Giant Solitary Osteochondroma of the Inferior Medial Calcaneal Tubercle: A Case Report and Review of the Literature

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Small osteophytes are frequently encountered in the foot and ankle, and are not to be confused with true osteochondromas, which are relatively uncommon in this region. They most often affect long bones of the appendicular skeleton but may involve flat bones as well. Osteochondromas are benign osseous neoplasms with a distinct hyaline cartilage cap originating from the physis and cease growing with skeletal maturity. Osteochondroma are often treated conservatively unless they become symptomatic, painful, demonstrate rapid or new growth, enlarge after skeletal maturity, and/or exhibit signs of malignant transformation. In this report, we present a case of a giant (8 cm \times 4.2 cm \times 2.1 cm) osteochondroma in an adult occurring on the inferior medial tubercle of the calcaneus that underwent excision, with 3.5 years of follow-up without recurrence. To our knowledge this is the largest osteochondroma affecting the inferior medial tubercle of the calcaneus. This case demonstrates that large osteochondromas may occur in the foot, and also confirms that benign osteochondroma growth may occur in adulthood. A detailed review of osteochondroma occurrence in the foot is presented along with a review of the diagnostic work-up to evaluate for malignant transformation. Level of Clinical Evidence: 4 (The Journal of Foot & Ankle Surgery 47(3):206–212, 2008)

Key Words: osteochondroma, chondrosarcoma, calcaneus, bone tumor

True osteochondromas are uncommon in the foot and ankle. They are benign osseous neoplasms that typically extend from the metaphyseal or metadiaphyseal region of long bones of the appendicular skeleton and are most commonly found around the knee (1). They mostly occur solitarily or in rare situations they may occur as multiple lesions within a hereditary disorder (multiple hereditary exostoses [MHE]) (2). When they occur in the foot and ankle, they tend to be small innocuous lesions that are typically managed conservatively. They have a distinctive radiographic appearance as an exophytic osseous lesion that grows perpendicular to its parent bone. They also have a distinctive

hyaline cartilaginous cap, which may rarely undergo malignant transformation.

Osteochondroma is the most common benign tumor of bone, representing approximately 36% to 41% of the benign bone tumors (3). It is estimated that osteochondroma affects 2% to 3% of the general population (4–6). They grow during childhood into adolescence, at a rate mirroring the growth of the normal skeleton and cease growing with skeletal maturity (7). They are typically identified in patients younger than 20 and extensive osteochondroma growth into adulthood is rarely reported (8–10). In many cases, the osteochondroma itself is nonpainful and becomes symptomatic for a variety of reasons, including fracture, bursae formation, pain, neurologic compromise, interference with joint motion, joint malformation or malalignment, vascular compromise, and/or malignant transformation (11–13).

In the foot and ankle, osteochondromas are typically identified earlier than other regions because of the low proportion of subcutaneous tissue in the region, which may make an osseous mass more noticeable and symptomatic. We present a single case of a giant osteochondroma in an uncommon location occurring in the foot arising from the inferior medial calcaneal tubercle. The purpose of this manuscript is to illustrate that large benign osteochondroma may occur in the foot, and to provide a detailed review of the diagnostic work-up to evaluate for malignant transformation.

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FIGURE 1 Preoperative photograph of bilateral heels. The left heel is enlarged due the underlying calcaneal osteochondroma.

Case Report

A 40-year-old female presented with a 6-month history of a painful bulging mass in the area of her left heel. She described the pain as achy and has not noted any gradual increase in size since she first identified the enlargement. She complained of difficulty standing at her job in a delicatessen. The only treatment involved self-prescribed ibuprofen, which alleviated some of the discomfort. Past medical history was unremarkable. Past surgical history involved only wisdom tooth extraction. She related a smoking history of 1.5 packs per day for 20 years.

Physical exam demonstrated a firm nodular mass in the region of her plantarmedial rearfoot (Figure 1). The skin was supple without any overlying lesions or pigmentation. The posterior tibial and deep peroneal pulses were present and normal in character. The digits demonstrated instant capillary refill. The heel was painful with deep palpation and there was full supple range of motion of the ankle joint and rearfoot joints.

