

CASE REPORT

Primary Giant Synovial Osteochondroma of Ankle

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ABSTRACT

Background: Giant synovial osteochondroma (SOC) is a clinical variant of synovial chondromatosis wherein the size of the osteochondral nodule exceeds 2 cm. This is a rare clinical entity and finds its description in a few case reports or case series only.

Case description: A 28-year male patient presented with swelling of the ankle region for 5 months duration. Plain radiograph of the ankle showed well circumscribed, increased soft tissue density lesion in the posterior aspect of tibiotalar joint and talus. Magnetic resonance imaging (MRI) of the ankle showed a smoothly margined lesion close to the tibiotalar joint which was heterogeneously hypointense on T1-weighted images and hyperintense on T2-weighted images. Hypointense foci within the lesion in both images were suggestive of ossification. Intra-operatively, the swelling was noted to arise from the tibiotalar joint capsule with no evidence of bony erosion. The mass was excised in toto, and wound closure was done in layers. Microscopic examination of the mass showed predominant chondroid element with irregularly distributed chondrocytes in lacunae and evidence of enchondral ossification along with cancellous bone mass suggesting a synovial osteochondroma. On postoperative follow-up at 2 months and 9 months, movements around ankle joint were full range and pain-free with no reappearance of swelling. Imaging including plain radiograph and MRI revealed no evidence of recurrence at 9 months.

Conclusion: Giant SOC can present itself with the variety of symptoms. Imaging is helpful in ascertaining the diagnosis. The treatment of the condition remains surgical only. Post surgery, recurrence remains a possibility.

Clinical significance: Giant SOC should form one of the clinical possibilities in swellings arising from synovial joints as early diagnosis of the condition can prevent arthritic changes to progress further.

Keywords: Ankle, Giant synovial osteochondroma, Primary.

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BACKGROUND

Giant SOC is a clinical variant of synovial chondromatosis wherein the size of the osteochondral nodule exceeds 2 cm and may reach up to 20 cm.¹ This unusual entity finds its description in few case reports or case series, and only two cases have been reported for ankle region including one secondary osteochondroma of subtalar joint as searched on Pubmed and Ovid SP search engines.^{2,3} We present here, a case of primary giant synovial osteochondroma of the tibiotalar joint.

CASE DESCRIPTION

A 28-year male patient presented to our center with complaints of swelling in right ankle region for 5 months duration which was insidious in onset and gradually progressive. The swelling caused restricted movements of the ankle joint and difficulty in wearing high ankle boots for 2 months. The patient did not have any constitutional symptoms including fever, weight loss, anorexia, etc.

Clinical examination revealed a diffuse, non-tender, firm, smooth, fixed, non-fluctuant, swelling in right ankle region posterior to the lateral malleolus, measuring about 7 cm x 5 cm x 3 cm (Fig. 1). There was restricted but pain-free plantar flexion of 20 degree and full range dorsiflexion. There was no evidence of any tendon dysfunction around ankle joint.

The patient was evaluated with a plain radiograph of the ankle with anteroposterior and lateral views which showed well circumscribed, increased soft tissue density

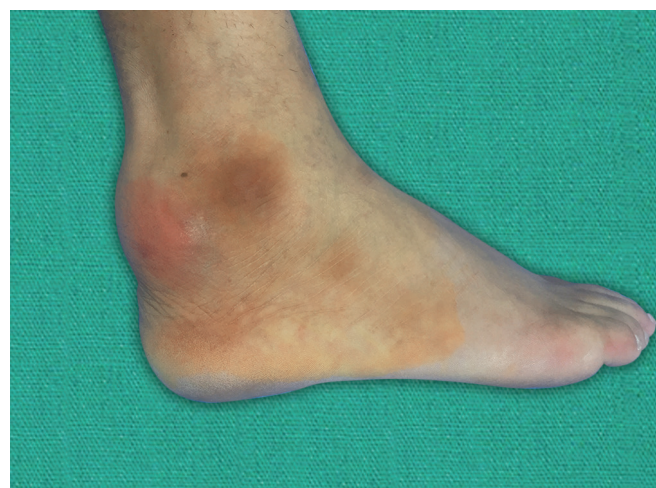


Fig. 1: Clinical photograph of the swelling over posterolateral aspect of right ankle joint

