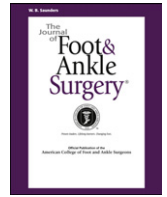




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Case Report

Asymptomatic Synovial Chondromatosis of the Ankle: An Incidental Finding

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ABSTRACT

Synovial chondromatosis is an uncommon, benign lesion of nodular cartilaginous neoplastic development of the synovium that can lead to loose bodies and arthritic degeneration if left untreated. Although very rare, malignant transformation to chondrosarcoma can occur. Primary and secondary forms of synovial chondromatosis also exist, and each has distinct clinical, radiographic, and histologic characteristics. In this article, we describe a case of extensive primary synovial chondromatosis of the ankle that was asymptomatic until just before presentation, and that was treated by means of open synovectomy with excision of the osteochondromatous lesions within the joint.

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First described by Leannac in 1813, synovial chondromatosis is a relatively rare, generally benign, monoarticular lesion consisting of multiple nodular cartilaginous bodies once thought to be the result of hyper-metaplastic development of the synovium. More recently, some investigators (1, 2) have come to consider the lesion to be the result of a benign, neoplastic process (1, 2); one that can lead to the presence of loose intra-articular bodies and subsequent degenerative joint changes if left untreated (1–8). The exact prevalence of ankle involvement with synovial chondromatosis is unknown, although it is thought to be a rare entity (3, 4). Larger joints are more commonly affected, with the knee being involved in up to 65% of reported cases (2). Other common sites include the hip, elbow, and shoulder, although cases have been reported to involve the wrist and interphalangeal and temporomandibular joints, as well as extra-articular locations (1–3, 7, 8). Males in the third to fifth decades of life are affected twice as often as are females (2, 4, 5, 9). Trauma has been considered the inciting factor in up to 50% of reported cases (1, 5), and surgical excision is considered to be the definitive treatment, although recurrence has been reported in up to 23% of patients (2, 8, 10). Although synovial chondromatosis is generally considered a chronic, progressive entity, with some cases reported to have spontaneously resolved (1, 2), malignant transformation is thought to occur in approximately 5% of cases associated with an extended clinical course and recurrent lesions (1, 2, 7, 8, 10).

In this article, we describe the case of an adult female who presented with right heel pain at the insertion of her Achilles tendon that,

upon clinical and imaging examinations, incidentally revealed the presence of asymptomatic synovial chondromatosis localized to her right ankle. The heel pain, we determined, was a result of the space-occupying lesion within the ankle, and the chondromatosis was treated by means of surgical excision. The patient responded well to surgical excision with partial synovectomy, and heel pain had resolved by post-operative week 3.

Case Report

A 58-year-old-female presented with a chief complaint of the sudden onset of pain localized to the posterior aspect of her right heel, aggravated by standing and walking. There was no history of recent trauma. She wore over-the-counter shoe inserts, and occasional warm soaks with Epsom salts had been performed with only minimal relief. Her past medical history included uterine carcinoma that was in remission, sarcoidosis with ocular manifestations, no current active lung disease (although approximately 20 years earlier she had pulmonary complications). In addition, the medical history was positive for right kidney disease, gout, hypertension, sciatica, and osteoarthritis. The surgical history included hysterectomy for the treatment of uterine carcinoma, left shoulder replacement, and repair of a traumatic laceration to the peroneus brevis that had occurred more than 50 years earlier. She had been a smoker for a 25-year duration, although she had quit smoking approximately 9 years before presentation to our clinic, otherwise the social history was unremarkable. Her family history was positive for diabetes mellitus affecting her mother and 2 female siblings.

Physical examination of the symptomatic area revealed mild tenderness to palpation over the tendo-Achillis insertion into the right heel, and the biomechanical evaluation demonstrated bilateral

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ankle equinus and flatfoot. Ankle range of motion was symmetrical bilaterally. Incidentally, soft tissue edema was noted overlying the anteromedial aspect of the right ankle, along with an underlying painless, indurated, mobile nodule that measured approximately 2.5×1.5 cm. The lesion was firm, nonpulsatile, and did not exhibit percutaneous transillumination of light. The affected area also demonstrated no deviation in skin temperature, hydration, or hair growth in comparison with the same areas on the contralateral lower extremity (Fig. 1). When questioned, the patient denied any pain associated directly with the palpable mass and the immediately adjacent surrounding tissues, although she did recall having intermittent swelling localized to the right ankle for several months before presentation. The remainder of the physical examination was unremarkable.

