Case report

Giant extraskeletal osteochondroma of foot. A case report with review of literature

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1. Introduction

Osteochondromas are the most common benign tumor of bone. They usually arise from the growth plates of the metaphyseal area of the long bones, especially near the joint. They are composed of bone, having a cartilage cap and are connected with the medullary cavity often having a stalk. They increase in size during childhood and adolescent period and cease to grow after skeletal maturity. Extraskeletal osteochondromas, however, are uncommon variants of soft tissue chondromas which should be distinguished from conventional osteochondromas. Although they are histologically similar, extraskeletal osteochondromas are not attached to a parent bone. They most commonly occur in the digits (hands and feet) and surrounding the knee, and present as a small discrete calcified mass, that rarely exceed 2–3 cm in its greatest dimension. Other less common reported locations include within the thigh, hip, buttock, and the nape of the neck. The reported recurrence rate was suggested to be between 5% and 18%. We present a patient with a large extraskeletal osteochondroma (10 × 8 × 4 cm) of the plantar midfoot.

2. Case report

A 25-year old male patient presented at our clinic with a painful mass on the planter aspect of his right foot. The mass had grown

gradually in size for the past 1 year, and was associated with pain during walking. The patient had history of the foot, run over by tractor trolley one and half year back for which patient took rest and medications for week, no records of any fracture were found. Patient had no complaints after a week. Six months after the incident patient noticed swelling on the planter surface which was initially painless but as the mass grew over a period, however, it progressively caused an increasing degree of pain and discomfort during weight bearing and ambulation (Figs. 1–5).

Physical findings showed a firm, fixed and non tender mass in the medial aspect of the planter surface, measuring approximately 10 × 8 cm in size.

Plain radiographs showed a well-circumscribed, calcified mass at the medial plantar arch of the foot. Magnetic resonance imaging revealed a relatively large lesion with bone marrow signal intensity but no connection to bone. It was intimately related to the flexor tendons, and had produced a mass effect on the quadratus plantae muscle.

An excision biopsy was planned and performed. The mass was approached directly through the plantar aspect of the foot. It was well encapsulated and was easily dissected away from the other structures. The mass was confirmed intraoperatively to be exclusively extraskeletal, extra-articular, and extrasynovial. The mass was intimately related, but not attached, to the flexor tendons.

Grossly, it measured 10 × 8 × 4 cm, had multiple lobulations, was yellowish white, and appeared to contain both bony and cartilaginous tissue. On the cut surface, the tumor was mainly composed of whitish cartilage with focal bony hard areas. There were no intraoperative complications. A drain was temporarily placed for the prevention of hematoma. Postoperative radiographs were obtained. The postoperative course was uneventful. The

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Fig. 1. Lateral radiograph of the right foot demonstrates a large osseous mass of the plantar arch.

patient remained non-weight-bearing in a splint for 3 weeks postoperatively.

A histopathologic evaluation revealed a lesion composed of both bone and hyaline cartilage, with a partial cap of hyperplastic cartilage consistent with an extraskeletal benign osteochondroma.

3. Discussion

Conventional osteochondromas are the most common benign bone tumor, representing approximately 36% to 41% of benign bone tumors.¹ A “true” osteochondroma.

uncommonly occurs in the foot and ankle.² In contrast, extraskeletal osteochondromas are rarely occurring growths that arise within the soft tissue which are not contiguous with the bone or joint, and commonly occur in the hand and feet. Cases of extraskeletal chondroma are estimated to account for about 1.5% of all benign soft-tissue tumors.³

Extraskeletal osteochondromas are thought to originate from the synovial tissue of the joints, tendon sheaths, or bursae in the hands and feet, although their exact pathogenesis is unclear.⁴

One hypothesis holds that extraskeletal osteochondromas occur as a result of metaplasia of mesenchymal fibroblasts of the loose connective tissues while other theory states that it originates from pluripotent cell lines derived from the joint synovium, tenosynovium, or connective tissue.⁵ Adipose-tissue metaplasia was also suggested as a cause, attributable to the metamorphosis from a lipoma into an extraskeletal osteochondroma.⁴ Repeated trauma was also suggested to be an event provoking the development of extraskeletal osteochondroma.^{4,6}

Fig. 3. (A, B) The soft tissue osteocartilagenous mass during surgical exposure.

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 few patients, the lesion is painful and tender, especially if located on the plantar side of the foot.

