A case of intraosseous angioleiomyoma

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We report a rare case of expansively growing angioleiomyoma in the proximal tibia of a 64-year-old woman. MRI revealed a well-defined area of low signal intensity on T1-weighted image and high intensity on T2-weighted image. Angiographs showed hypervascularity and vascular poolings. Microscopic examination after incisional biopsy showed an angioleiomyoma with bundles of nerve fibers.

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Angioleiomyoma of the extremity is usually found in the skin and subcutaneous tissue (Enzinger and Weiss 1988). We report a case of angioleiomyoma expansively growing in the proximal tibia. To our best knowledge, this is the first report of an intraosseous angioleiomyoma in the extremities.

Case report

A 64-year-old woman suffering from traumatic chronic subdural hematoma for two months was referred by a neurosurgeon for evaluation of acute pain in the left knee joint. There was a spherical, firm and fixed lump about 1 cm in diameter in the subcutaneous tissue at the medial infrapatellar region. The overlying skin appeared normal and there was no erythema. She could not recall when she first noticed this tumor. Laboratory data including a complete blood count, sedimentation rate and chemistry profile were normal. Chest radiographs were normal. Radiographs of the left knee showed a lesion with sclerotic margins in the proximal part of the tibia, and marginal calcification of the lump. Tomographs confirmed the radiolucent lesion which had marginal sclerosis and a central calcification (Figure 1). Magnetic resonance imagings (MRI) disclosed signals typical of soft tissue tumor since the signals had a low intensity which resembles that of muscles on T1-weighted image and high intensity on T2-weighted image. A ^{99m}-technetium-hydroxymethylene diphosphonate bone scintigraphy showed increased uptake over the lesion. ^{67}-Gallium citrate scintigraphy was normal. Angiographs of the left popliteal artery demonstrated irregular hypervascularity and vascular poolings within the tumor lesion. There was no involvement of the arterial branches in circumferential soft tissue. An incisional biopsy was histologically diagnosed as angioleiomyoma.

Marginal excision of the lesion and reconstruction using a corticocancellous bone graft were carried out. At the operation, the tumor was completely encapsulated by a thin layer of fibrous tissue. The tumor was embedded in the sclerotic medullary bone of the tibia, but a part protruded from the surface of the eroded cortical bone. The superficial portion of the tumor was embedded in the medial patellar retinaculum. The tumor was 5 x 3 x 1 cm in size, was elastic, and whitish yellow. A cross section revealed a whitish parenchymatous tissue with a central calcified lesion.

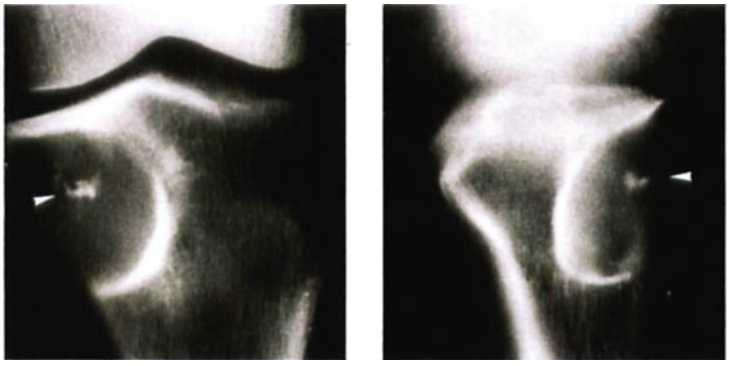
On histologic examination, the tumor was composed of many tortuous vascular channels with thick muscular walls and interlacing smooth muscle fibers. Foci of calcification and myxoid changes were also observed. Necrosis, cellular atypia or mitoses were not found. Bodian's staining revealed bundles of nerve fibers in the tumor.

Postoperatively the patient made a good recovery with full relief of her symptoms. 2 years postoperatively, there was no evidence of recurrence.

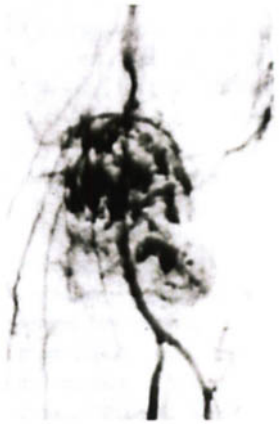
Discussion

Several authors have reported clinicopathologic studies of soft tissue angioleiomyoma (for review see Enzinger and Weiss 1988), while intraosseous angioleiomyomas are extremely rare. Only in the mandibular region have a few cases been reported (Rhatigan and Kim 1976, Goldblatt and Edesess 1977, McMillan et al. 1985).