Radiographs revealed a large exophytic sessile osseous lesion involving the weight-bearing portion of the inferior and medial aspect of the heel (Figure 2). The mass measured 4.3 cm in greatest dimension and extended into the porta pedis. Its margins were smooth and did not appear to affect the structure of the calcaneus. There were no soft tissue calcifications or cortical erosions. Magnetic resonance imaging (MRI) was obtained, involving axial and sagittal and coronal T-1—weighted images, pre- and post-gadolinium sagittal and coronal T-2—weighted sequences with fat saturation. A large pedunculated bone lesion involving the inferior medial calcaneal tubercle was easily identified, measuring 8 cm × 4.2 cm × 2.1 cm. It was well demarcated with a distinct cortical rim and a variable thickness cartilage cap (measuring 0.2 cm to 1.8 cm). There were no destruc-

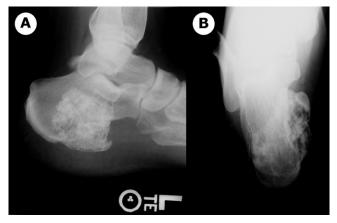


FIGURE 2 (A) Lateral preoperative weight-bearing radiograph of left rearfoot demonstrating a large exophytic osseous mass. (B) Axial radiograph reveals that the osseous growth is located medially

tive changes of the body of the calcaneus. Isointense bone with scattered areas of hypointensity images were visualized on the T1-weighted images. On T2-weighted images there were multiple area of increased signal (Figure 3). With gadolinium, heterogeneous enhancement was seen throughout the lesion. The MRI findings could not exclude sarcomatous change.

Based on the radiographic and MR imaging, a secondary chondrosarcoma could not be excluded. Further work-up included a total-body technetium 99 bone scan and a limited lower extremity bone scan (Figure 4). Remarkable findings were mild focal uptake at the plantar medial tubercle of the calcaneus correlating with the lesion and mild uptake at the left sternoclavicular joint associated with degenerative joint disease. Chest x-ray was normal without any pulmonary lesions and a chest computerized tomogram (CT) did not identify any pulmonary nodules or metastatic lung disease. A CT of the foot was not ordered as it would not have offered any additional information that would direct care.

The patient underwent incisional biopsy and the area of the cartilage cap was targeted for microscopic evaluation. Intraoperative fluoroscopy was correlated with the MR imaging to sample the cartilage cap. This biopsy revealed both hyaline cartilage and lamellar bone; however, classic endochondral ossification was not seen. A thickened cartilage cap with low to moderate cellular hyaline cartilage was present. Loosely grouped chondrocytes were present without any significant atypia or definitive myxoid change. Fragments suggestive of necrosis with loss of nuclei were identified. Necrotic chondroid matrix was present beneath the cartilage cap. Because of the heterogeneity of the specimen, sarcomatous degeneration could not be excluded.

Approximately 2 months after the initial biopsy, the patient underwent complete excision of the mass. A more immediate removal was not necessary to exclude sarcoma-

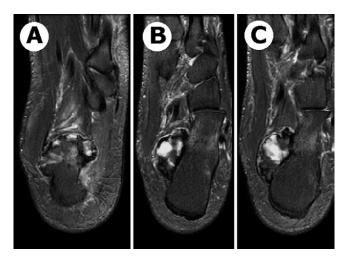


FIGURE 3 Preoperative transverse plane T2-weighted magnetic resonance imaging demonstrating the large osseous mass originates from the plantar medial calcaneal tubercle and extends medially along the medial calcaneal wall into the porta pedis. The mass is cauliflower-like and somewhat pedunculated. Scattered areas of lobulated locules of increased signal intensity are seen illustrating the cartilage component are seen in images A, B, and C. (A) There is some soft tissue edema of the displaced plantar musculature. (B, C) The bone lesion is not invasive to the medial cortical wall of the calcaneus.

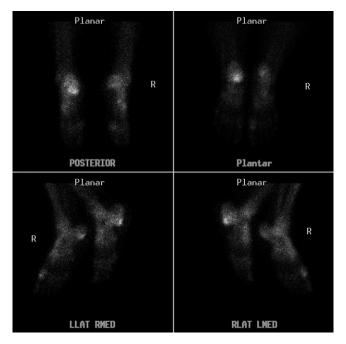


FIGURE 4 Limited technetium bone scan demonstrating mild focal uptake (*arrows*) at the inferior medial calcaneal tuberosity corresponding with a portion of the large osseous lesion identified on radiographs.

tous degeneration because the biopsy did not demonstrate any mitotic figures, a feature present in an aggressive lesion. Moreover, the radiographic findings and advanced imaging demonstrated findings with that of a stable benign lesion, as there was not any surrounding destruction. While chondro-

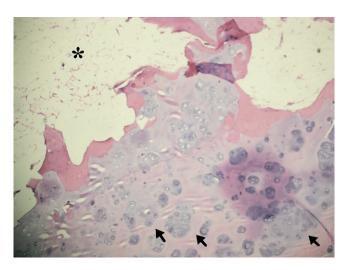


FIGURE 5 Osteochondroma photomicrograph (magnification ×4; hematoxylin and eosin stain) identifies the marrow space (*asterisk*) and hyaline cartilage cap with columns of chondrocytes (*arrows*).

sarcoma was in the differential diagnosis, it was considered to be unlikely.