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Fig. 2: Plain radiograph of right ankle AP and a lateral view showing well-circumscribed soft tissue density lesion in the posterior aspect of right tibiotalar joint and talus. No erosions are seen in adjacent talus or calcaneum

lesion in the posterior aspect of tibiotalar joint and talus. Well defined nodular ossification was noted in the antero-inferior aspect of the lesion with no bony erosion (Fig. 2). The MRI of the ankle showed a smoothly margined lesion close to the tibiotalar joint which was heterogeneously hypointense on T1-weighted images and hyperintense on T2-weighted images (Fig. 3). Hypointense foci within the lesion in both images were suggestive of ossification. Short tau inversion recovery (STIR) images were heterogeneously hyperintense. There was moderate heterogenous post contrast enhancement (Figs 4 and 5). The radiological differentials included synovial hemangioma, pigmented villonodular synovitis, and lipoma arborescens along with osteochondroma of ankle joint.

Routine blood samples including inflammatory markers were found within the normal range. The patient was taken-up for complete excision under spinal anesthesia in the prone position, and the procedure was performed under tourniquet. A longitudinal incision was given directly over the swelling parallel to achilles

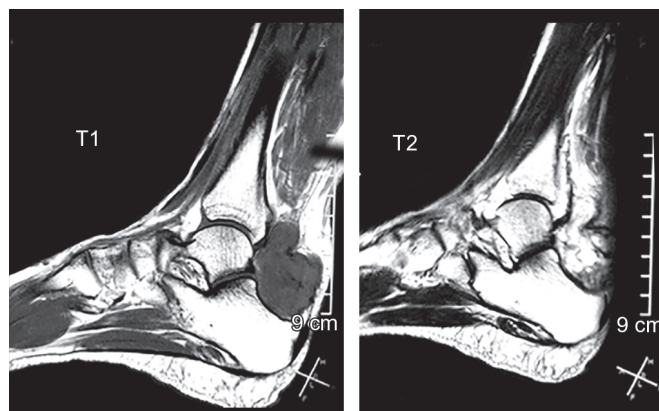


Fig. 3: MR images of ankle joint showing smoothly margined swelling in close relation to the posterior aspect of the tibiotalar joint. The lesion is heterogeneously hypointense in T1 weighted images and heterogeneously hyperintense in T2 weighted images. Hypointense foci were seen in both of them, suggesting ossification

tendon approx 8 cm in size. The swelling was noted to arise from structure deeper to tendons. Sural nerve was identified and protected during the dissection.

Further dissection was performed between flexor hallucis longus and Peronei. A 6.5 cm x 4.5 cm x 3.0 cm grey-white, bosselated tumor mass was noted compressing the surrounding structures hence making a false capsule around it (Fig. 6). The mass was noted to have communication with the tibiotalar joint capsule with no evidence of bony erosion. The mass was excised in toto, and wound closure was done in layers.

Microscopic examination of the mass showed predominant chondroid element with irregularly distributed chondrocytes in lacunae and evidence of enchondral ossification along with cancellous bone mass. Mature adipose cells were also noted between the bony trabeculae and lobules of cartilage. No osteoblastic or osteoclastic activity was noted. Examination of the periphery of a tumor confirmed the presence of pseudocapsule. These findings were suggestive of a SOC (Fig. 7).

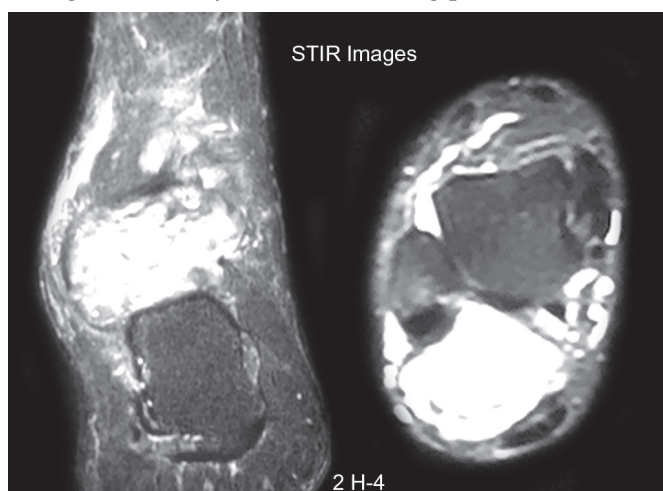


Fig. 4: The lesion is heterogeneously hyperintense in short tau inversion recovery (STIR) images

