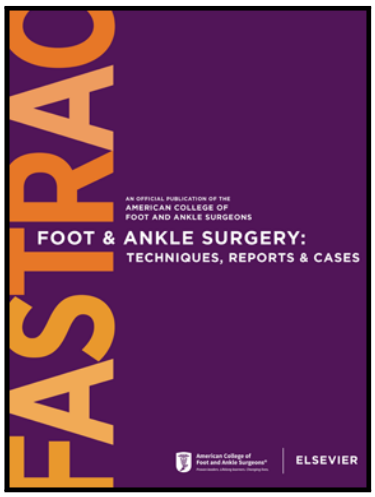

Synovial Osteochondromatosis of the Ankle Joint: A Case Study and Literature Review

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Abstract:

Synovial osteochondromatosis is a rare synovial proliferative disease that occurs because of chondral metaplasia within the synovium. This disease can occur intra- or extra-articularly.

Etiology is ultimately unknown. When conservative treatment fails, the cartilaginous nodules should be surgically removed. Although it has been reported all over the body, cases within the foot and ankle are far less common. We report a case of successful management of osteochondromatosis in an adult male that was surgically managed through a standard anterior ankle approach. At over two years of follow-up, he has since presented with no recurrence of synovial osteochondromatosis, significant reduction of pain post-operatively and has returned to full activity level.

Level of Clinical Evidence: 4

Key Words:

Ankle, Cartilage, Intra-articular, Nodule, Osteochondromatosis, Synovium

Introduction:

Synovial osteochondromatosis (SC) is a benign, tumor-like process of synovial tissue that was originally described by Henderson and Jones in 1923 (1). The disorder is an uncommon arthropathy of synovial joints characterized by the formation of intra-articular cartilaginous nodules in the synovial membrane (2). These nodules can detach and become loose bodies within the joint and may undergo secondary calcification and proliferation. It is believed to be caused by synovial metaplasia, although some indirect evidence may suggest a neoplastic origin. Synovial chondromatosis is more common in males, and current literature cites symptomatic presentation predominantly ranging from the third to fifth decade (3). The onset is usually insidious over a period of months to years.

Presenting complaints often include pain, swelling, sprains, paresthesias, and a palpable mass. The hip and knee are most often affected, with more than 50% of cases being reported in the knee (4). Synovial osteochondromatosis of the foot and ankle are exceedingly rare, with only a few cases reported in the literature. Locations reported include the tibiotalar, tarsometatarsal, metatarsophalangeal, calcaneocuboid, and naviculocuneiform joints (5). Radiographs frequently show calcified, juxta-articular bodies, but are often nonspecific. Computed tomography (CT) often shows mineralized densities in the corresponding region. Magnetic Resonance Imaging (MRI) typically shows loose bodies of low signal on T1-weighted images, high signal on T2-weighted images, and often, an associated joint effusion (6). However, the definitive diagnosis is achieved after histological examination of the synovial tissue (5).

Treatment involves surgical excision if conservative measures such as icing, resting, bracing, and anti-inflammatories fail. We report a case of synovial chondromatosis of the ankle joint treated by surgical excision and its outcome.

Case Report:

A 67 year-old male, with a past medical history of hypertension, bilateral knee joint osteoarthritis with subsequent right knee replacement, bilateral elbow joint osteoarthritis, and left hip joint osteoarthritis with subsequent hip replacement presented to our institution with a chief complaint of right ankle pain. Patient officiated high school volleyball in his free time and noted that over the 4 months prior to his visit the pain at the right ankle had progressed to a level of moderate pain. The patient denied any injury to the right foot or ankle during the 4 months prior to his appointment and stated that wearing an ankle brace did provide mild improvement in pain.

The dermatological, vascular, and neurological exams of the right foot, ankle and lower extremity were unremarkable. Musculoskeletal examination revealed tenderness to the right anterolateral, anterocentral and anteromedial aspects of the ankle joint. Palpable loose bodies were palpated within the capsule of the anterior right ankle. Tenderness on palpation was also appreciated along the course of the posterior tibial tendon and peroneal tendons. This tenderness was appreciated posterior as well as distal to their respective malleoli. Range of motion throughout the right foot and ankle joints was restricted with crepitus.