Standard weight-bearing radiographs of the right foot revealed soft tissue edema surrounding multiple nodules with punctate calcifications approximately 2-mm to 3-mm in diameter, likely cartilaginous in nature, located at the anterior aspect of the right ankle. No adjacent cortical disruption, osteophytic proliferation, or



Fig. 1. Soft tissue mass, anteromedial aspect of the right ankle.



Fig. 2. Weight-bearing lateral radiograph, right ankle, showing multiple calcified nodules anteriorly.

cystic formation was identified within the affected ankle (Fig. 2). Magnetic resonance (MR) images were obtained to further characterize the lesion, and our differential diagnoses included malignancy, pigmented villonodular synovitis (PVNS), and synovial chondromatosis. In particular, the presence of overt cortical destruction with adjacent marrow invasion would have been suggestive of possible malignancy (2). Contrast studies, which could have been helpful in delineating the lesions, were not used because of the patient's known renal disease. The noncontrast MR studies demonstrated synovial thickening with the presence of multiple intra-synovial round to ovoid bodies that were centrally isointense relative to muscle on T1-weighted sequences. On both T1- and T2-weighted

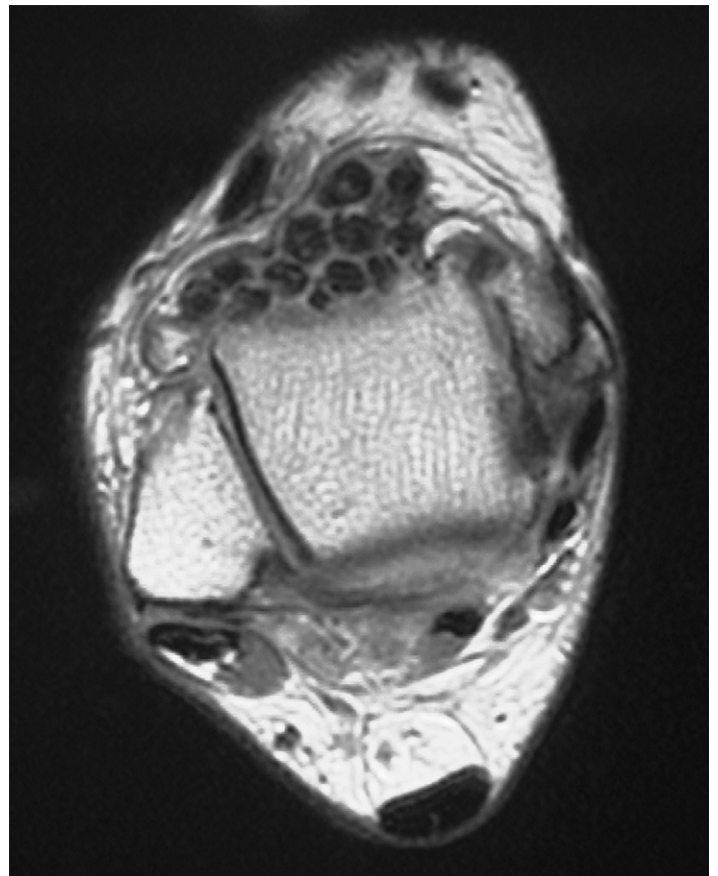


Fig. 3. T1-weighted axial magnetic resonance image scan showing multiple osteocartilaginous bodies, focal low-intensity areas within each body, well-defined rim with the central aspect isointense to muscle, with synovial hyperplasia.

images, focal areas of low signal intensity were seen within each body, which was suggestive of synovial chondromatosis. High signal intensity on T2-weighted sequences was also seen within the ankle, consistent with effusion (Figs. 3 and 4). Because there was no history of prior, direct ankle trauma or systemic inflammatory disease, primary synovial chondromatosis was strongly suspected. Consistent with the primary type of synovial chondromatosis, the imaging studies revealed multiple intra-articular bodies of similar size and shape with the absence of degenerative changes to the affected ankle. Based on the clinical symptomatology and imaging findings, the decision was made to proceed to the operating room for surgical inspection and excisional biopsy of the mass in an effort to make an accurate diagnosis and attempt to alleviate the initial complaint of posterior heel pain, which was thought to be directly related to the space-occupying lesions within the ankle.