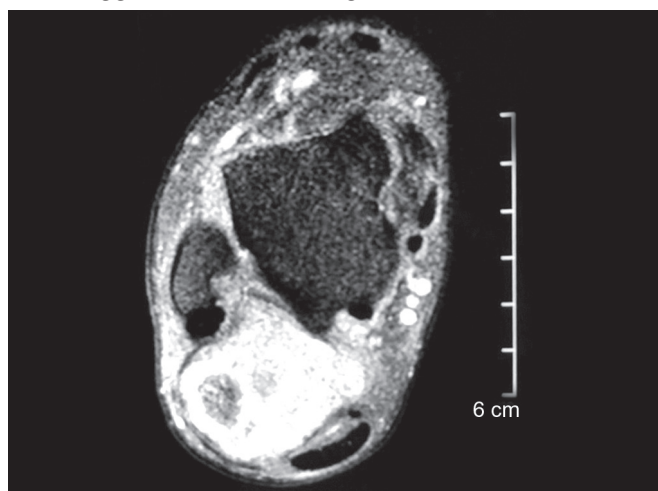
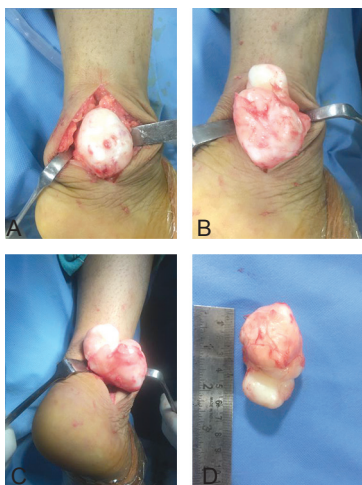


Fig. 5: T1 weighted fat suppression (FT) post-contrast image showing heterogenous moderate enhancement of the lesion.



Figs 6A to D: Intraoperative images of the lesion. Note the size of the lesion in image D

On postoperative follow-up at 2 months, 6 months, and 9 months, movements around ankle joint were full range and pain-free with no reappearance of swelling. Imaging including plain radiograph and MRI revealed no evidence of recurrence at 9 months (Figs 8 and 9).

DISCUSSION

Synovial chondromatosis is a rare condition and affects 1 in 100,000 with a slight male preponderance, mostly involving the large joints such as a knee, hip, elbow, and shoulder. Even the usual form of SOC is a rarity for ankle joint and occurs in less than 5% of cases.⁴

Giant SOC is a rare musculoskeletal neoplasm which has been described by many authors as a variant of synovial chondromatosis; while others consider it a separate entity altogether.⁵ The term giant SOC was first used by Edeiken¹ in the year 1994 and was added as phase IV in the Milgram's description⁶ of synovial chondromatosis. It is believed to have developed either from the coalescence of multiple small chondromas or the continuous growth and ossification of a single chondroma.