Initial radiographs of the right ankle revealed multiple irregular osseous bodies at the anterior aspect of the tibiotalar joint without overlying soft tissue swelling **(Figures. 1a&1b).** To further assess the extent of the bodies, an MRI of the right ankle was ordered and revealed ankle joint effusion with numerous round ossified loose bodies compatible with synovial osteochondromatosis. Severe osteoarthritis of the right ankle with bone-to-bone contact was also noted **(Figures. 2a-2c).** Computed Tomography (CT) imaging of the right ankle confirmed innumerable small calcified loose bodies in the anterior, anteromedial and anterolateral aspect of the ankle joint. The loose bodies measured up to approximately 0.8 cm. Distal tibia also demonstrated degenerative spurring along the anterolateral and posterior margin **(Figures. 3a&3b).** Due to failure of conservative management with continued pain, the patient underwent a right ankle arthrotomy with removal of synovial osteochondromatosis and anterior tibial cheilectomy.

Operative Technique:

Surgery was performed under general anesthesia in the supine position with a thigh tourniquet inflated to 275mmHg. Attention was then directed to the anterior ankle and at the

interval between the tibialis anterior (TA) tendon and the extensor hallucis longus (EHL) tendon, a linear incision was made. The incision was carried down through the anatomic layers taking care to protect neurovascular structures and obtain hemostasis with electrocautery. The incision was carried down to the level of the tibia and EHL tendon interval, and then taking care to maintain the TA tendon sheath, the interval was bluntly dissected with Metzenbaum scissors. The incision was carried down to the anterior aspect of the tibiotalar joint. The ankle joint capsule was incised giving access to a very large amount of loose body bony fragments **(Figures. 4a&b).** The bony fragments were removed without difficulty and sent to pathology for further examination. There was a large overlying lip of the anterior tibia and this was resected with an osteotome. The cartilage appeared grossly degenerative. Given the significant amount of cartilage loss, it was deemed not necessary to address the small lateral osteochondral defect as this was insignificant relative to the overall ankle arthritis. The ankle was taken through range of motion and there was no evidence of crepitus. The incision was irrigated with copious amounts of saline. The tourniquet was deflated, and hemostasis was obtained with electrocautery. Closure was performed in a layered fashion. The patient was placed in a cadillac splint

Post-Operatively:

Histopathological examination confirmed that the osteochondroid tissue fragments were consistent with osteochondromatosis. Following surgery, the patient was placed in a posterior splint for 2 weeks post-operatively and was instructed to be non-weight bearing. At the 2 week follow up, radiographs were obtained to confirm that all osteochondroid fragments had been removed. Sutures from the anterior ankle incision were also removed at this time. Patient was allowed to be weight-bearing as tolerated to the right leg in a Controlled Ankle Motion (CAM)

boot. At 3 week follow up, the patient was transitioned from a CAM boot to a regular shoe at the right leg with an ankle brace and allowed to walk as tolerated. Patient subsequently followed up at 6 weeks and 10 weeks post-operatively, at both times he reported no issues with range of motion, mobility, or weight-bearing at the right ankle. At the patient's last known follow-up, 12 months post-operatively, he reported no issues at the right ankle joint with regard to range of motion or mobility, and radiographs indicated no recurrence of SC loose bodies **(Figures. 5a&5b).**

Discussion:

Synovial chondromatosis (SC) of the foot and ankle is a rare condition with a rate of approximately 1 in 100,000 and is an even rarer entity in the foot and ankle, with few documented cases in literature (7). Synovial chondromatosis is an articular disease that is associated with cartilage fragments in the joint cavity. These fragments originate in the synovium and consist of hyaline cartilage (8). The symptoms are usually nonspecific and chronic. Symptoms generally include pain at rest or on movement, crepitus, joint stiffness, decreased movement and range of motion, and a locking or catching sensation. The physical findings vary with the joint involved and with the degree of involvement. There can be a localized or diffuse swelling of the joint involved with fixed or loose bodies palpable (9).