The giant osteochondroma was accessed through a medial incision. The neurovascular bundle was located directly over the lesion, and gently retracted and protected. The plantar fascia was detached from its insertion on the heel. Because osteochondroma originated from the inferior medial tubercle of the heel, it was necessary to detach the plantar fascia to facilitate the removal of this giant mass. Because of the size of the osteochondroma and location (which precluded wide expose), the mass was removed in several large pieces. The cartilage cap was clearly identified for pathologist review. Intraoperative fluoroscan was used to confirm appropriate removal. Closure was performed in a standard fashion, subcutaneous with absorbable sutures and skin with interrupted nonabsorbable sutures.

Microscopic evaluation of the entire lesion illustrated increased degeneration with aggregates of amorphous calcified debris and necrotic cartilage (Figure 5). The hyaline cartilage showed occasional binucleate chondrocytes. The cartilage merges with bone trabeculae with intervening fatty marrow space. Because there were significant degenerative changes that alter the architecture, it is more difficult to determine the extent of the cartilage cap and radiographic correlation was important. A final diagnosis of osteochondroma with degenerative changes was made. The pathology was reviewed and the diagnosis confirmed via consultation with the Division of Anatomic Pathology at the Mayo Clinic, Rochester, MN.

The postoperative course was uneventful. The patient was non-weight bearing for 4 weeks then increased activities accordingly. Follow-up radiographs at 3.5 years demonstrated resorption of residual ossicles remaining in the soft tissue at the time of surgery and bone remodeling of the



FIGURE 6 Postoperative lateral weight-bearing radiograph of the left foot approximately 3.5 years after the giant osteochondroma was excised. There is some flattening of the arch via a mild naviculocuneiform sag, which may be due to loss of the plantar fascia windlass effect on the arch.

resection site (Figure 6). The last follow-up visit of 3.5 years postexcision did not reveal any recurrence of the lesion. The patient had difficulty standing long periods and was unable to return to her previous line of employment and eventually declared permanent disability.

Discussion

Osteochondromas are common benign tumors of bone that typically do not affect the foot. Large or giant osteochondromas may be concerning in this region and may prompt removal because they may interfere with function. Additionally, a large osteochondroma may be suspicious for malignant transformation based purely on size alone. A better understanding of osteochondroma as it relates to the foot will better guide the foot and ankle surgeon when presented with these bone tumors.

The diagnostic work-up for osteochondroma is straightforward. Radiographs are often diagnostic alone. However, other imaging modalities, such as CT, MRI, or bone scanning, may be necessary for surgical planning and/or to exclude sarcomatous degeneration. Radiographically, they appear as sessile or pedunculated osseous growths in continuity with the underlying cortex and medullary cavity of the parent bone (14). In long bones, they appear to emanate from the metaphyseal region and have a cauliflower-like appearance. Pedunculated osteochondromas tend to grow away from the nearest joint (15). Sessile lesions may be radiographically similar to Olliers disease, myositis ossificans, parosteal osteosarcoma, and juxtacortical chondrosarcoma, which may warrant further imaging and investigation (16, 17). Radiographic features of malignant transformation of osteochondroma are listed in Table 1.

Malignant transformation is rarely known to occur with osteochondroma, and is estimated to occur in less than 1% to 2% of cases of solitary osteochondroma (11, 15, 18). Patients with MHE are at a greater risk for malignant

TABLE 1 Radiographic features of a peripheral chondrosarcoma

Thickened cartilage cap > 2 cm
Scattered calcified foci in the cartilaginous portion, particularly in the adjacent soft tissue
Irregular osteochondral interface
Calcified or noncalcified soft tissue mass
Focal areas of radiolucency within the interior of an osteochondroma
Destruction or pressure erosion on an adjacent bone

Adapted from Lee KC, Davies AM, Cassar-Pullicino VN. Imaging the complications of osteochondromas. Clin Radiol 57(1):18–28, 2002.

degeneration, which has been suggested to occur in 5% to 25% of cases (4, 19–21). Clinical features suspicious for malignant degeneration include new onset of pain in a previously stable lesion, rapid or new growth, growth beyond skeletal maturity, and/or large lesions (1, 22). Pain and enlargement prior to skeletal maturity "seldom indicates malignancy" as the tumor growth parallels that of the physis (10, 13). Lesions greater than 8 mm and/or a cartilage cap greater than 1 cm should undergo histopathologic evaluation. Cartilage cap thickness greater than 1 to 2 cm in adults and 2 to 3 cm in growing children suggests malignant transformation (11, 13, 15, 21).

