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Extraskeletal Osteochondroma of the Great Toe in a Teenager

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ABSTRACT

Osteochondromas are common, benign surface tumors of bone, composed of the cartilage-capped bone confluent with the medullary canal of the metaphyseal bone. Extraskeletal osteochondromas have the same gross appearance and histologic characteristics as a typical osteochondroma but do not have any boney attachment to the surrounding osseous structures. They are rare and most frequently reported in the middle-age and older adults. We present the first case of an extraskeletal osteochondroma of the foot reported in a teenager. Our patient was a 17-year-old male complaining of a slow-growing mass along the medial border of the great toe that he first noted at the age of 14 years. The increasing size of the mass and frequency of complaints with shoe wear prompted medical attention. Imaging studies showed an ossified 1-cm boney mass with trabecular detail, located on the medial aspect of the great toe at the level of the interphalangeal joint, without any connection to the surrounding structures. An excisional biopsy revealed a well-circumscribed, easily removable mass, which proved to be an extraskeletal osteochondroma both clinically and histologically.

Osteochondroma is the most common bone tumor, comprising 10% to 15% of all boney tumors and nearly 20% to 50% of all benign bone tumors (1). Structurally, they consist of a boney outcropping from the cortex of the native bone, confluent with the medullary canal and with a cartilage cap of varying depth that is histologically arranged like the physeal cartilage. For typical osteochondromas, the proposed source of the cells is from the perichondrial ring of LaCroix at the growth plate (2). For extraskeletal osteochondromas, the etiology is less well understood.

Although soft tissue osteochondromas, also known as extraskeletal osteochondromas, have the same gross and histologic structure as typical osteochondromas, they do not have any boney attachment to the surrounding osseous structures. These rare soft tissue neoplasms usually arise in the absence of trauma, adjacent to synovial joints or tendon sheaths. In addition, unlike typical osteochondromas, whether they are solitary or those associated with multiple hereditary exostoses, soft tissue osteochondromas typically occur in adults between the third and sixth decades of life. Soft tissue osteochondromas are slow growing and asymptomatic; however, they can become bothersome owing to the mechanical compression of the surrounding structures (3,4). These lesions may be mistaken for a malignancy because of the de novo appearance in adulthood, with a tendency to enlarge.

We present the second reported pediatric case of an extraskeletal osteochondroma of the foot. This is the first instance of this diagnosis made in a teenager—unique in the rarity of the diagnosis, as well as in the location of the mass and the age of the patient.

Case Report

A 17-year-old male presented to our center with a slow-growing, well-circumscribed mass of 3-year duration, located on the medial side of the right great toe at the level of the interphalangeal joint (IP). The patient denied any history of trauma. Initially, the toe mass caused occasional discomfort, but for the last 12 months, the mass was increasing in size and became more bothersome with shoe wear. The family sought a second opinion because a prior physician raised the specter of malignancy.

The patient had no personal or family history of tumors, metabolic conditions, or rheumatologic conditions, and he had no symptoms beyond his localized toe discomfort. Physical examination revealed a hard, painless, and well-circumscribed 1-cm mass along the medial and slightly plantar aspect of the great toe at the level of the IP joint (Fig. 1). The mass was firm and somewhat mobile, deep to the subcutaneous tissue, and not fixed to the underlying structures. The skin was slightly thickened but free from the underlying mass.

Plain radiographs showed a well-delineated, ossified 1-cm mass, with trabecular detail located on the medial aspect of the great toe at