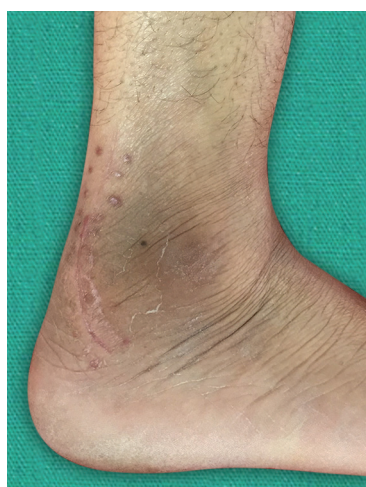


Fig. 8: Postoperative clinical photograph at 6 weeks

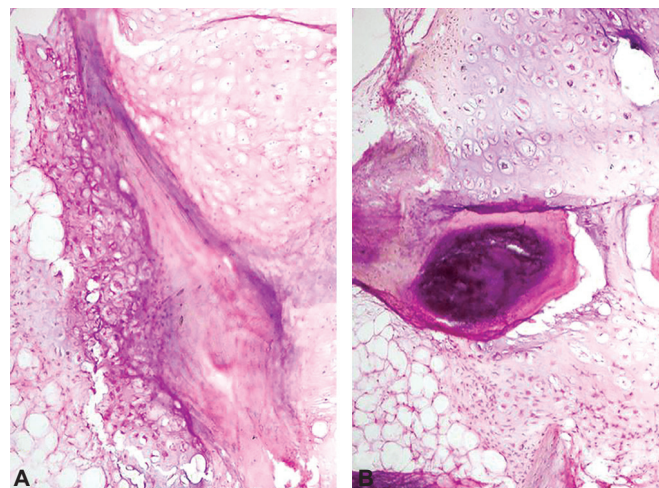


Fig 7A and B: H & E stained section showing (A) lobulated hyaline cartilage with evidence of enchondral ossification and (B) hyaline cartilage with unevenly distributed chondrocytes and foci of ossification. Fat spaces are seen along with compressed connective tissue of synovial membrane in the edge of the section

Giant SOC has been classified into primary and secondary groups based on etiology with definitive histopathological differences between these two varieties.⁷ Secondary osteochondroma is relatively common and has been attributed to various factors like trauma, osteonecrosis, osteoarthritis, osteochondritis dissecans, neuropathic arthritis, and rheumatoid arthritis.⁸ There are several case reports available for secondary SOC of different joints however the primary variety is even less reported in English language literature.⁹

Giant SOC usually presents itself as insidious onset gradually progressive swelling which becomes symptomatic due to loss of movements at adjacent joint or the compressive erosion of the joint cartilage and hence arthritis. This tumor has been reported in the elbow by Al-Najjim et al. in a middle-aged male patient causing ulnar nerve neuropathy due to mechanical effects.¹⁰ Lui et al. have reported secondary giant SOC of the subtalar



Figs 9A and B: Postoperative plain radiograph anteroposterior and lateral views at 09 months

joint in a 12 years old female patient as a clinicoradiologically aggressive tumor.¹¹ Two case reports are mentioning the presence of giant SOC in the Hoffa fat pad around the knee joint.^{2,12} Fornaciari et al. reported intra-articular giant SOC of ankle and knee joint in their case report. Yu et al. reported extra-articular giant SOC in the proximal thigh of a middle-aged female patient.¹³

Plain radiography may remain normal in early stages however with increasing size of the lesion; increasing soft tissue shadow may be noticed along with the evidence of enchondral ossification in the form of ring and arc calcification. It also helps to assess the arthritic changes of the joint, if any. Magnetic Resonance Imaging of the lesion shows typically the changes mentioned above.

Treatment of the condition remains surgical excision. Evaluation of the joint cavity can be done arthroscopically however the size of the lesion generally mandates open excision biopsy.

Malignant transformation of synovial chondromatosis to chondrosarcoma has been reported in approx 5% of the cases.¹⁴ However, the malignant transformation rate could not be found for giant SOC in the literature because of a rarity. Recurrence of giant SOC remains a possibility following surgical intervention.¹³

In the presented case, we were able to attain a good outcome. However, our follow-up on the case has been only 9 months duration. A longer period of follow-up is desirable to know the long-term outcome of surgical intervention and early detection of recurrence if any.

CONCLUSION

Giant synovial osteochondroma is a rare benign musculoskeletal neoplasm which can present itself with the variety of symptoms. Imaging is helpful in ascertaining the diagnosis. The treatment of the condition remains surgical wherein the joint cavity inspection can be done arthroscopically, but size generally mandates open excision. Post surgery, recurrence remains a possibility.

CLINICAL SIGNIFICANCE

Though a less common neoplasm, giant SOC should form one of the clinical diagnoses in swellings arising from synovial joints as early diagnosis of the condition

can prevent arthritic changes to progress further. In the presented case, there was a relatively short duration of the symptoms; with prompt intervention, we have been able to achieve good functional outcome without any arthritis.

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