The diagnostic work-up for malignant degeneration typically involves multiple imaging studies. Osteochondromas may be visualized with nuclear imaging as focal uptake of radionucleotide adjacent to the growth plate, especially in skeletally immature individuals. Longstanding stable lesions in adults may not demonstrate any uptake. A total body bone scan may detect asymptomatic deeply seated lesions. Nuclear imaging has not been useful for identifying malignant degeneration. Hudson et al (23) determined that increased uptake may be seen with malignancy; however, a normal study may be seen with malignancy as well. Hendel et al (24) reviewed 22 cases of osteochondroma with sarcomatous transformation and were unable to use bone scinitigraphy to differentiate between the 2 lesions. Rather, the specific utility of nuclear imaging is to simply identify deeply seated lesions that would otherwise go undetected, which is most important with patients with MHE who are at greater risk for malignant degeneration.

MRI is paramount in the work-up of symptomatic or suspicious osteochondroma because the extent of the lesion as well as any soft tissue involvement may be visualized as well as cartilage cap depth and location may be determined.

Continuity of an osteochondroma with its parent bone may be identified. The cartilage cap demonstrates signal characteristics typical of hyaline cartilage: high signal intensity on T2-weighted images and low to intermediate signal intensity on T1-weighted images (13). MRI may also detect an adjacent associated traumatic tenosynovitis or tendon rupture. Increased T2 signal within the soft tissue

surrounding may be seen as a nonspecific finding, especially if the osteochondroma is located in an area of increased mechanical irritation. Bursa formation is a known occurrence and may be confused with a large cartilage cap with increased water content (5, 25); however, a benign bursa simulating a large cartilage cap will demonstrate rim enhancement with gadolinium (25). A thick cartilage cap with "lobulated locules of increased signal intensity on T2weighted images" may be suggestive of malignant transformation (11). Lesions with a thick cartilage component are suspicious to have undergone malignant transformation (21). In contrast, CT does not accurately allow for cartilage cap measurement with cartilage cap thickness less than 2.5 cm (26). Fast contrast-enhanced MR imaging may "assist in differentiation between benign and malignant cartilaginous tumors" (27, 28).

Chondrosarcoma is the most common malignant neoplasm arising from osteochondroma (29, 30). In a literature review, Willms et al (31) identified 50 cases of osteochondroma malignancy with 94% developing chondrosarcoma. Other reported malignancies arising within an osteochondroma represent less than 10% of such malignant transformations; they include osteosarcoma, fibrosarcoma, and spindle-cell sarcoma (31, 32). Simon and Springfield (15) and Mirra (17) suggest that the incidence of malignant degeneration into chondrosarcoma is directly proportional to cartilage volume in the underlying preexisting benign lesion.

Osteochondromas of the extremities demonstrate a low rate of malignant transformation because these lesions become symptomatic necessitating investigation or excision long before malignancy may develop. Malignancies arising within osteochondroma have a propensity for the pelvis, most likely because they are deeply seated and grow unnoticed before being identified (15, 30). Nonetheless, any bone with a true osteochondroma may be at risk. Willms et al (31) included 1 case of a calcaneal osteochondroma malignant transformation in a literature review of 50 cases. Malik et al (22) reported on a solitary osteochondroma of the calcaneus transforming into a chondrosarcoma. In a review of 75 cases of chondrosarcoma secondary to osteochondroma, Garrison et al (29) identified 1 calcaneal chondrosarcoma in a patient with MHE.

Malignant transformation of osteochondroma into chondrosarcoma may be a well-differentiated low-grade tumor that is difficult to detect histopathologically. The structure and architecture of the cartilage cap is disrupted with malignancy, and may be seen as intermixed thin fibrous septa resulting in lobulation. In some cases, the diagnosis is based on correlating the histopathologic findings with the clinical history, and radiographic and advanced imaging findings.