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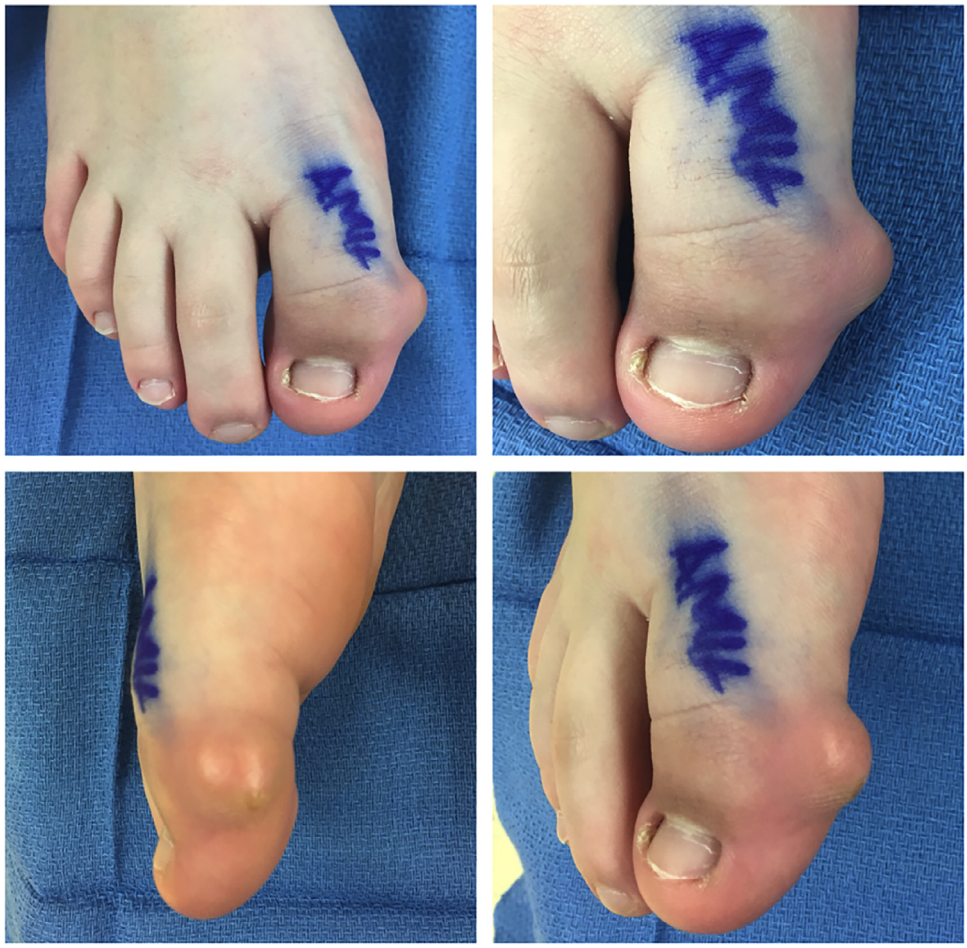

Fig. 1. Gross examination of well-circumscribed 1-cm mass of the great toe at the interphalangeal joint.

the level of the IP joint, without any connection to the surrounding structures (Figs. 2 and 3). There was no scalloping or cortical destruction of the adjacent distal or proximal phalanx or any other imaging characteristics that would suggest an aggressive change or boney destruction.

After discussion of the risks, benefits, and alternatives, the patient was taken to the operating room for excisional biopsy of the mass. A 2-cm incision was made along the medial boarder of the great toe, overlying the protruding mass. Just below the level of the skin, there was a hard, gray-pink spherical mass with smooth margins and a shiny surface (Fig. 4). The mass was not attached to any underlying structures, such as tendons and the nearby IP joint. It was not confluent with bone and instead encapsulated within the soft tissue only. The mass was removed without difficulty, and the skin was sutured with 3-0 nylon sutures with a soft overlying dressing. He was made weightbearing, as tolerated. Postoperative radiographs confirmed full excision of the mass (Fig. 5).

On histopathologic examination, multiple cut sections of the specimen were described as a benign osteochondroma with a core of cortical bone (Figs. 6 and 7). The boney mass had a thin, mildly nodular, and relatively thin cartilage cap with columns of chondrocytes described as

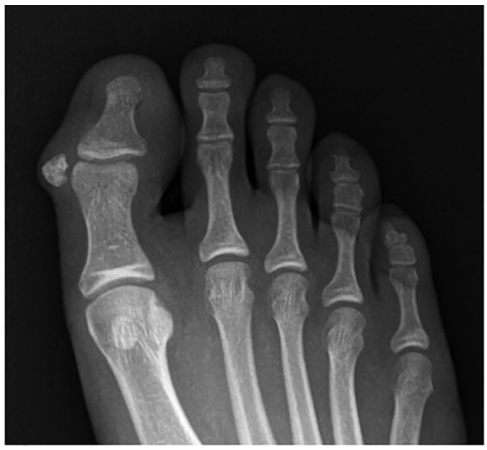

Fig. 2. Magnified view of plain radiograph (anteroposterior view) showing calcified mass of the great toe.

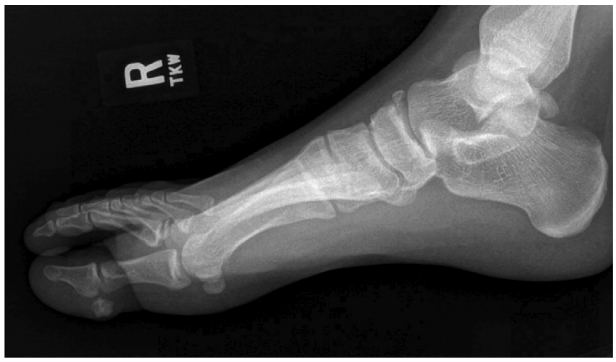

Fig. 3. Plain radiograph (lateral view) showing calcified mass of the great toe.

