Case report

Intraosseous leiomyoma of the calcaneum: An unusual bone tumor of foot and review of literature[☆]

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ABSTRACT

Leiomyoma is a benign tumor of smooth muscle origin and commonly diagnosed in the uterus, gastrointestinal tract, skin, and mucous membranes. To the best of our knowledge, the only reported intraosseous leiomyomas in extremities occurred in the proximal aspect and distal aspect of the femur, in the tibia, and in the ulna. We are not aware of any previous reports of intraosseous leiomyomas in the foot. The radiograph of the intraosseous leiomyoma shows unilocular or multilocular lytic lesion with sclerotic rim. Due to lack of definitive radiological features on magnetic resonance imaging and computed tomography diagnosis of this rare tumor is established with histopathological study and immuno-histochemistry markers. Smooth muscle spindle cells and positive immunohistochemistry markers for muscle cells is hall mark for the diagnosis. The treatment of intraosseous leiomyoma is surgical intervention by excision with wide margin and curettage followed by filling the cavity. The diagnosis of this tumor is challenging due to its extraordinarily rare incidence. Intraosseous leiomyoma should be included in the differential diagnosis of intraosseous lesion with benign radiographic feature. We report of the first published case of primary intraosseous leiomyoma of calcaneum in a 22-year-old male patient.

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1. Introduction

Leiomyoma is a benign tumor of smooth muscle origin and commonly diagnosed in the uterus, gastrointestinal tract, skin, and mucous membranes.¹ Intraosseous leiomyoma is a rare bone tumor and primary leiomyomas of bone in axial skeleton involving mandible, temporal bone and ribs have been reported in medical literature.² To the best of our knowledge, the only reported intraosseous leiomyomas in extremities occurred in the proximal aspect and distal aspect of the femur, in the tibia, and in the ulna.^{3–8} We are not aware of any previous reports of intraosseous leiomyomas in the foot. The radiograph of the intraosseous leiomyoma shows unilocular or multilocular lytic lesion with

sclerotic rim. Due to lack of definitive radiological features on magnetic resonance imaging and computed tomography diagnosis of this rare tumor is established with histopathological study and immuno-histochemistry markers. Smooth muscle spindle cells and positive immunohistochemistry markers for muscle cells is hall mark for the diagnosis. The diagnosis of this tumor is challenging due to its extraordinarily rare incidence. Intraosseous leiomyoma should be included in the differential diagnosis of intraosseous lesion with benign radiographic feature. We report of the first published case of primary intraosseous leiomyoma of calcaneum in a 22-year-old male patient.

“The authors have obtained the patient's informed written consent for print and electronic publication of the case report.”

2. Case study

A 22 year old male-year male labourer presented with pain and swelling over right heel for one-year duration. He had no relevant trauma or constitutional symptoms suggesting systemic illness. Physical examination revealed diffuse swelling over right

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Fig. 1. Lateral radiograph showing an expansile lytic lesion with sclerotic margin in calcaneum (white arrow).

calcaneum. On palpation local temperature was within normal limit, tenderness was elicited over calcaneum and there were no skin changes. Ankle and foot had full range of movement. The remainder of the physical examination was unremarkable. His laboratory studies, including differential and white blood count, were normal. Plain radiograph revealed an expansile lytic lesion with sclerotic margin in calcaneum (Fig. 1). MRI showed a well circumscribed lesion in calcaneum (Fig. 2). The lesion was expansile and well margined with iso-intense intense signal on T1-weighted images and iso-intense to high intense in signal on T2-weighted images. Post-contrast fat-suppressed T1-weighted image showed homogenous enhancement of the mass. There was no perilesional edema and no extension into the soft tissue. Needle biopsy was performed and the histopathology showed spindle cells in whorled pattern. There was no cellular atypia, mitoses and necrosis. Surgical planning was done and lateral approach for the

Fig. 2. A sagittal Proton density fat suppressed MR image shows a well defined neoplasm with signal intensity higher than muscle (black arrow).

