

Case Report

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Volume 1 : Issue 2

Article Ref. #: 1000ROJ1109

Article History

Received: August 5th, 2016

Accepted: September 15th, 2016

Published: September 15th, 2016

Citation

Kobashi Y, Munetomo Y, Baba A, Yamazoe S, Mogami T, Miyauchi J. Two case reports of angioleiomyoma of the finger. *Radiol Open J*. 2016; 1(2): 55-59. doi: [10.17140/ROJ-1-109](https://doi.org/10.17140/ROJ-1-109)

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Two Case Reports of Angioleiomyoma of the Finger

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ABSTRACT

Angioleiomyoma represents a benign stromal tumor, which usually occurs in the subcutaneous tissue of the extremities. The angioleiomyoma in the finger is rare. We report 2 rare cases of an angioleiomyoma in finger. Case one, was a 72-year-old man with a mass on the radial side of distal phalanx of the left third finger. Case two, was a 70-year-old man with a mass on the ulnar side of interphalangeal joint of the left thumb. Both cases showed isointense to hypointense mass lesion on both T1 and T2-weighted images and were pathologically diagnosed with an angioleiomyoma. Heterogeneously isointense to hypointense on T2-weighted MR image showed two components: smooth muscle tissue punctuated with thick-walled vessels and/or hyalization on pathological finding. When much hyalization is included, it will not be enhanced on gadolinium-enhanced fat suppressed T1-weighted magnetic resonance (MR) image. Both T2-weighted and gadolinium-enhanced fat suppressed T1-weighted MR image findings should be considered to predict tumor composition.

KEYWORDS: Angioleiomyoma; MRI; Hyalization; Myxoid.

INTRODUCTION

Angioleiomyoma in the extremities is a benign smooth muscle neoplasm arising from the small blood vessels of the muscle. It was first described in 1937 by AP Stout,¹ who reviewed the literature on solitary cutaneous and subcutaneous leiomyomas and added 15 more cases from his own clinic-pathologic findings.^{1,2} We found 40 articles about magnetic resonance imaging (MRI) findings in angioleiomyoma using PubMed online. Many authors reported that angioleiomyomas showed different types of signal intensity on MRI. There are very few reports about pathological and radiological correlation in angioleiomyomas. We report the pathological and radiological correlation of 2 cases of angioleiomyoma in the finger seen in our hospital and observed on MRI using a microscopy coil.

CASE REPORTS

Case 1

A 72-year-old man came to our observation because of the presence of a mass on his left middle finger, which had been hurting for the recent two weeks. The mass was present for approximately 30 years without pain or increase in size. On physical examination, the mass was slightly mobile, approximately 1 cm in size and located subcutaneously just adjacent to the radial side of distal phalanx of the left third finger. Overlying skin was intact. Range of motion of the proximal and distal interphalangeal joints of the third finger was complete. He had no history of trauma on the site of lesion. Radiograph of the finger showed an oval-shaped soft tissue mass lesion on the radial side of the distal phalanx. There was no associated calcification or

bony involvement (Figure 1). An excision biopsy was proposed. Before the surgery, a finger MRI was performed.

The finger MRI (1.5 Tesla Achiva, Philips, Netherlands) using a microscopy coil (micro 47 grant coil) showed a heterogeneously hypointense mass lesion located in the subcutaneous tissue layer adjacent to the distal phalanx of the left middle finger on both T1-weighted (TR 405 msec, TE 15 msec, Thickness 1.5 mm, FOV 60 mm) (Figure 2a) and T2-weighted (TR 2500 msec, TE 90 msec, Thickness 1.5 mm, FOV 60 mm) (Figure 2b) coronal images. On gadolinium-enhanced fat-suppressed T1-weighted coronal and axial images (TR 470 msec, TE 13 msec, Thickness 1.5 mm, FOV 60 mm), the mass lesion showed enhancement in the margin especially in the superior

area (Figures 2c-2d arrows). A fibro-proliferative neoplasm, an angioleiomyoma or a calcium pyrophosphate dihydrate crystal deposition disease (CPPD) like gout was initially suspected. Tumor resection was performed and the circumscribed mass was confirmed to be located within the subcutaneous fat.