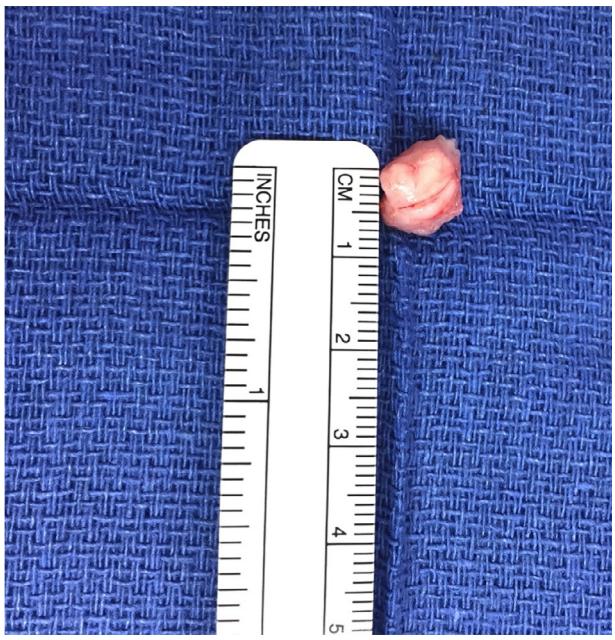

Fig. 4. Excised smooth, shiny well-circumscribed 1-cm boney mass.

disorganized but without an evidence of atypical changes. Endochondral ossification was present at the base of the cartilage cap. The final diagnosis of extraskeletal osteochondroma was confirmed.

Postoperative recovery was uneventful and without complication. The patient's symptoms resolved without evidence of recurrence at his 2-week, 3-month, and 1-year postoperative follow-up appointments.

Discussion

Osteochondroma is a benign boney tumor with a cartilage cap that has a histologic appearance similar to the growth cartilage of the physis. It is believed to arise from the perichondral ring and typically develops in the metaphyses adjacent to the growth plate. The tumor may grow up in size until the closure of the growth plate, at which point it ceases to enlarge (3). Renewed growth of an osteochondroma in adulthood may herald a malignant transformation into a chondrosarcoma. Although, unlike typical osteochondromas, there are no reports of late malignant transformation of an extraskeletal osteochondroma, local recurrence rates have been reported as high as 18% following local excision (4).

A soft tissue or extraskeletal osteochondroma is made of mature bone with cartilage cap; however, there is no connection to the underlying boney structures. Most often, it occurs between 30 and 60 years of age (4,5). Reports of these soft tissue neoplasms are detailed through case reports, most often arising in the hands (70%) and feet (25%); however, less frequent sites of the thigh, hip, knee, buttock, neck, and wrist have also been reported (6–11).

Case reports of an extraskeletal osteochondroma in the foot are reported in great variety and in different locations, including the soft tissue beneath the calcaneus (12–14), near the ankle (14), the plantar arch (15–17), within the toe web space (18), and plantar to the metatarsals of the foot (19–20). A single case was reported where the mass caused a secondary stress fracture owing to an unusually large extraskeletal osteochondroma between the fourth and fifth metatarsals (20). All of these reports have occurred in adults; however, a single congenital pediatric case detailed an obvious third web toe mass present since birth, enlarging in size since the age of 3 years and ultimately excised in a 9-year-old male owing to mechanical symptoms (21). However, unlike this, our pediatric patient identified a de novo mass of the foot later in life, at the age of 14 years, ultimately prompting excision at the age of 17 years.

Osteochondromas of the great toe have been reported in the form of subungal exostosis after trauma or subungal osteochondromas—a recognized variant of osteochondroma. Unlike extraskeletal osteochondromas, subungal osteochondromas are continuous with the underlying boney structures, affect the nail bed, and are often preceded by trauma, making their diagnosis common in the great toe, unlike our patient (21,22).

Clinically, extraskeletal osteochondroma presents as a slow-growing, nonpainful mass. The initial differential diagnosis may include osteochondroma, subungal osteochondroma, extraskeletal osteosarcoma, synovial chondromatosis, myositis ossificans, synovial cell sarcoma, and tumoral calcinosis (7). On imaging, an extraskeletal osteochondroma presents as a lobulated, well-demarcated osseous mass not congruent with the nearby bone (13). The histologic examination is characterized by mature hyaline cartilage encasing around both lamellar and trabecular bones, sometimes with a central area of necrosis (15). The clinical, radiologic, and histologic features of the present case excluded the possibility of the lesions listed in the differential diagnosis.

