CASE REPORT

Extraskeletal Osteochondroma of the Foot

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Extraskeletal osteochondroma, an infrequently encountered benign lesion, is described in 1 patient who presented with a slow-growing, relatively painful mass on the dorsal side of his right foot, in the 3rd toe web. Radiographs and magnetic resonance imaging studies demonstrated a radio-opaque extraskeletal mass. Excision biopsy revealed a well-defined and lobulated, firm, calcified, extraskeletal mass, which was easily enucleated. Histologic examination revealed mature chondroid tissues with bland-looking chondrocytes and ossified tissues. The diagnosis of an extraskeletal osteochondroma should be considered when a discrete ossified mass is localized in the soft tissues of the distal extremities. A case of pathologically proven extraskeletal osteochondroma is presented together with clinical, radiologic, and magnetic resonance imaging findings, and a literature review. [*J Chin Med Assoc* 2010;73(1):52–55]

Key Words: calcification, extraskeletal osteochondroma, magnetic resonance imaging, mature chondroid tissues

Introduction

Extraskeletal osteochondroma is a relatively rare, slow-growing, benign soft tissue tumor, commonly arising within the soft tissues of hands and feet, and presenting as a small discrete calcified mass, that rarely exceed 2–3 cm in its greatest dimension.[1,2] This tumor has no predilection for either gender, and it occurs mostly in patients 20 years or older with peak occurrence during the 3rd and 6th decades, and usually without antecedent trauma.[2] Because of its variable and worrisome radiological features and histological presentation, it is usually mistaken for an osteosarcoma.[1,2] A case of extraskeletal osteochondroma of the right foot measuring 5 cm in diameter is described along with its clinical and radiographic findings, and a review of the literature.

Case Report

A 62-year-old male patient presented at our clinic with a painful mass on the dorsal side of his right foot. The mass had grown gradually in size for the past 5 years, and was associated with numbness and pain during walking. Physical findings showed a firm, fixed and tender mass in the 3rd toe web, measuring approximately $5 \times 4 \times 4$ cm in size. Plain radiography of the right foot showed an ossified soft-tissue mass around the proximal phalanx of the 4th toe, with no significant joint or bony destruction noted (Figure 1). Magnetic resonance imaging (MRI) scans of the right foot with routine sequences, including T1- and T2-weighted images, revealed that the lesion and its adjacent soft tissue were moderately enhanced, with amorphous hypointense and non-enhanced ossified contents present (Figure 2). A total excision of the mass was performed. Intraoperative findings revealed a well-defined and lobulated firm mass with calcifications, which was easily enucleated. On the cut surface, the tumor was mainly composed of whitish cartilage with focal bony hard areas. Microscopically, most of the tumor displayed mature chondroid tissues (Figure 3) in which bland looking chondrocytes were seen. There were foci of ossifying tissue composed of mature lamellar bone (Figure 4), which were compatible with an extraskeletal osteochondroma. The patient was discharged on the 3rd postoperative day in a stable and healthy condition. He had regular follow-up visits over the following 18 months and there was no recurrence of the tumor. A complete chromosomal study of our patient was also performed, which showed no clonal alteration of chromosomes (Figure 5).

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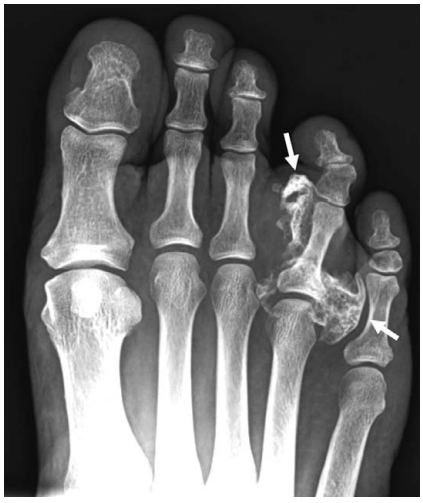

Figure 1. Anteroposterior radiograph of the right foot showing an ossified soft tissue mass at the proximal phalanx of the 4th toe.

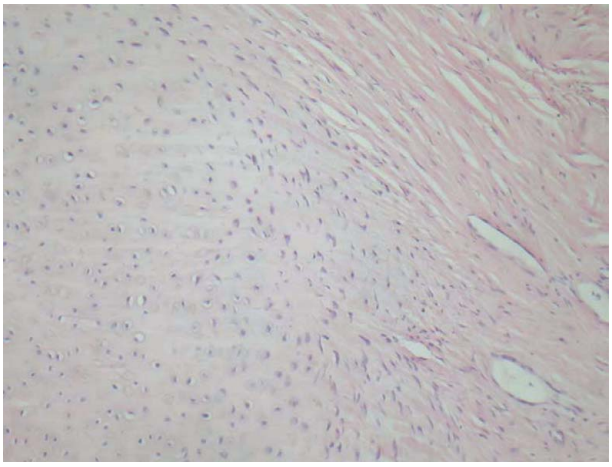

Figure 3. Histopathologic examination of the mass shows mature chondroid tissue with bland-looking chondrocytes (hematoxylin & eosin, 40×).

