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Case Report

Primary synovial osteochondromatosis of the ankle: A case report and review of the literature ☆

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ABSTRACT

Primary synovial osteochondromatosis of the ankle is a rare and benign disease of the young adult. It is characterized by the formation of multiple nodules of hyaline cartilage under the synovium. It is usually asymptomatic but can sometimes be revealed by pain or joint mobility disorders. X-rays and CT scans can easily diagnose calcified osteochondromas, but ultrasound and MRI can provide a quicker diagnosis by visualizing non-calcified foreign bodies. Surgery under arthroscopy remains the best therapeutic choice, however, this pathology still causes recurrence in less than 1/3 of cases.

Introduction

Primary synovial osteochondromatosis is a rare, benign condition characterized by the of synovial tissue and the formation of cartilaginous and osteocartilaginous bodies in the capsule. The cause of the metaplasia is unknown. It most commonly involves large joints, such as the knee, hip, and shoulder. Ankle involvement is unusual. We report a case of a young patient diagnosed as primary synovial osteochondromatosis of ankle. We will discuss the clinical data, the imaging findings, and the different therapeutic options and compare our results with the literature.

Case presentation

An 18-year-old man, with a history of a fracture of the phalanx of the big toe of the right foot treated orthopedically 3 months ago. He came to the ER with a 2-year progressive mechanical pain of the right ankle. The patient was hemodynamically sable. The musculoskeletal examination revealed a standing position without support, normal walking, and no deformity of the forefoot on inspection. On palpation, the ankle presented painful points opposite the 2 malleoli and the tibiotalar joint. The ankle's joint amplitudes were respected. The examination of the other peripheral joints and the spine was normal.

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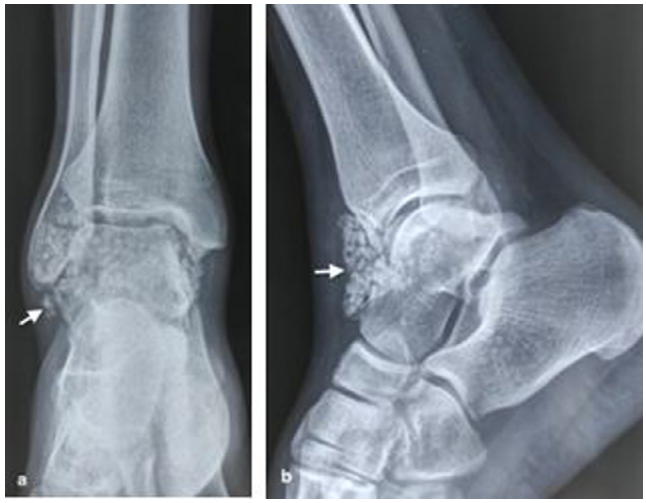

Fig. 1 – Front (A) and side (B) X-ray of the right ankle showing multiple peri-articular chondromas of different sizes.

A biological check-up was carried out and has shown no abnormalities. Initially, ankle radiographs were preformed (Fig. 1) showing periarticular calcified foreign bodies related to the chondromas of different sizes. Subsequently, an MRI was performed, confirming the presence of these chondromas developed in the anterior and lateral compartments of the tibiotalar joint (Fig. 2). There was associated an enhancement of the synovial and a moderate intra-articular effusion.

The patient underwent a surgical procedure of the ankle with extraction of the foreign bodies. Post-surgical radiograph showed a satisfying result (Fig. 3). The patient left the hospital after a short period of observation. Histologically, it was a synovial hyperplasia with a congestive chorion and minimal mononuclear element associated with ossified cartilaginous tissue realizing little cellular nodules without atypia compatible with osteochondromatosis. The post-operative follow-up was simple.

Discussion

Primary synovial chondromatosis also known as osteochondromatosis, chondrometaplasia, or synovial chondrosis is a mono-articular condition characterized by the sub-synovial formation of multiple nodules of hyaline cartilage [1,2]. It is a rare, benign but frequently recurrent disease [3]. Milgram distinguishes 3 phases in the osteochondromatosis cycle: Phase 1: intrasynovial disease without free foreign bodies. Phase 2: active synovial proliferation with free foreign bodies. Phase 3: multiple free osteochondromatous bodies without a synovial disease [4,5].

This pathology mainly affects the knee in more than half of the cases, then in decreasing order of frequency: the elbow, the hip, the shoulder, and the ankle [6,7]. Our young patient had a rare location which is the ankle.

This condition affects men slightly more than women and is most often discovered in adulthood between the ages of 20 and 50 [7].

It may be discovered incidentally on X-ray, but most often patients complain of mechanical pain, joint swelling, or limited mobility [6]. Our patient complained of progressive ankle pain which led to preform and X-rays examination.

Imaging provides an early diagnosis of osteochondromatosis from stage 2 (Milgram's stage), where the foreign bodies are free, and is only diagnosed in standard radiographs at the stage of calcified chondromas that appear in the para-articular area [4]. Ultrasound allows a very early diagnosis by visualizing non-ossified chondromas. They appear echogenic and mobile during compression [8]. In our case, the osteochondromas were calcified, which facilitated their visualization on the radiographs, this is why additional ultrasound was not performed.