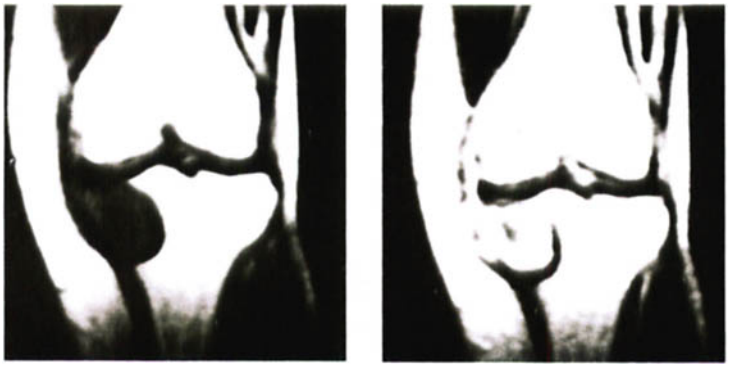
Figure 1. A 64-year-old woman with an expansively growing angioleiomyoma in the left proximal tibia.



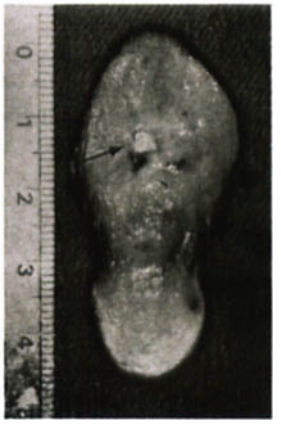
Tomographs showing radiolucent lesion of the proximal epimetaphyseal region of the tibia with a sclerotic inner border and a central calcification (arrow heads).



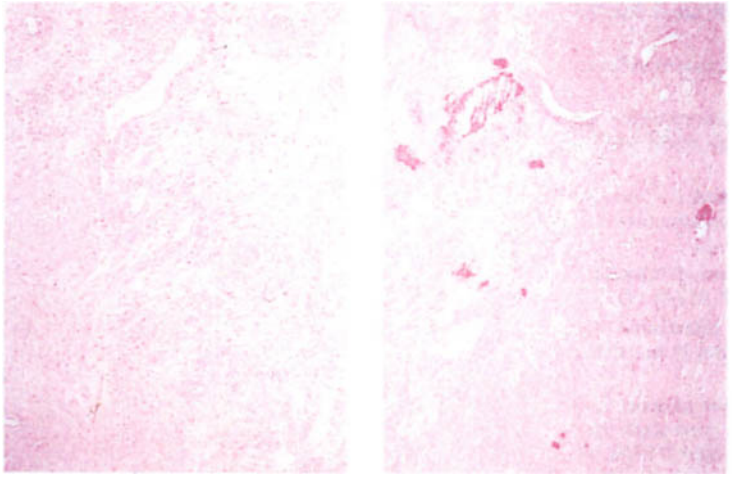
Angiograph in late phase showing the irregular hypervascularity and vascular poolings within the tumor mass.



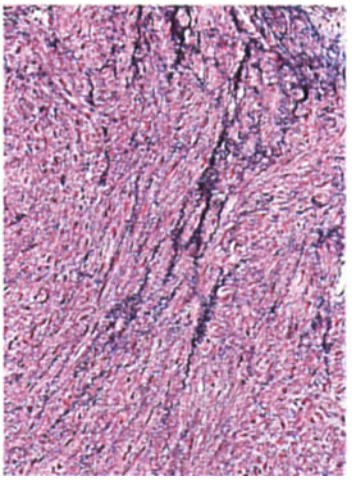
Coronal T-1 (TR 500msec; TE 300msec) (left) and T-2 (TR 2000msec; TE 900msec) (right) weighted MRI, the former demonstrating a soft tissue mass of a relatively low signal intensity replacing tibial bone, the latter the soft tissue mass of a high signal intensity with a central area having a low signal intensity.



Gross appearance of excised tumor showing a solid cut surface and a central calcified lesion (arrow).



Low-power view of angioleiomyoma. Tortuous vascular channels with thick muscular walls and interlacing smooth muscle bundles predominate (left). Foci of calcification are observed in the tumor (right), HE, ×25.



Bundles of nerve fibers are shown in the tumor by Bodian's staining, ×80.

The origin of angioleiomyoma of soft tissue in extremities is believed to be the tunic media of vessel walls in the subcutaneous tissue (Botte and Silver 1990). In previous cases of angioleiomyomas inside bone (Rhatigan and Kim 1976, McMillan et al. 1985) the tumor was thought to have arisen from the smooth muscle cells of vessel walls in bone. Evans and Sanerkin (1965) stated as to the histogenesis of primary leiomyosarcoma of tibial bone that it arose from the muscle in the media of a blood vessel, either de novo or through malignant change in a pre-existing angioleiomyoma. Genakos et al. (1987) suggested that if bone involvement of angioleiomyoma was apparent, leiomyosarcoma should be suspected. However, no cellular atypia or mitoses were observed in our case. We suppose that the tumor arose from the smooth muscle cells of vessel walls inside the cancellous bone.

The precise mechanisms of pain production of angioleiomyoma remain obscure. Duhig and Ayer (1959) stated that myxomatous degeneration resulting from contraction of the tumor vessels could be associated with the pain. On the other hand, Enzinger and Weiss (1988) pointed out the existence of nerve fibers. Recently, nerves were identified immunohistologically within the capsule and interstitium of angioleiomyoma (Fox et al. 1990). In our case, both focal myxoid change and nerve fibers were found.

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