Fig. 3. This photomicrograph shows tumor composed of spindle cells in whorled pattern (Stain, hema-toxylin and eosin; original magnification, 100 $\times$).

calcaneum was used and the lesion was exposed and observed. Examination of the cortex revealed an expansile mass lesion. The specimen consisted of a nodular well-circumscribed firm, tan, rubbery mass and histologic studies were performed (Figs. 3 and 4). The tumor composed of spindle cells in whorled pattern. There was no cellular atypia, mitoses and necrosis. Immunohistochemistry markers for smooth muscle actin and desmin were positive and neural markers S-100 and CD 34 were negative (Fig. 5). The

Fig. 4. This photomicrograph shows tumor composed of spindle cells in whorled pattern without cellular atypia, mitoses and necrosis (Stain, hema-toxylin and eosin; original magnification, 400 $\times$).

Fig. 5. Immunohistochemical staining using desmin shows strong cytoplasmic positivity in the tumor cells (original magnification, 400 $\times$).

Fig. 6. Lateral radiograph of calcaneum showing an healed lesion and void filled with bone cement.

patient showed no signs of local recurrence during four years follow up period (Fig. 6).

3. Discussion

Leiomyoma is a benign smooth muscle tumor seen in the female reproductive system (uterus) and gastrointestinal system. The histopathology shows smooth spindle cell stroma and positive immunohistochemistry staining for muscle markers.¹ Intraosseous leiomyoma in axial skeleton involving mandible, temporal bone and ribs and appendicular skeleton involvement of ulna, tibia, proximal femur and distal femur are reported in medical literature^{2–9} (Table 1).

The postulated theory for the origin of this intra-osseous tumor is it originates from the smooth muscle tissue in the vascular system of the bone.^{2–9}

Intraosseous leiomyoma presents with a painful swelling. There is female preponderance and are seen in skeletally mature individuals.

Histopathological examination of leiomyoma is having spindle cells with elongated nuclei, eosinophilic cytoplasm arranged in whorled or fascicular pattern.^{1–9} The benign tumors with similar histological features are nerve sheath tumors, non ossifying

fibroma, fibrous histiocytoma. Immunohistochemistry stains are helpful for the diagnosis of these tumors. Leiomyoma stains positive for muscle markers i.e. desmin, smooth muscle actin and the markers S100, CD 34 are negative.^{1–9} Bundles of smooth muscle strongly staining for smooth muscle actin are diagnostic of leiomyoma. The differentiating features between intraosseous leiomyoma and leiomyosarcoma are lack of features of malignant spindle cell tumor, cellular atypia, mitosis and necrosis.^{7–10}

The radiological features of intraosseous leiomyoma are well defined osteolytic expansile lesion, sclerotic margin.^{2–5} MRI shows a well circumscribed lesion with iso-intense signal on T1-weighted images and iso-intense to high intense signal on T2-weighted images.^{2–9} Post-contrast fat-suppressed T1-weighted image showed homogenous enhancement of the mass. There was no perilesional edema and no extension into the soft tissue.

The differential diagnosis includes of intraosseous leiomyoma includes giant cell tumor, aneurysmal bone cyst, intraosseous schwannoma, chondroblastoma, xanthofibroma, spindle cell hemangioma.

Giant cell tumor of calcaneum shows areas of expansile bone lesion with destruction and erosion of bone cortex. Giant cell tumor around foot is often aggressive disease and requires proper treatment to prevent recurrence. The histopathologic examination of giant cell tumor shows mono-nucleated cells with multinucleated giant cells.¹¹

Aneurysmal bone cyst of calcaneum shows multiseptated lesion with fluid levels and extraosseous soft tissue extension. Histopathological study shows blood filled cavities lined by fibrous septation lined with endothelial cells.¹²

Intraosseous schwannoma of calcaneum shows a well-circumscribed, multilocular osteolytic lesion with peripheral sclerotic margin.¹³ There is thinning of the cortex and absence of soft-tissue invasion, periosteal reaction and calcification. Immunohistochemistry study of schwannoma is positive for protein S100 and negative for desmin and smooth muscle actin.¹³