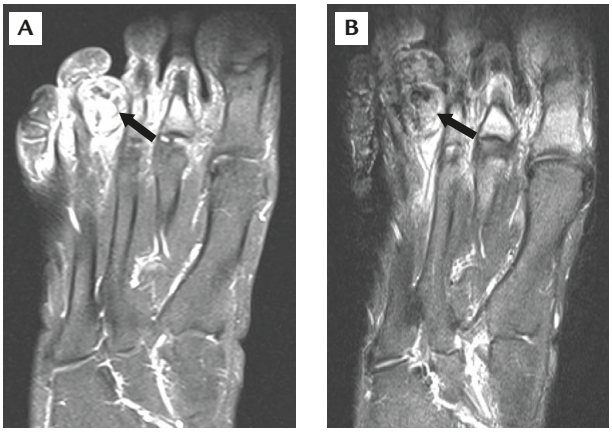

Figure 2. Magnetic resonance imaging demonstrates a well-encapsulated hypodense mass with amorphous ossification over the 3rd toe web with no bony joint involvement (T1- and T2-weighted images with contrast and fat suppression).

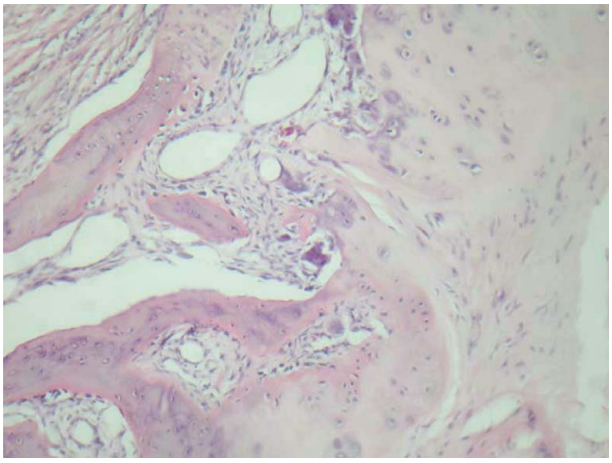

Figure 4. Microscopic examination of the same mass shows foci of ossified tissue with mature lamellar bone (hematoxylin & eosin, 400×).

Discussion

Osteochondroma is a common benign bone lesion representing approximately 20–50% of all benign bone tumors and 10–15% of all bone tumors.[3] These benign tumors are rare in soft tissues, and if found are without osseous or intra-articular involvement, thus their description as extraskeletal osteochondromas. There are other terms used to described this benign solitary cartilaginous mass in the extrasynovial tissue, including extraskeletal or soft tissue chondroma, extraosseous chondroma, tenosynovial chondroma, chondroma of soft parts, and cartilaginous tumor of the soft tissue.[4]

Extraskeletal osteochondroma is a slow-growing tumor with the highest incidence in the hands and feet. The foot is the second most common location after the fingers, where more than 80% of soft tissue chondromas are found.[5] Sporadic cases have been reported in the buttocks, thighs, knees, pharynx, oral cavity, parotid gland, skin, and the nape of the neck.[6–10] The precise cause of extraskeletal osteochondroma is still unknown, with 1 theory that it originates from pluripotent cell lines derived from the joint synovium, tenosynovium, or connective tissue,[1,11] while other theories postulate that it originates from metaplasia of tendon sheaths in the hands and feet.[1,11] Another theory is that extraskeletal osteochondroma arises from fibroblasts in the connective tissue distant from bone and joints because of unknown stimuli.[9] Recently, nonrandom clonal alterations of chromosomes 6, 11, and 12 have been reported in soft tissue chondromas, including supernumerary ring chromosomes containing

