

Case Report

ANGIOLEIOMYOMA OF UTERUS – A RARE BENIGN SMOOTH MUSCLE TUMOUR

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ABSTRACT

Uterine angioleiomyoma (vascular leiomyoma) is an uncommon smooth muscle tumor derived from blood vessels and is almost exclusively reported in the subcutaneous soft tissues of extremities. When seen in the female genital tract, the lesion is very rare. The radiological and clinical characteristics that distinguish uterine angioleiomyoma from conventional leiomyoma are not well documented in literature. Herein, we describe a 32-year-old woman with complaints of menorrhagia and pelvic pain. Ultrasonography showed an intramural mass suggesting a leiomyoma. This mass was surgically excised through an open myomectomy procedure. The pathologic findings were characteristic of angioleiomyoma and the differential diagnosis with conventional leiomyoma is discussed. Angioleiomyoma should be considered a rare variant of uterine smooth muscle tumors due to its specific potential to cause heavy menstrual bleeding by virtue of its vascular nature and due to its increased risk of causing hemorrhage during surgical excision.

Keywords: Angioleiomyoma; CD34; Leiomyoma; Menorrhagia; Myomectomy; Uterus; Vascular leiomyoma

INTRODUCTION

Angioleiomyoma is also known as vascular leiomyoma, and is derived from the smooth muscle of the vascular wall. These tumors are most frequently found in the subcutaneous tissue of the extremities, particularly the lower extremities, and only rarely occur in the uterus. Because the clinical and imaging characteristics of uterine angioleiomyoma can be virtually indistinguishable from those of a typical leiomyoma, and because of its rich vasculature, menorrhagia, pelvic pain and intraoperative bleeding may occur. Thus, this article brings to light a rare mimicker of typical leiomyoma, and serves to help clarify relevant clinicopathologic findings.^[1-4]

CASE REPORT

A 32 year old female patient had a complaint of heavy bleeding per vagina and lower abdominal pain for the last 6 months. There was no intermenstrual or postcoital bleeding or loss of weight. She had no other relevant past medical or surgical history. General and systemic examination was also non-revealing. On pelvic examination, the uterus was

slightly enlarged. Ultrasonography done confirmed a well defined solid mass in the uterine cavity suggesting leiomyoma. A provisional diagnosis of uterine leiomyoma was thus made and the patient was posted for open myomectomy.

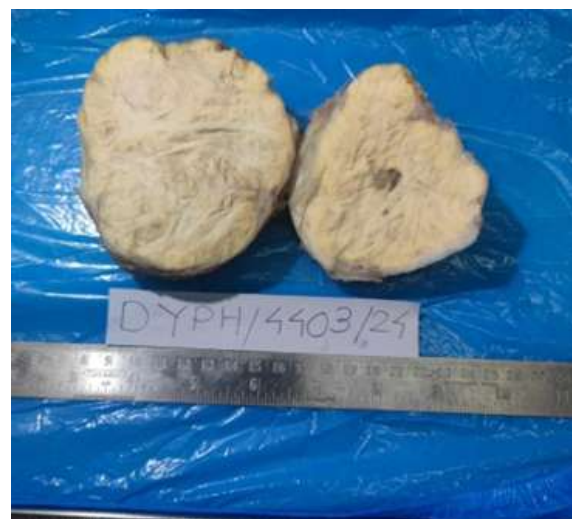


Figure 1: Gross photograph of the uterine mass showing a well-circumscribed intramural tumour with a firm, grey-white cut surface and focal haemorrhagic areas.

A discrete, firm intramural mass was received in 10% neutral buffered formalin and measured 12 × 9.5 × 8 cm in greatest dimensions. The external surface was smooth. Sectioning the mass revealed a solid, grey-white (tan) background with foci of hemorrhage and trabeculations. At low power, the mass contained fascicles of bland smooth muscle cells with cigar-shaped nuclei and eosinophilic cytoplasm. Within these fascicles there were numerous thin walled, capillary-sized vascular channels scattered throughout the mass. No atypia was seen, there was no coagulative necrosis, and there was no significant increase in mitotic activity. CD34 stained the endothelial lining of the vascular channels, confirming a diagnosis of uterine angioleiomyoma. [Figure 1-3]