Synovial osteochondromatosis is typically classified according to its usual progression through three stages as described by Milgram, in 1977. In phase I, metaplasia of the synovial

intima occurs. Active synovitis and nodule formation is present, but no calcifications can be identified. In phase II, these nodules detach from the synovium to become loose bodies within the joint. The loose bodies are primarily still cartilaginous. In the first 2 stages, plain radiographs will show no gross abnormalities of the affected joint (5). Phase III is characterized by multiple loose bodies within the joint space with none remaining within the synovial tissue lining. Synovitis has typically resolved at this phase of the disease (8). It is at this stage that multiple intra- or extra-articular opaque loose bodies of various sizes are typically demonstrated as the loose bodies have calcified. Our patient was in stage III.

The classification may be further broken down into a primary versus secondary type. The process is considered primary if it occurs in an otherwise normal joint. Primary synovial osteochondromatosis has no common predisposing factors, represents de novo metaplasia of the synovium, and has a greater chance of recurrence (10). Secondary synovial osteochondromatosis is more common and is usually associated with osteoarthritis or a history of trauma. Other predisposing factors include osteochondritis dissecans, avascular necrosis, rheumatoid arthritis, and neuropathic osteoarthropathy (11).

The differential diagnosis includes other disorders that may give rise to loose bodies such as degenerative joint disease, osteochondritis dissecans, neurotrophic arthritis, tuberculous arthritis (rice bodies), and osteochondral fractures. A variety of synovial diseases should be included in differential diagnosis such as synovial hemangioma, pigmented villonodular synovitis, synovial chondrosarcoma, and synovial sarcoma (12). Magnetic Resonance Imaging (MRI) imaging can be of some use in distinguishing between SC and other differential diagnoses. Typical findings of SC on T1-weighted images include an appearance of the loose bodies with a low-intensity peripheral rim and a central intensity comparable to that of the soft

tissues. These findings are also evident on T2-weighted images. During the final stage of SC, loose bodies may be characterized by low-intensity rims with homogeneous hyperintensity close to the bone marrow on T1-weighted images and loss of signal on T2-weighted images (13).

The aim of any treatment for SC consists of decreasing pain and limiting the development of early osteoarthritis if possible. The only definitive treatment for SC consists of surgical removal of all accessible loose bodies at the affected joint and synovectomy when the synovial membrane is seen to be producing more bodies. The calcified bodies seen on plain radiographs may not be readily found in the joint cavity because they can be encased in the synovial membrane, they can be located in a bursa, or they may have not yet been liberated from the synovial membrane (12). Generally, in the early stage of the disorder, synovectomy is adequate. Synovectomy, together with loose body removal, is indicated in the 2nd stage of the disease. For later stages, generally, loose body removal will suffice (9).

The treatment method for SC in our case study consisted of the classical approach of open ankle arthrotomy with debridement and removal of all loose bodies. Although not mandatory, CT or MRI scans were obtained prior to surgery to assist in identifying and localizing the multiple loose bodies. Open arthrotomy has the advantage of visualization and access. However, it has higher risk of complications in addition to longer postoperative rehabilitation. Arthroscopic debridement of loose bodies is another option for surgical intervention of SC. Arthroscopy has the advantage of quicker rehabilitation and reduced pain, as well as fewer complications; however, visualization and access within the joint could be technically difficult depending on location and orientation of joint bodies (13). Both open and arthroscopic treatments have been documented as successful treatment options, so the choice of

intervention can be dependent on specific patient circumstances as well as the surgeon's comfort level with either modality.

It is important to note, there is conflicting evidence on whether performing a synovectomy can prevent recurrence or malignancy. Successful treatment has been described with and without synovectomy (14). In general, for SC, the recurrence rate has been estimated to be from 11.5% to 37.5% after operative treatment (5). Most recurrences occurred after more than 5 years after surgery. Recurrence is assumed to be the result of incomplete pathologic tissue removal, namely incomplete synovectomy. Irrigation of the joint during surgical intervention with 3% H2O2 has been described as a possible adjunctive intervention to prevent recurrence of SC, as it acts as a chemical cauterization and is not believed to be associated with added surgical complications (15).

Malignant transformation of SC into low-grade chondrosarcoma has been documented with up to 5% occurrence noted (9). Malignant transformation is closely connected to recurrence rate. Malignant transformation usually occurs many years after operative treatment, therefore long-term follow up is necessary to make the right conclusion about the rate of this serious outcome (6).