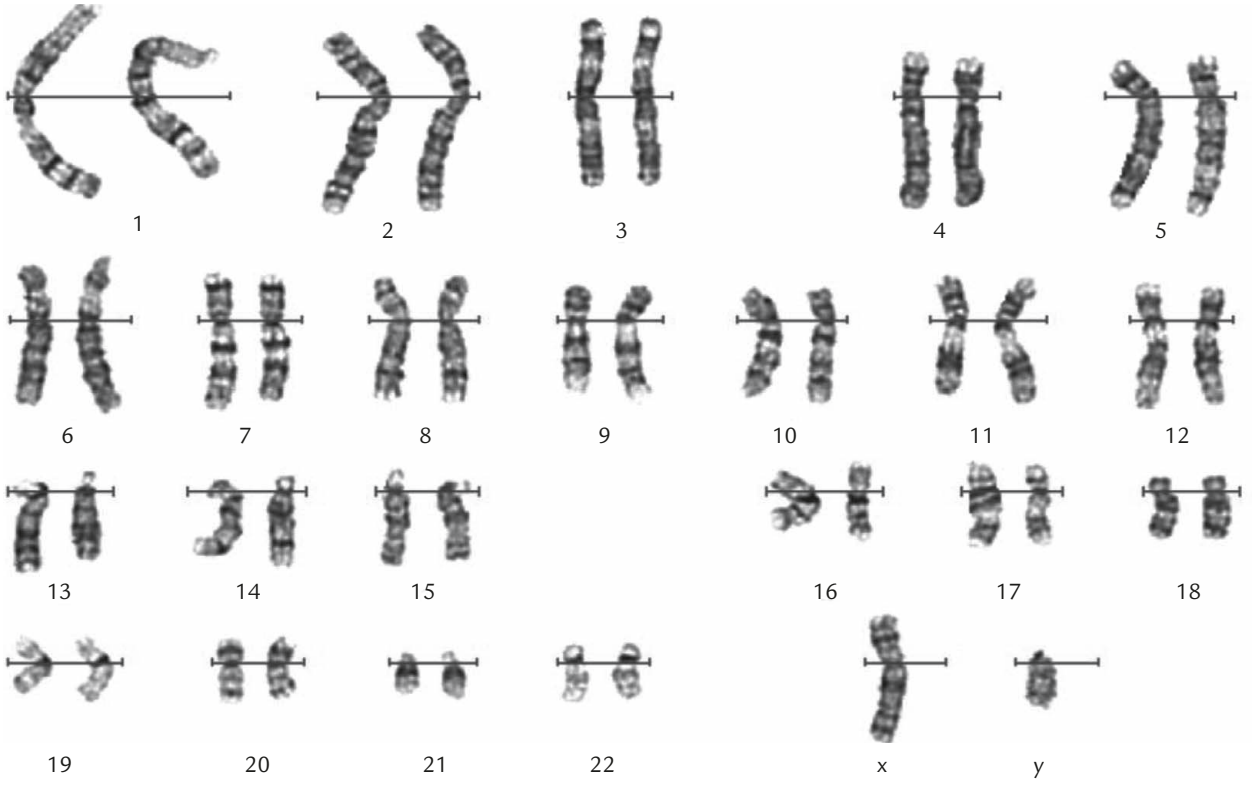

Figure 5. Chromosomal report reveals no chromosomal abnormalities.

material from chromosome 12.[12] In another study, molecular analysis showed that the so-called *HMGA2* gene located at 12q15 appears to be involved in soft tissue chondromas and other cartilaginous tumors and also in lipomas.[13] However, in the chromosomal study performed on our patient, no clonal alterations of chromosomes were noted, therefore clonal alternations of chromosomes might not be routinely identified in patients with soft tissue chondromas. There are reports[10] describing cellular atypia, although no malignant transformation or metastatic lesions have been demonstrated.

Clinically, extraskeletal osteochondroma presents as a painless, slowly enlarging nodular soft tissue mass that is usually present for a variable time before diagnosis. However, in 20% of patients, the lesion is painful and tender, especially if located on the plantar side of the foot.[14] Bilateral extraskeletal osteochondromas have been reported but the majority of extraskeletal osteochondromas are solitary. Multiple lesions more likely to be synovial chondromatosis.[15] Current recommended treatment for extraskeletal osteochondroma consists of total excision with histopathological confirmation and close follow-up for recurrence. Local recurrence is rare with a reported rate of 18%, and is best treated with re-excision.[1,2]

Chung and Enzinger reported 56 cases in which follow-up was available for 1 year or more. Ten patients reported recurrences after initial excision.[1] In the 54 cases of Dahlin and Salvador, there was recurrence in 8 patients during an observation period of 1–10 years.[2] Therefore, recurrence is a potential problem.

Radiologically, an extraskeletal osteochondroma presents as a well-defined soft tissue mass, often with central calcification or areas of ossification that are not attached to the periosteum or cortex of a bone and are not found within the confines of a joint capsule or tendon sheath. Computed tomography demonstrates the extraskeletal location and central dense area of calcification or ossification of an extraskeletal osteochondroma.[7] On MRI, the mass appears as an intermediate signal intensity on T1-weighted images. On T2-weighted images, it appears as a high signal intensity lesion as seen in areas of calcification. MRI is best for delineating the border of the mass, thus distinguishing it from a sarcoma.[11]

The main differential diagnoses, such as periosteal chondroma, myositis ossificans, lipomatous lesion,

pseudomalignant osseous tumor, ossifying fibromyxoid tumor, soft-tissue osteochondroma, tumoral calcinosis, synovial chondromatosis, synovial sarcoma, soft tissue chondrosarcomas and extraskeletal osteosarcoma, should be reserved for discrete soft tissue masses with mature ossification. The clinicopathological and radiological features of the present case excluded the possibility of these lesions. Although mature ossification is usually associated with a benign lesion, sarcomatous lesions such as synovial sarcomas, soft tissue chondrosarcomas and osteosarcomas can also present with dense calcification and ossification.[11] Thus, close histopathological and clinical correlation are necessary to differentiate a chondrosarcoma from an extraskeletal osteochondroma.

In conclusion, an extraskeletal osteochondroma should be considered when a discrete, ossified mass is localized in the soft tissues of the distal extremities. Although cellular atypia are described on histologic examination, there is no known malignant transformation or metastatic lesion. Marginal excision is the recommended treatment of choice, with preservation of the adjacent bone and soft tissue structures. A local recurrence rate of up to 18% has been reported, and such recurrence is best treated with re-excision.

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