CT scans without intra-articular contrast injection are of little use, but arthroscanning is still interesting in the case of free chondromas. MRI is often preferred to the previous imaging. It allows to detect cases of pure synovial chondromatosis by allowing to visualize foreign bodies and to specify their location. The appearance on MRI depends on the stage of the disease; chondromas generally appear in T1 hypo intensity.

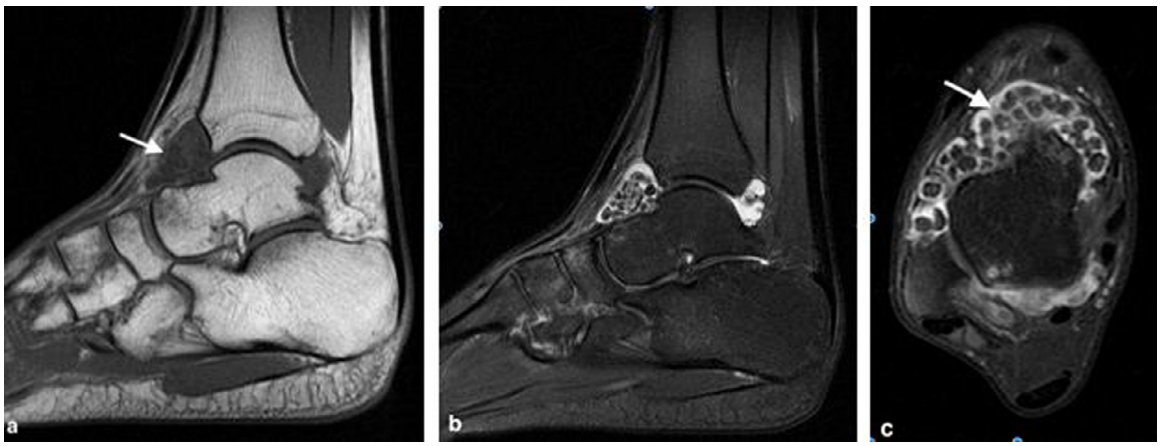

Fig. 2 – MRI of the right ankle showing intra-articular foreign bodies (white arrow) they appear hypointense on T1-weighted images (A), hypointense on T2 weighted images (B, C).

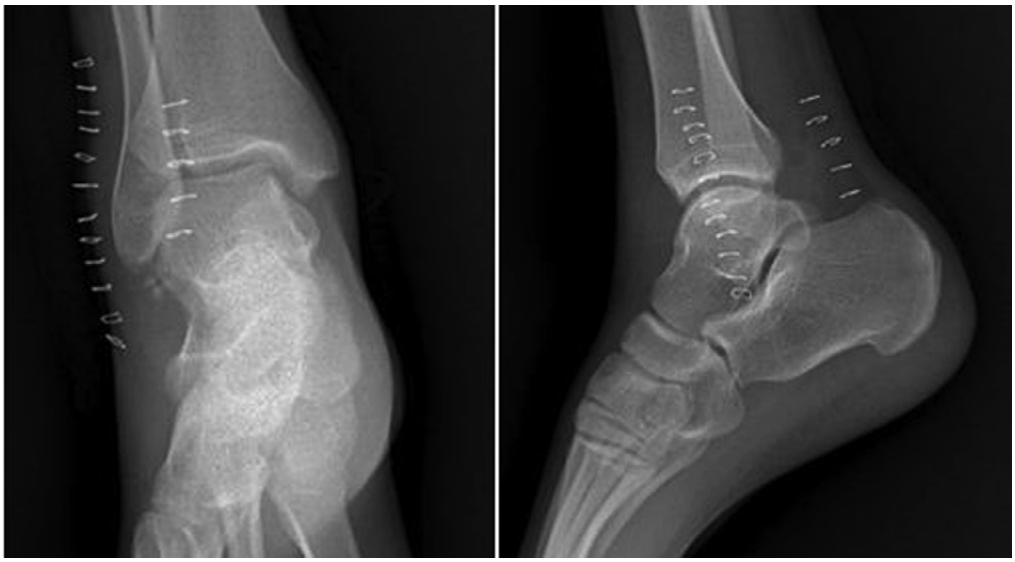

Fig. 3 – Front and side post-surgical radiograph of the right ankle after extraction of the foreign bodies.

In T2, the cartilage bodies are in hyper intensity. Injection of gadoline contrast medium results in signal enhancement of the inflammatory synovium, which may reveal the chondromas [2]. In our case, MRI allowed an accurate assessment of the location of foreign bodies, thus facilitating their surgical management. Direct arthro-MRI is rarely performed and offers the same results.

The treatment of choice for synovial chondromatosis is surgical resection. Synovectomy is indicated if active synovitis is present [1]. In the absence of symptomatology, therapeutic abstention is the rule. Arthroscopy has developed considerably in the surgical treatment of this condition because of its lower morbidity, but it does not allow treatment of the extra-articular extension of osteochondromas, hence the use of imaging [9].

The recurrence rate of this disease is still non-negligible up to 23% and would be related to an incomplete synovectomy.

Conclusion

Primary osteochondromatosis of the ankle is a rare condition of the young subject that can sometimes be disabling and require the least invasive surgical treatment. Imaging plays an important role in localizing the osteochondromas especially on MRI.

Patient consent

An informed consent was obtained from the patient.

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