Although the pathogenesis of extraskeletal osteochondroma remains unknown, several theories have been proposed. Three different theories of lesion formation were presented by Lynn and Lee (23): 1) cells capable of cartilage production migrate from the nearby bone and deposit into the soft tissue, where they proliferate; 2) precartilaginous tissue within the tendon activates from a dormant stage and begins proliferation; and 3) unknown factors stimulate synovial cells to undergo

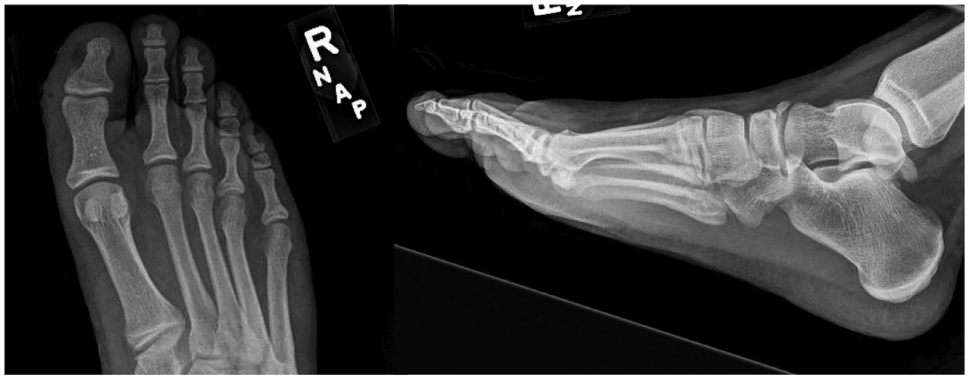

Fig. 5. Magnified view of plain radiographs (anteroposterior and lateral views) showing full excision of the calcified mass of the great toe.

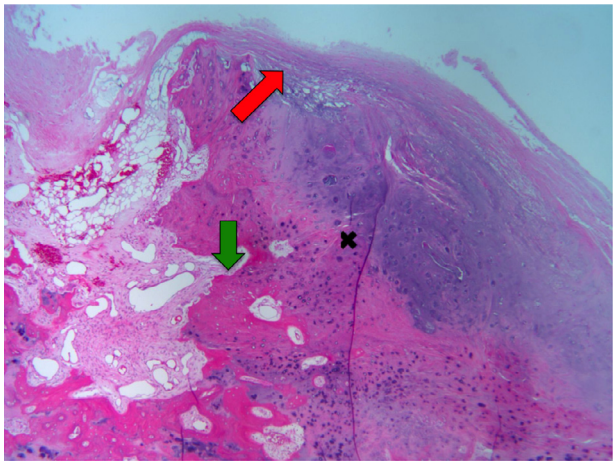

Fig. 6. Photomicrograph shows mature hyaline cartilage (×), fibrous capsule at the periphery (red arrow), and bone/endochondral ossification (green arrow) (magnification × 10; hematoxylin and eosin stain).

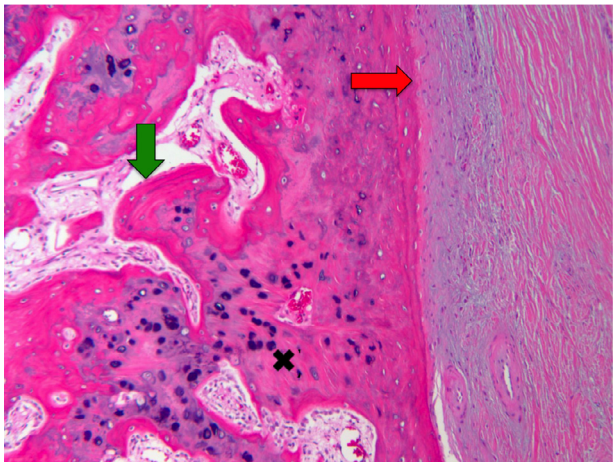

Fig. 7. Photomicrograph shows mature hyaline cartilage (×), fibrous capsule at the periphery (red arrow), and bone/endochondral ossification (green arrow) (magnification × 100; hematoxylin and eosin stain).

metaplasia to produce cartilage. The consistent agreement in the current literature is that trauma does not provoke the formation of extraskeletal osteochondromas (23); however, no consensus has been reached on the causation or true origins of their development.

In conclusion, a unique case of extraskeletal osteochondroma of the great toe in a teenager is presented here. It is the first case reported in a patient of this age and in this specific location. Extraskeletal osteochondroma should be considered with the identification of a slow-growing, benign mass located in the soft tissue near a bone or joint. Extraskeletal osteochondromas should be identified and monitored, with surgical excision indicated should symptoms present. Although there is no evidence to suggest malignant transformation, a local recurrence rate of up to 18% has been reported, and it is best treated by re-excision (3).

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