Chondroblastoma of calcaneum shows a radiolucent lesion with a sclerotic margin with stippled calcification and septations.¹⁴ The histopathologic study of chondroblastoma shows cellular proliferation with chondroblastic cells embedded in immature benign cartilage matrix.¹⁴

Xanthofibroma of calcaneum shows a well demarcated expansile lesion with hypointense signal on T1 weighted images

Table 1

Review of literature on intraosseous leiomyoma involving axial skeleton.

Study	Age/sex	Site	X-ray features	MRI features	Surgery	Comments
Taxy et al. (1981)	37/F	Tibia (Periosteum)	Normal	–	Excision	2 cm lesion Bone Scan showed positive uptake
Braun et al. (1994)	54/F	Neck of Femur	Osteolytic lesion	–	Curettage and bone grafting	Disseminated peritoneal leiomyomatosis
Zikria et al. (2004)	31/F	Ulna	Osteolytic lesion	–	Enbloc resection	8 mm lesion
Laffosse et al. (2007)	42/F	Fibula	Multilocular osteolytic lesion with fine rim of sclerosis	Iso-intense on T1 and high signal on T2 images	Curettage and bone graft	2 cm lesion 2 year follow-up
Aisner et al. (2008)	37/M	Tibia	Intracortical Osteolytic lesion	High signal intensity on T1 and Iso-intense on T2	Enbloc resection	2 cm lesion 2 year follow-up
Chien et al. (2012)	57/M	Distal Femur	Osteolytic lesion	Iso-intense on T1 And Iso to high-signal intensity on T2 with fat saturation	Enbloc resection	7 cm lesion 2 year follow-up
Current Study (2016)	22/M	Calcaneum	Multilocular osteolytic lesion	Iso-signal intensity on T1 And High-signal intensity on T2	Curettage and cementing	Labourer patient 3 cm lesion 4 year follow-up with no recurrence

and hyperintense signal on T2-weighted Images.¹⁵ Histopathological examination shows areas of bone and soft tissue infiltrated by multinucleated giant cells, hemosiderin laden macrophages, cholesterol cleft and hemorrhages.¹⁵

Monostotic fibrous dysplasia shows an expansile, well-marginated osteolytic lesion. Monostotic fibrous dysplasia is having isointensities and high intensities on T1 and T2 weighted images, respectively and the lesion was enhances with gadolinium.¹⁶ The histopathologic examination shows fibrous tissue and foci of irregular woven bone.¹⁶

Spindle cell hemangioma of calcaneum is showing areas of large cavernous blood vessels and intermingled spindle tumor cells. Immunohistochemistry of spindle cell hemangioma are positive for α -smooth muscle actin and negative for D2-40 and Ki-67 markers.¹⁷ Magnetic resonance imaging shows low intensity on T1-weighted imaging, high intensity with mixed low and intermediate intensity on T2-weighted imaging and marginal enhancement on gadolinium enhanced, T1-weighted fat-suppression imaging.¹⁷

The treatment of intraosseous leiomyoma is surgical intervention by excision with wide margin and curettage followed by filling the void with bone graft.^{1–9} Laffosse et al. performed curettage of intraosseous leiomyoma of fibula.³ Enbloc resection or excision has been performed for intraosseous leiomyoma involving the ulna, tibia, proximal femur and distal femur.^{3–9} In this present study curettage of the calcaneum tumor was performed and the cavity was filled with bone cement. Regular follow-up is necessary to observe the risk of recurrence and none of the reported cases of axial intraosseous leiomyoma has showed local recurrence.^{2–9}

4. Conclusion

The diagnosis of this tumor is challenging due to its extraordinarily rare incidence. Intraosseous leiomyoma should be included in the differential diagnosis of intraosseous lesion with benign radiographic feature.

Conflicts of interest

The authors have none to declare.

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