Histologically, extraskeletal osteochondromas show a hyaline cartilage cap peripherally that is covered by a thin fibrous capsule and mature trabecular bone centrally.^{7,8,9} At the core are areas of prominent endochondral ossification, along with areas of lamellar and trabecular

Fig. 2. Preoperative MRI scan showing osteocartilagenous mass with some soft tissue edema and cartilaginous cap.

Fig. 4. Figure 4 (A, B, C) Macroscopically, lobulated osteocartilagenous mass with thin capsule removed, measuring $10 \times 8 \times 4$ cm.

Fig. 5. (A, B) Histopathology (magnification 40X; hematoxylin-eosin stain) illustrates findings consistent with an extraskeletal osteochondroma. Marrow space (asterisks) and hyaline cartilage cap (arrows).

bone. The thickness of the cartilage rim varies, and was reported to be as thin as 0.2 cm, and as thick as 1.2 cm.⁴ The cartilage may be hypercellular, and may demonstrate cellular atypia having hyperchromatic nuclei.⁴

Radiographs show a well-circumscribed, discrete, often lobulated mass with areas of calcification and ossification.⁹ MRI confirms extra-osseous involvement,⁴ and is helpful in the diagnostic work-up of symptomatic or suspicious osteochondroma to locate the extent of lesion, soft tissue involvement and to determine the thickness of cartilage cap which can rule out sarcoma. A cartilage cap generally demonstrates low-to intermediate signal intensities on T1-weighted images, and high signal intensities on T2-weighted images.

The differential diagnosis of an extraskeletal osteochondroma includes conventional osteochondroma, synovial chondromatosis, myositis ossificans, periosteal chondroma, periosteal chondrosarcoma, synovial sarcoma, and chondrosarcoma.^{4,8,9,10}

Extraskeletal osteochondromas are most frequently mistaken for conventional osteochondromas. Conventional osteochondromas typically are outgrowths of the long bones at the metaphysis, and are continuous with the medullary cavity of the parent long bone.^{4,9}

Synovial chondromatosis is characterized by multiple cartilaginous and osteocartilaginous nodules found within the synovium, and it may present as loose bodies inside the joint as well. Synovial chondromatosis has a diffuse growth pattern, and its cartilage proliferation is located subsynovially, which helps distinguish this lesion from an extraskeletal osteochondroma.^{4,9}

Myositis ossificans is often heterogeneous or amorphous in its early development and may increase or decrease in size within a few weeks. This lesion contains both cortical and medullary bone when mature, and it may adhere to the underlying bone. Radiographically, it is usually easy to distinguish because of its

peripheral ossification with central lucency, referred to as the “zonal phenomenon”.⁴

Periosteal chondromas occur on the surface of long bones, and create cortical erosions. Histologically, periosteal chondromas are viewed as well-defined lobules of hyaline cartilage, covered by periosteal connective tissue. They also exhibit hypercellularity, plump nuclei, and binucleated cells.⁴

Malignant processes may also be radiographically confused with extraskeletal osteochondromas. Synovial sarcoma is a malignant soft-tissue sarcoma that occurs primarily

in middle-aged adults. It involves painful masses most commonly occurring in the lower extremity (about 70% of cases). Their size typically ranges from 3 to 5 cm. Radiographically, scattered amorphous calcification and ossification within the soft-tissue mass are signs of malignancy. Histologically, a malignant process is usually obvious.

Malignant transformation in solitary osteochondroma is rare (1–2%).^{4,9} Chondrosarcoma is the most common malignant neoplasm arising from these lesions. The clinical features indicating malignant degeneration include new onset of pain, rapid growth, growth after skeletal maturity or large lesions. Cartilage cap thickness greater than 1–2 cm in adults and 2–3 cm in growing children often suggests malignant transformation.¹¹

4. Conclusion

Extraskeletal osteochondromas are benign, slow growing, and often painless lesions, and should be identified to avoid unnecessary aggressive surgical treatment.

We present a case of a giant extraskeletal osteochondroma originating in the arch of the foot, along with a review of the literature. The present case made an uneventful recovery following excision. To the authors' knowledge, this is the largest isolated extraskeletal osteochondroma of foot ever reported in literature. It was an atypical presentation in view that it presented in the skeletally mature patient, showed benign nature despite its large size, grew rapidly and was associated with trauma.

An extraskeletal osteochondroma should be considered when a discrete, well-defined osseous mass without any direct continuity

with the adjacent bone or joint is present in the soft tissues of the distal extremities. Asymptomatic osteochondroma in the foot may be treated conservatively unless they become symptomatic, painful, demonstrate rapid or new growth, 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. 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.⁹

Conflict of interest

None.

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