal is relatively nonspecific and can be present in both arthritides and malignancy (20). Moreover, a low signal on T1-weighted images can be demonstrated in gout, as well as PVNS (21), making the diagnosis of gout in difficult cases even more challenging.

Tophaceous gout was not considered as the provisional diagnosis in this case for the following reasons. First, with respect to age, the patient was relatively young, and the first flare of gout in males is reported to be after age 40 years. Second, the patient complained of dull, aching, and diffuse 2/10 pain, 1 to 2 times a year, to the area of the mass, which is an atypical presentation for gout flares. Third, the location was the sinus tarsi bilaterally. Fourth, there were extensive erosions in the tarsal bones, mimicking a neoplasm. Fifth, no intraosseous calcification was seen on the radiographs.

The sinus tarsi is an atypical location for tophaceous gout formation. Post-traumatic tophaceous gout formation has been reported in the literature (22) because regional inflammation caused by the injury might trigger the precipitation of soft tissue tophaceous gout. However, this patient only reported a right ankle sprain that he sustained from playing basketball. He did not report any injury to the left foot or ankle and demonstrated normal subtalar joint ROM bilaterally.

In conclusion, in more challenging cases of gout in which a malignant neoplasm is suspected based on imaging, we recommend performing an excisional or incisional biopsy to rule out any malignancy. Moreover, if gouty tophus is noted during surgery, the patient should still undergo surgical debridement and curettage with application of allogenic bone graft to prevent further osseous erosion and decrease pain. Few retrospective and prospective studies have demonstrated surgical management of gouty tophus in the foot and ankle such as

curettage, debridement, and free flaps (23), excision of tophi with arthrodesis (24), and the arthroscopic soft tissue shaver technique (25), which are all acceptable surgical procedures. In this case report, we presented a 23-year-old male with tophaceous gout mimicking synovial sarcoma in the sinus tarsi who underwent curettage and packing of defect with allogenic bone graft. The patient has returned to full activity with no limitations and is pain free after 1 year of follow-up.

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