No involvement of the adjacent lateral collateral ligament of the distal interphalangeal joint was observed. On microscopic examination, the mass showed well-circumscribed fascicles of mature smooth muscle cells surrounding vascular lumina, lined by normal appearing endothelium (Figure 3a), predominantly in the peripheral area of the tumor and mostly in the stromal hyalization (Figure 3b). The pathological diagnosis was an angioleiomyoma.



Figure 1: X-ray of the left 3rd finger, AP view. Oval shaped soft tissue density mass lesion without calcification is visualized along the distal phalanx of the third finger (arrow).

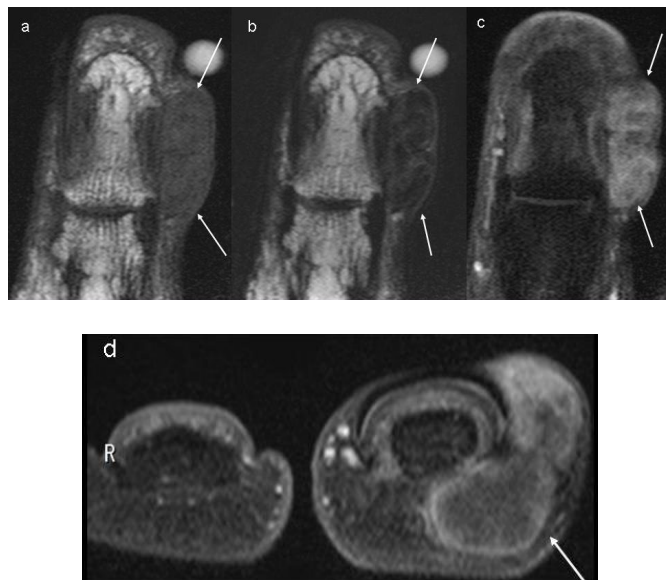


Figure 2: The third finger MRI using a microscopy coil (micro 47grant coil). The mass lesion shows very low signal intensity with intermediate signal septum on both T1-weighted (a) and T2-weighted (b) coronal images (arrows). On contrast study, the mass lesion is well enhanced (c, arrows).

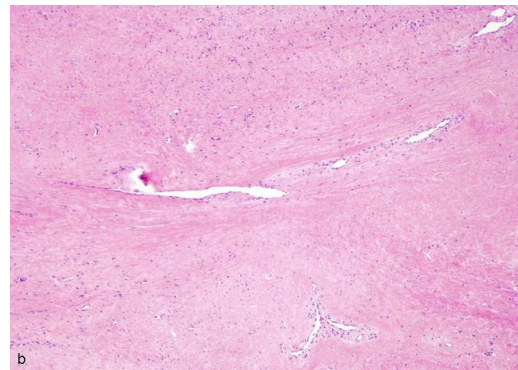
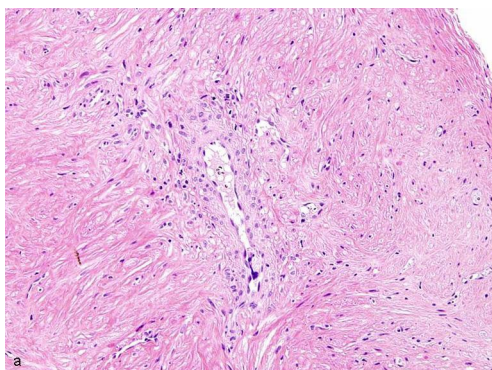


Figure 3: Tumor cells with H-E stain x200). The tumor consists of smooth muscle tissue punctuated with thick-walled vessels (a) and is predominantly visualized in the peripheral area. Most are occupied in the stromal hyalization (b).

