#### **Case Report**

# **Vulvar Angioleiomyoma**

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#### **Abstract**

Vulvar angioleiomyoma is a very rare neoplasm. We describe the case of a 49-year-old woman who presented with a small, firm, tender labial mass. Intraoperatively, the lesion appeared hypervascular and was excised using sharp dissection. Histological and immunohistochemical analyses were necessary to make the diagnosis. The report of this extremely uncommon gynecologic lesion is important to make clinicians aware of the possibility of vulvar angioleiomyoma.

Keywords: Angioleiomyoma, case report, labial mass, pathology, vascular leiomyoma, vulvar mass

#### INTRODUCTION

Angioleiomyoma (otherwise known as vascular leiomyoma) is a very uncommon, benign mesenchymal neoplasm. This mass can arise anywhere in the body but generally occurs in the subcutaneous tissue of the lower extremities. Angioleiomyomas originate from smooth muscle cells of arterial and venous walls and typically present as a solitary, subcutaneous painful nodule. This lesion is most common in the fourth to sixth decades of life.<sup>[1]</sup>

This tumor has been rarely described in the female urogenital tract, although a few reports on angioleiomyoma of the uterus<sup>[2]</sup> have been published. Presentation in the ovaries,<sup>[3]</sup> pelvic retroperitoneum,<sup>[4]</sup> broad ligaments,<sup>[5]</sup> female periurethra,<sup>[6]</sup> and vulva<sup>[7]</sup> are even more unusual. As a result, angioleiomyoma is not commonly included in the differential diagnosis for labial mass, which makes preoperative diagnosis difficult. Here, we present the case of a 49-year-old woman whom we diagnosed with angioleiomyoma of the labia majora.

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# CASE REPORT

A 49-year-old, gravida 10, para 8 with diabetes mellitus, hypertension, hyperlipidemia, and obesity presented to our clinic with a 5-year history of a rubbery, mildly tender right labial mass measuring approximately 3 cm × 2 cm. A reading of the magnetic resonance imaging (MRI) of the pelvis reported an "oblong shaped and peripherally enhancing 2 cm cystic focus in the subcutaneous tissue of the inferior right labial fold which may represent a nonspecific subcutaneous cyst, versus a Bartholin or other small glandular cyst."

We excised the labial mass using careful sharp dissection. Grossly, the lesion appeared hypervascular, white in color, and spongy in texture [Figure 1]. Microscopically, the mass was well-circumscribed, with "nerve twigs" identified within and at the edge of the lesion. Ki-67 did not show increased proliferative activity. The vascular component of the labial mass stained positive for CD31 and CD34, which are markers found on endothelial cells and expressed by

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Figure 1: Intraoperative image of vulvar angioleiomyoma

vascular tumors.<sup>[3]</sup> The intervening stroma showed prominent staining for smooth muscle actin, caldesmon, and desmin. The histological and immunohistochemical results were consistent with the diagnosis of angioleiomyoma. Physical examination 8 months after surgery revealed no evidence of recurrence.

The patient gave verbal consent to the de-identified publication of this report.

#### DISCUSSION

Although leiomyoma are the most common tumors found in the human body,[8] subtypes such as angioleiomyoma are especially uncommon.<sup>[9]</sup> Reports of vulvar angioleiomyoma are especially scarce; [7] therefore, this lesion lacks a typical clinical presentation which makes preoperative diagnosis difficult.[4] The diagnosis of angioleiomyoma should be considered in patients presenting with a firm, tender vulvar mass. MRI findings are nonspecific.[2] Histologically, angioleiomyoma is similar to leiomyoma in that they are comprised of interlacing smooth muscle cells, but microscopy also demonstrates regularly distributed small blood vessels in angioleiomyoma.[10] Immunohistochemical markers and assays for smooth muscle cell components assist in achieving a pathologic diagnosis, [5] as was the case for the only other published report of vulvar angioleiomyoma.<sup>[7]</sup> This neoplasm has benign features and a good prognosis, [3] with recurrence unlikely after resection.[1]

## CONCLUSION

Angioleiomyoma is uncommon in general, but published documentation of vulvar lesions of this kind is incredibly rare. This report provides an example of vulvar angioleiomyoma and provides insight into its clinical, radiographic, and histological presentation. This report may help clinicians consider this diagnosis when encountering a firm vulvar lesion with microscopic evidence of vascularity and histochemical markers consistent with vascular and smooth muscle tumors.

#### Ethical approval and declaration of patient consent

The IRB approval of this study was exempted. The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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#### **Conflicts of interest**

There are no conflicts of interest.

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