Surgical removal of osteochondroma should be considered on a case-by-case basis accounting for the age of the patient, skeletal maturity, presentation and duration, pain

and/or associated symptoms, size of the lesion and cartilage cap, location, and the presence of MHE. Excision is indicated if the lesion is symptomatic, and may be considered even with mild symptoms. Small stable asymptomatic lesions should be treated conservatively. It is recommended that asymptomatic lesions be evaluated every 2 years during growth and every 3 to 5 years thereafter or when symptoms arise (27, 31, 32). Surgical of excision of small superficial osteochondroma are usually straightforward. Larger lesions may require more preoperative planning, especially when they are deeply situated, adjacent to neurovascular structures, impinging on a nearby joint, and/or suspicious for malignant transformation. Suspicious lesions for malignant transformation should undergo biopsy and the cartilage component should be targeted for histopathologic examination.

One-stage removal is indicated for stable symptomatic lesions. A marginal resection is adequate and demonstrates a low rate of recurrence. The entire lesion should be excised flush to the parent bone. Any remaining cartilage cap may result in recurrence, especially in growing lesions. In younger patients with secondary joint deformation, skeletal remodeling may return the structure to normal anatomy after the osteochondroma has been removed (16). This process has been particularly reported involving the ankle joint (16, 33–36).

Simple excision is not without risks and complications. Wirganowicz and Watts (37) evaluated 285 osteochondroma resections and reported a complication rate of 10%, although a majority of these complications occurred around the knee. Of the 19 foot and ankle cases included, there was only 1 complication of an intraoperative iatrogenic fibular fracture. The majority of extrinsic complications reported occur as a result of osteochondroma resection in an extremity and include adventitious bursae, tenosynovitis, tendon rupture, joint stiffness of surrounding joints, neuropathy, joint subluxation, synostosis, rotational deformity, parent bone fracture, osteochondroma stalk fracture, and premature physeal closure.

Major vascular structures adjacent to an osteochondroma are at risk for a serious associated event including arterial thrombosis, arteriovenous fistula, pseudoaneurysm, claudication, acute ischemia, phlebitis, and arterial rupture (6). Although these reports involve lesions occurring around the knee, the risk still remains theoretically significant with regard to the foot and ankle (6). Chalstrey (38) reported on a single case of a posterior tibial artery aneurysm due to a tibial osteochondroma. In general, with regard to a vascular incident associated with osteochondromas, surgical intervention is recommended when an osteochondroma is discovered in the direct vicinity of a major vessel, especially surrounding a mobile joint (39).

Osteochondromas are uncommon on the foot, and when they do occur they tend to involve long bones or tufts of the phalanges as subungual exostoses (40-42). While small exophytic bone growths are common on the calcaneus, they are often improperly referred to as osteochondroma. Plantar and posterior heel spurs are traction spurs and not true osteochondromas. Akmaz et al (43) reported on a calcaneal osteochondroma involving the plantar body. Nogier et al (10) recently reported on a large calcaneal osteochondroma occurring on the inferior lateral calcaneal tubercle. Similarly, this case also involved an extensive growth in adulthood, further demonstrating that benign growth of osteochondromas may occur after skeletal maturity. The peroneal tubercle has also been reported as a site for osteochondroma presentation, although it is unclear if these occurrences represent a true osteochondroma versus hypertrophied tubercles (44-46). Murphey and colleagues (21) list the following conditions as osteochondroma variants: subungual exostosis, dysplasia epiphysealis hemimelica, turret and traction exostoses, bizarre parosteal osteochondromatous proliferation, and florid reactive periostitis.

With regard to this case presented, the lesion was successfully removed and did not demonstrate recurrence at 3.5 years follow-up. However, the patient had not returned to her previous level of activity and went onto disability. One reason for the cause of the pain and disability may simply be due to the soft tissue disruption from excision of this large osteochondroma. Another possibility is that the nerves within the porta pedis were traumatized during the excision and rendered this patient with a baseline level of pain. It is also possible that detachment of the plantar fascia from its insertion, which was necessary for removal, may somehow be contributing to her residual pain through loss of the windlass effect on the arch. Overall complications associated with surgical excision of osteochondroma are low. Larger lesions involving the foot may result in significant long-term disability when excised.

In conclusion, symptomatic osteochondroma in the foot should be treated conservatively unless they become symptomatic, painful, demonstrate rapid or new growth, enlarge after skeletal maturity, and/or exhibit signs of malignant transformation. Large lesions are more likely to become symptomatic in the foot due to the low proportion of subcutaneous tissue in the region. We present a case of a giant osteochondroma involving the calcaneus, originating from the inferior medial calcaneal tubercle. To our knowledge this is the largest osteochondroma affecting the inferior medial tubercle of the calcaneus.

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