In the operating room, a 3-cm longitudinal incision was made over the anteromedial aspect of the ankle, where immediately upon entry into subcutaneous tissue a large mass with lobulated adipose tissue was identified and dissected free. Embedded in this fibroadipose tissue were several fine, multicolored, silklike fibers 2.0 cm or shorter in length. These did not appear to be consistent with tendon, ligament, or joint capsule, or bone that would be expected in this area (Figs. 5 and 6). As blunt dissection was carried deeper, a second mass consisting of lobulated fibroadipose tissue with the same silklike fibers was encountered, and dissected free. These findings were incidental, and we speculated that the material could have been retained suture from the previous peroneus brevis repair, over 50 years ago per past surgical history, that had since migrated and subsequently become encapsulated in the form of a fibrolipomatous mass. Parallel to the initial incision, the synovium was longitudinally

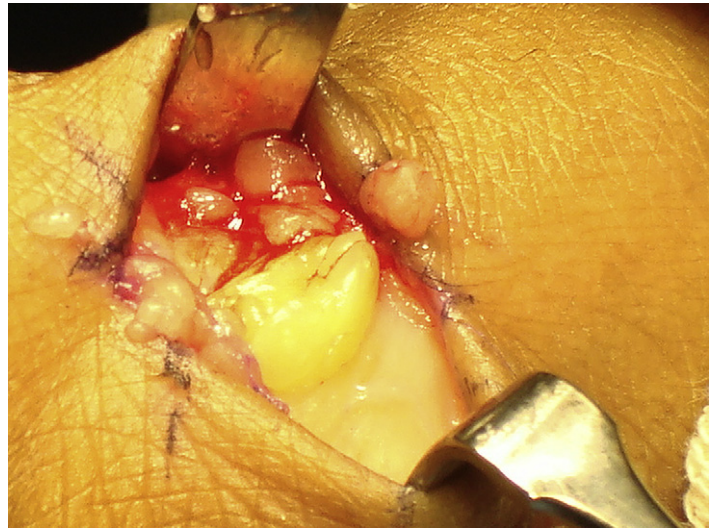


Fig. 5. Fibroadipose lobule with stringlike material.

incised allowing entry into the ankle to enable inspection via direct visualization. Manipulation of the ankle from medial to lateral revealed numerous mixed osseous-cartilaginous fragments that appeared to be freely mobile within the joint cavity, extending across the entirety of the anterior chamber of the ankle. A total of 38 distinct specimens were extracted individually with a forceps, and no resistance from soft tissue adherence was encountered with any specimen. All of the specimens were ovoid, firm, pearly white, mixed osseous, and cartilaginous tissue ranging from 2 to 5 mm in diameter (Fig. 7). Intraoperative C-arm fluoroscopy was used to ensure that all calcified loose bodies were removed. Partial synovectomy was then performed at the anterior ankle secondary to synovial hypertrophy. Layered closure allowed for accurate approximation of the synovium, subcutaneous tissues, and skin.

The gross pathological examination of the specimens was reported as multiple ovoid portions of white-tan osteocartilaginous tissue measuring $4.5 \times 3.3 \times 1.0$ cm in aggregate, a yellow lobulated soft tissue mass measuring $2.5 \times 2.5 \times 0.5$ cm in aggregate, and 5 linear portions of pale-tan stringlike material ranging from 1.0 to 2.0 cm in length, all less than 0.1 cm in diameter. Histologic findings included