Case 2

A 70-year-old man reported to our facility with a painless nodule adjacent to the ulnar side of interphalangeal joint (IP) of the left thumb. The nodule was present for approximately one year, with slow progressive growth. On physical examination, the nodule was rubbery in consistency, located subcutaneously and 1 cm in size. Overlying skin was intact. Range of motion of the left thumb was complete. Radiograph showed an oval-shaped soft tissue mass adjacent to the IP joint (Figure 4 arrow) of left thumb. An MRI (1.5 Tesla Achiva, Philips, Netherlands) was performed using a microscopy coil (micro 47 grant coil) in order to exclude the presence of a malignant tumor. It showed isointense nodule on both T2-weighted (TR 2550 msec, TE 90 msec, Thickness 1.5 mm, FOV 60 mm) (Figure 5a arrow) and T1-weighted (TR 400 msec, TE 15 msec, thickness 1.5 mm, FOV 60 mm) (Figure 5b arrow) image, and moderate enhancement on gadolinium-enhanced fat-suppressed T1-weighted im-

ages (TR 480 msec, TE 13 msec, Thickness 1.5 mm, FOV 60 mm) (Figure 5c arrow). The margins of the nodule were indistinct on MRI. We suspected an angioleiomyoma, a fibroproliferative neoplasm or a neurogenic tumor. Tumor resection was performed and a circumscribed mass was identified with in the subcutaneous fat layer. No involvement of the lateral collateral ligament of the IP joint was observed. Microscopically, it was composed of small vessels with smooth-muscle thickening of their walls (Figure 6). A little zonal distribution of hyalinization was noted. The pathological diagnosis was compatible with a solid type angioleiomyoma.

DISCUSSION

Angioleiomyoma in lower extremities occurs more frequently in women, while its presence at the head level, in the trunk or upper extremities is more common in men.^{3,4} The peak incidence is in the 4th to 6th decades of life. Pain and tenderness are more



Figure 4: X-ray of the left thumb, AP view. Oval shaped soft tissue density mass lesion is visualized at level of subcutaneous area of the IP joint (arrow). No bone erosive change is seen.

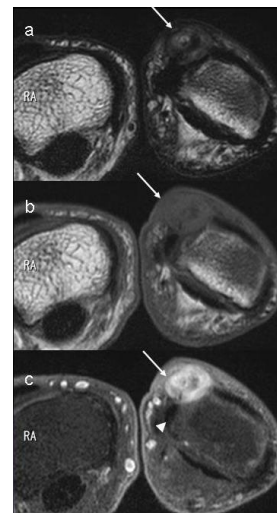


Figure 5: Thumb MRI using a microscopy coil (micro 47 grant coil). The mass lesion shows low signal intensity with a target like appearance on both T2-weighted (a, arrow) and T1-weighted (b, arrow). It is well-enhanced on gadolinium-enhanced fat suppression T1 weighted axial image (c, arrow). Medial lateral collateral ligament of the IP joint is indistinct (c, arrowhead).

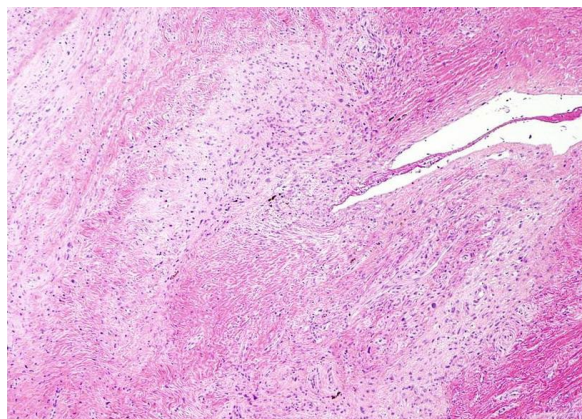


Figure 6: Tumor cells with H-E stain x100). Many small vessels with smooth-muscle thickening of their walls are visualized and are scattered among the stromal hyalinization.

frequent when the tumor is located in the lower extremities than in the upper extremities, head or neck. Duhig and Aye⁵ reported that 27 of 61 patients with this tumor complained of these symptoms.