In conclusion, a comprehensive literature review and a case report with involvement of the ankle joint of synovial osteochondromatosis has been provided. Our patient was treated surgically, with no signs of recurrence to this date. Synovial osteochondromatosis is a rare condition, and a thorough clinical and radiological workup is the key to appropriate treatment.

Informed Patient Consent

☒ Complete informed consent was obtained from the patient for the publication of this study and accompanying images.

Declaration of interests

☒ The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements: None

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Figures:

Figure 1a:

Figure 1b:

Figure 2a:

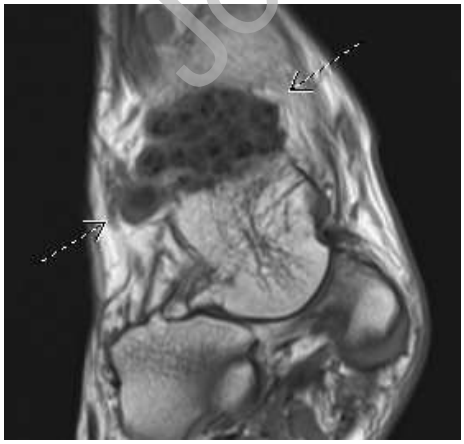
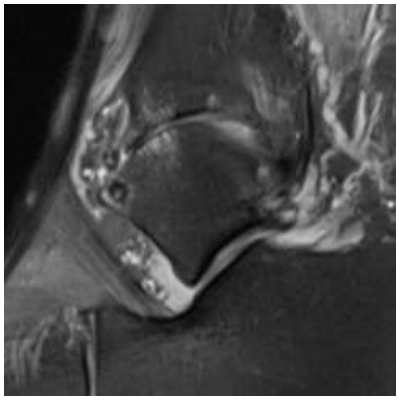


Figure 2b:



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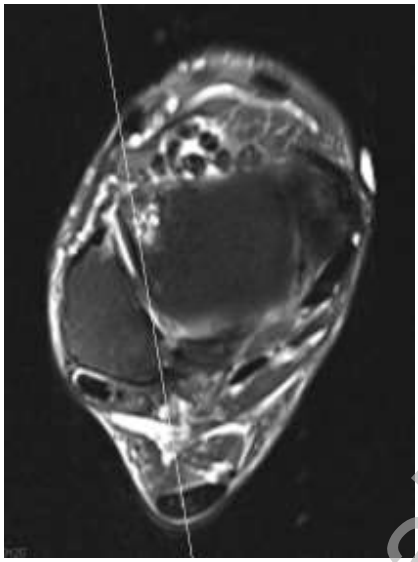

Figure 3a:

Figure 3b:

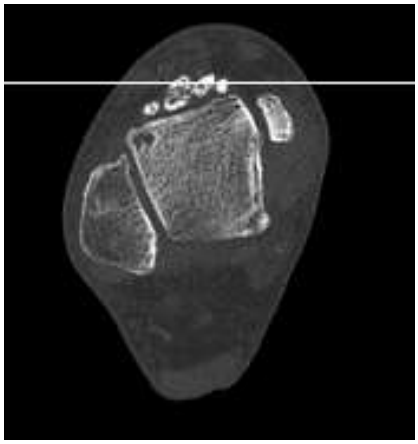

Figure 4: Intraoperative right ankle images

Figure 5a:

Figure 5b:

Figure Legends:

Figure 1a: Preoperative Anterior Posterior right ankle radiograph
Figure 1b: Preoperative Lateral right ankle radiograph
Figure 2a: Preoperative T1 Axial right ankle MRI
Figure 2b: Preoperative T2 Sagital right ankle MRI
Figure 2c: Preoperative T2 Axial right ankle MRI
Figure 3a: Preoperative right ankle Coronal CT demonstrating osteochondral defect
Figure 3b: Preoperative right ankle Axial CT demonstrating loose bodies
Figure 4a: Intraoperative right ankle image demonstrating loose bodies in the synovium after an ankle dissection
Figure 4b: Intraoperative right ankle image demonstrating the amount and size of the loose bodies

Figure 5a: 12-month postoperative anterior posterior right ankle radiograph
Figure 5b: 12-month postoperative lateral right ankle radiograph