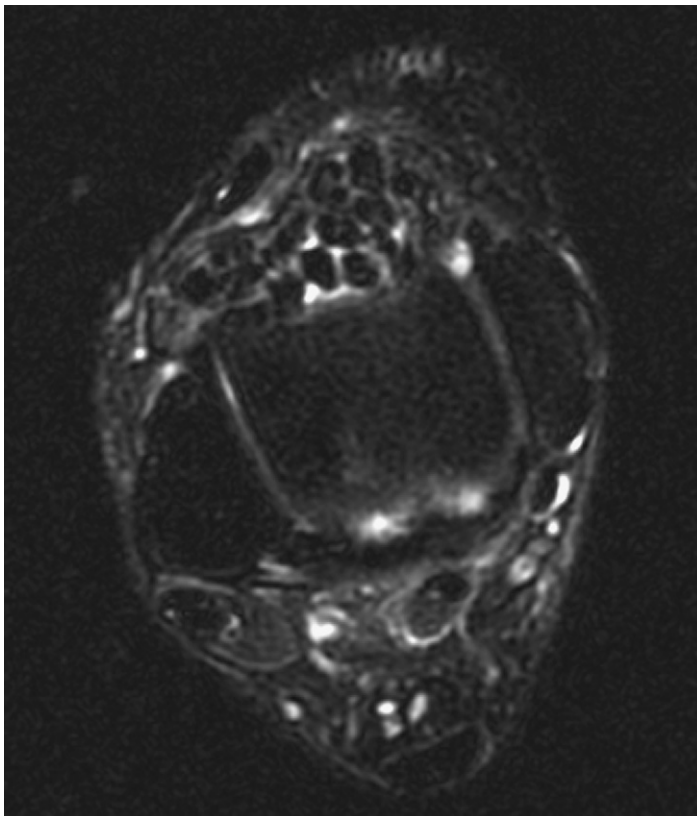


Fig. 4. T2-weighted fast spin echo (FSE) axial magnetic resonance image scan showing multiple osteocartilaginous bodies, diffuse low-intensity areas similar to that seen on the T1-weighted images.



Fig. 6. Additional fibroadipose lobule with stringlike material.



Fig. 7. Osteocartilaginous bodies removed from ankle joint.

multiple osteocartilaginous fragments from the synovium (Figs. 8 and 9). Also noted was the presence of mature fibroadipose tissue that was negative for acute inflammation and consistent with a lipoma, along with “stringlike foreign material,” as reported by pathology. Following open synovectomy with excisional biopsy of the multiple lesions, the patient progressed very well. Her posterior ankle pain resolved at 3 weeks postoperative, and the patient remained symptom-free, without clinical or radiographic evidence (Fig. 10) of recurrence at the 1-year follow-up examination. Because there is a high rate of recurrence, our ongoing plan is to continue to periodically monitor the patient for the chance of recurrence.

Discussion

Although the exact mechanism remains unclear, synovial chondromatosis is thought to evolve from spontaneous undifferentiated stem-cell proliferation in the stratum synoviale. It is thought that mesenchymal cells at the synovium-articular cartilage junction

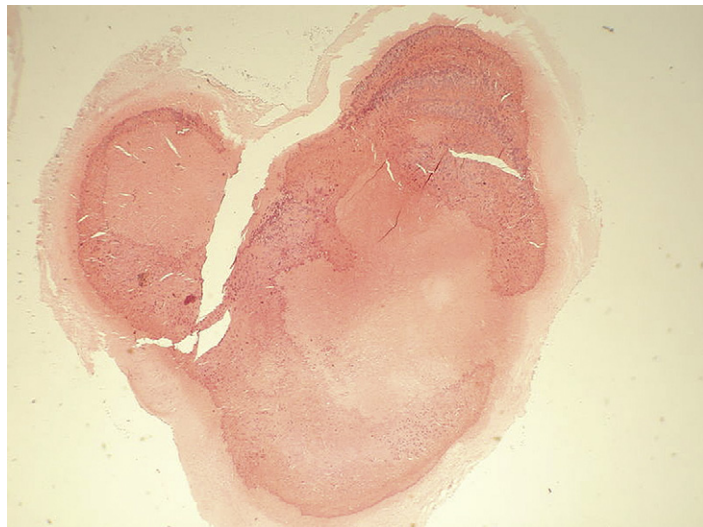


Fig. 8. Histology, original magnification $\times 10$, hematoxylin and eosin stain, depicting lobulated hyaline cartilage with surrounding synovium.