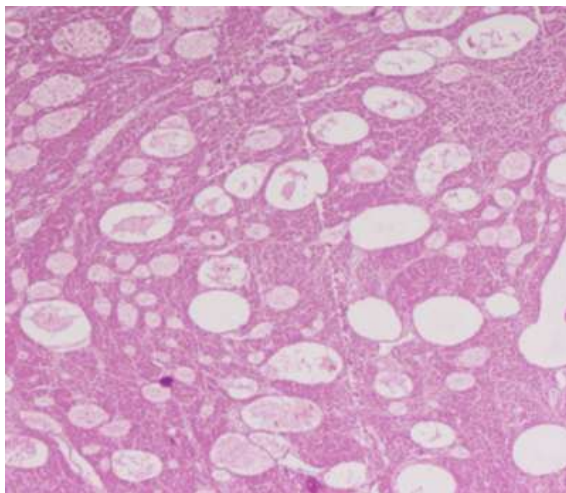


Figure 2: H&E section showing smooth-muscle bundles with numerous thin-walled capillary-sized vessels, consistent with capillary angioleiomyoma.

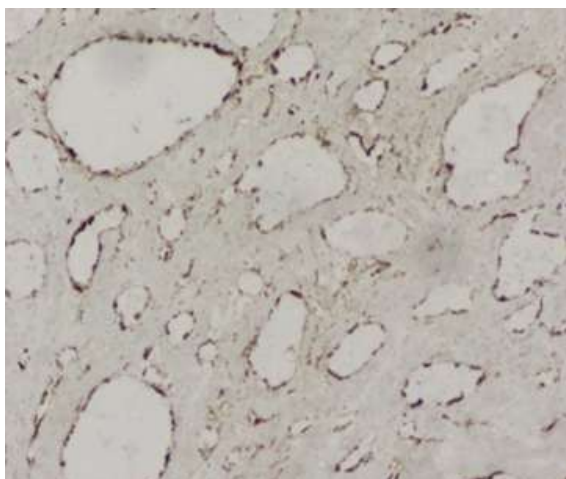


Figure 3: CD34 immunostain highlighting endothelial lining of proliferating vascular channels, supporting a diagnosis of angioleiomyoma.

DISCUSSION

Angioleiomyoma is a smooth muscle tumor arising from the tunica media of vessels. It is a common lesion in the soft tissues of the skin. Uterine

angioleiomyoma is a rare tumor and is often under-recognized, being misdiagnosed as conventional leiomyoma pre-operatively in the majority of reported cases, and therefore only identified at histology with further confirmation by immunohistochemistry. In the current study, imaging suggested the diagnosis of a leiomyoma and it was only confirmed by microscopy following which the immunohistochemistry supported the final diagnosis thus highlighting the potential for diagnostic confusion.^[5]

Patients with smooth muscle tumors may present with abnormal uterine bleeding or pelvic pain due to the vascular nature of the lesion. Many prior clinicopathologic reports have cited menorrhagia as a symptom of the lesion. Severe complications have been rarely reported in conjunction with the bleeding, so awareness of this possible consequence should be kept in mind. The tumor in the current case was well circumscribed and showed large hemorrhagic foci consistent with its clinical presentation. It was composed of unremarkable smooth muscle cells in a fascicular arrangement with prominent blood vessels; and there was no appreciable atypia, necrosis, or increased mitotic rate.^[6]

The differential diagnosis is a conventional leiomyoma with secondary vascular changes or degeneration, hemangioma, angiomatoid lesions or other smooth muscle tumors with prominent vascularity. The key points to look for are the pattern of vascular channel density and distribution and the bland appearance of the smooth muscle. Immunohistochemistry is useful and endothelial markers such as CD34 are helpful in highlighting the vascular channels and confirming the vascular nature of the lesion. Despite being a benign entity, large size and vascularity may cause clinical suspicion of malignancy or non-uterine pelvic neoplasm, which can lead to difficulty in diagnosing these lesions.^[7]

Wide local excision of a uterine angioleiomyoma appears to be a therapeutic procedure, associated with a good outcome. No malignant degeneration of leiomyomas has been reported in the literature to date. This rare case suggests that, clinically, the recognition of a uterine angioleiomyoma is primarily a warning for increased intra-operative bleeding and underlines the fundamental role of histopathology for an exact diagnosis.

CONCLUSION

Uterine angioleiomyoma is a rare benign smooth muscle neoplasm that closely resembles conventional leiomyoma clinically and radiologically. The definitive diagnosis is based on the histopathological findings with confirmation by appropriate immunohistochemistry. The recognition of this rare tumour is important for accurate diagnosis, as there may be risk of increased bleeding because of its highly vascular nature.

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