Angioleiomyomas in the hand are rare and even more uncommon in the fingers. They account for 5-12% of all hand tumors. Prasad R et al⁶ reported that there are less than 200 cases of terminal phalanx angioleiomyoma described in the literature.

The most definitive diagnostic method to make a diagnosis is from histologic analysis of an excisional biopsy, together with a confirmatory immunohistochemical evaluation. Characteristics of angioleiomyoma on CT scan have not been well described yet. However, MRI has been frequently used as it is more sensitive for discerning between the soft tissue layers and can demonstrate a non-specific, well defined, round or oval mass in the subcutaneous or dermal tissue.⁷ Ashley DW et al⁸ reported that angioleiomyomas are usually either isointense or hyperintense if compared to skeletal muscle on T1-weighted MR image (low signal on magnetic resonance imaging) and heterogeneous on T2-weighted MR image (high signal on magnetic resonance imaging), and often have a hypointense fibrous capsule as the peripheral rim.

Some studies have reported about the pathological and radiological correlation of Angioleiomyoma.⁹⁻¹² Hwang et al¹⁰ suggested that the smooth muscle and the numerous vessels corresponded to the high signal intensity areas, while the fibrous tissue appeared iso signal intensity on T2-weighted MR images. Yoo et al¹¹ suggested that the presence of tortuous vascular channels surrounded by smooth muscle bundles and areas of myxoid change could explain the heterogeneity of signal intensity of the tumor on T2-weighted images. Matsuyama et al¹² reported that some flow void is suggestive of vessels along and/or within the tumor and can help in the diagnosis of angioleiomyoma. Some tumors showed predominant myxoid change and hyalization corresponding to the higher signal intensity on T2-weighted image than the remaining part of mass.⁹ However, in case 1, only hyalization component was visualized in the center area of the tumor on pathological findings and it showed heterogeneously hypointense signal on T2-weighted MR image. It may suggest that myxoid component reflects hyperintense signal and hyalization reflects hypointense signals on MRI. However, myxoid and hyalization occurring in angioleiomyomas might be secondary to the circulatory disturbance. In addition, small vessels with smooth-muscle thickening on pathological finding were consistent, with marginal enhancement area on gadolinium-enhanced fat suppression T1-weighted MR image. On the other hand, in our case 2, little hyalization areas were visualized in the tumor and showed isointense on T2-weighted MR image. Hyper cellular component composed of many small vessels with smooth-muscle thickening gave isointense on T2-weighted MR image. As the evidence, the tumor of case 2 moderately enhanced on gadolinium-enhanced fat suppressed T1-weighted MR image as compared to the tumor of case 1. Basing on the diagnostic

evidences obtained from our 2 cases, if we are able to consider both findings of T2-weighted and gadolinium-enhanced fat suppressed T1-weighted MR image, we could predict the tumor composition.

The differential diagnosis of a well-defined, enhancing, subcutaneous nodule or mass with T2-hyperintense to isointense signals includes synovial sarcoma, other low-grade soft tissue sarcomas, haemangioma, neurogenic tumour and nodular fasciitis. In addition, Low-grade sarcomas such as synovial sarcoma and low-grade myxofibrosarcoma may be slow in growing and appear well-circumscribed on MRI, giving the misleading impression that the lesion is well-localised. Haemorrhage, which may be seen as T2-hypointense, may be present in synovial sarcomas.

CONCLUSION

Angioleiomyoma of the finger is a rare tumor. Heterogeneously isointense to hypointense on T2-weighted MR image shows 2 components on pathological findings: a smooth muscle tissue punctuated with thick-walled vessels and/or hyalization. When much hyalization is included, it will not be enhanced on gadolinium-enhanced fat suppressed T1-weighted MR image. Both T2-weighted and gadolinium-enhanced fat suppressed T1-weighted MR images findings should consider in order to predict the tumor composition.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

CONSENT

The authors obtained written informed consent from the patients for submission of this manuscript for publication.

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