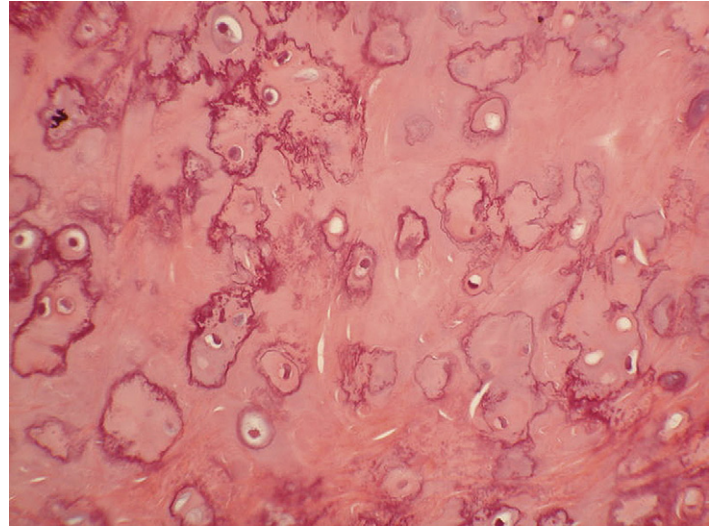


Fig. 9. Histology, original magnification, hematoxylin and eosin stain, depicting nuclear atypia with absence of mitoses.

proliferate, forming nodular foci of hyaline cartilage that often detach and remain as loose bodies within the synovial folds and articular cavity, and the nodules may calcify and even ossify over time (1, 3–5, 7, 10). In addition to associations with type 2 collagen and chromosome 6 abnormalities, and familial tendencies, cytogenetic aberrations with the potential for malignant transformation support the current notion that primary synovial chondromatosis represents a neoplastic process (1, 2, 10).

The clinical presentation of synovial chondromatosis is often insidious with gradual progression of nonspecific symptomatology, including a vague history of several years of joint pain, swelling, and stiffness. Pain is ultimately present in 85% to 100% of reported cases. In addition, the history often reveals a sensation of clicking, locking, or catching associated with the involved joint. Furthermore, common findings associated with synovial chondromatosis noted on physical examination include point-tenderness, diminished quantity and quality of joint motion, and the presence of a palpable mass adjacent to the joint (2, 3, 5, 7). Secondary synovial chondromatosis, on the other hand, is usually related to traumatic injury of the articular



Fig. 10. Weight-bearing lateral radiograph, 1 year postoperative.

hyaline cartilage, inflammatory arthritis, or an infectious process within the involved joint (2, 7).

Radiographic findings of multiple intra-articular bodies of similar size and shape, with a “ring and arc” pattern of chondroid mineralization are strongly suggestive of synovial chondromatosis (1, 2). MR images can be used to demonstrate the degree of synovial thickening while helping to distinguish between synovitis and effusion. The appearance of intra-articular bodies is, however, variable and dependent on the degree of calcification and/or ossification. Most commonly, calcific bodies demonstrate intermediate signal intensity on T1-weighted images, although low signal intensity is also observed. T2-weighted images and/or water-weighted images typically reveal high internal signal intensity. Furthermore, intravenous contrast can enhance visualization of these characteristics (2–4, 6, 7, 11). By comparison, computerized tomographic (CT) scans optimally depict calcified bodies while allowing for better visualization of extrinsic bone erosion, and therefore may be the preferred imaging modality in comparison to MR scans (2, 9). Secondary chondromatosis has greater variation with regard to the size of the chondral lesions with distinct joint abnormality, whereas primary synovial chondromatosis has an increased number of similar-sized lesions evenly distributed throughout the joint, and is associated with extrinsic bone erosion in 20% to 50% of cases (2). Histopathologic examination reveals a “cobble-stone appearance” of lobulated hyaline cartilage surrounded by synovium and some degree of nuclear atypia, without presence of mitosis (1, 2, 12). Secondary synovial chondromatosis lacks nuclear atypia while having concentric rings of hypocellular cartilage (2).

As a condition that somewhat rarely involves the ankle, the case of primary synovial chondromatosis presented in this article is instructive in that it was an incidental radiographic discovery in a middle-aged female who presented with a chief complaint of posterior heel pain, without mention of any abnormalities to the affected ankle. Other than a palpable, mobile, soft tissue mass with localized edema, the physical examination was unremarkable and the past medical history of sarcoidosis, gout, and uterine carcinoma seems to have been unrelated. Plain films and MR imaging revealed a characteristic appearance, namely multiple similar-appearing lesions without adjacent degenerative changes, indicative of primary synovial chondromatosis. We did not feel that the patient's chief complaint was

directly related to the synovial chondromatosis, and our decision to treat the chondromatosis surgically was based on the initial complaint of posterior heel pain, seemingly potentiated by the space-occupying lesions of the synovial chondromatosis within the associated ankle joint. The patient responded well to open excisional biopsy with partial synovectomy and was asymptomatic at the 1-year follow-up examination, with plans for additional follow-up on an annual basis.

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