Human Pathology: Case Reports (2015) 2, 83–86

ELSEVIER

Human PATHOLOGY Case Reports

http://www.humanpathologycasereports.com

Lipomatous angiomyofibroblastoma: A 20-year literature review ☆

Julie Guilmette MD [a,*], Philippe Gauthier MD, FRCSC [b], Kurosh Rahimi MD, FRCPC [a]

[a] *Department of Pathology, Centre Hospitalier Universitaire de Montréal, Montreal, Quebec, Canada*
[b] *Department of Gynaecologic Oncology, Notre-Dame Hospital, Montreal, Quebec, Canada*

Received 15 January 2015; accepted 27 January 2015

Keywords:
Angiomyofibroblastoma;
Lipomatous;
Vulva;
Vagina

Abstract Lipomatous angiomyofibroblastoma (LAMF) is a rare superficial muco-cutaneous tumor of the female genital tract in women of reproductive age and early menopause. We report the case of a 36-year-old woman who presented with an asymptomatic 1 cm round vulvar mass. The tumor showed both hypercellular and hypocellular areas composed of spindle and epithelioid cells set in a loose fibromyxoid background. Anastomosing networks of small to medium sized dilated vessels, surrounded by condensed spindle cells, were distributed equally throughout the tumor. Mature adipocytes comprised more than half of the tumor's volume. The neoplastic cells were reactive for both desmin and vimentin and did not express CD34, smooth muscle actin, S100 protein and muscle-specific actin. Making the right diagnosis may be challenging to medical professionals as LAMF may mimic both clinically and histologically several other benign and malignant gynecological lesions. LAMF has a benign course without evidence of local recurrence or metastatic potential.

1. Introduction

Angiomyofibroblastoma (AMF) is an unusual benign mesenchymal neoplasm of the female genital tract which arises as a superficial soft tissue tumor in women of reproductive age and in early menopause. Over the last decades, nine cases of AMF with a significant content of adipose tissue have emerged in the literature and became known as the lipomatous variant of AMF (LAMF).

2. Case report

A 36-year-old patient presented for her annual gynecological exam with a genital mass. Upon examination, a superficial 1 cm round vulvar mass localized on the left labium minus was revealed. The patient was otherwise asymptomatic. Initial clinical diagnosis included an inclusion cyst. The mass was excised under local anesthesia.

3. Pathological findings

Macroscopic examination revealed a 1.1 × 1.0 × 0.9 cm subcutaneous polypoid mass. Cut section revealed a solid rubbery mass white to yellow in color.

☆ Disclosure information: All authors had nothing to disclose. No funding was obtained.

* Corresponding author at: Département de pathologie et biologie cellulaire, N-528, Faculté de médecine–Université de Montréal, C.P. 6128, succursale Centre-ville, Montréal, QC H3C 3J7, Canada. Tel.: +1 514 434 4423; fax: +1 514 343 5755.
E-mail address: Julie.guilmette@umontreal.ca (J. Guilmette).

http://dx.doi.org/10.1016/j.ehpc.2015.01.002

On histological examination, the polypoid dermal lesion was covered with a normal squamous epithelium and contained alternating hypocellular and hypercellular areas composed of intermediate sized spindle to ovoid cells set in a loose fibromyxoid stroma (Fig. 1A). Mature adipocytes, located in the center of the lesion, occupied more than 50% of the total mass (Fig. 1B). Anastomosing networks of small to medium sized dilated vessels, surrounded by a cuff of condensed spindle cells creating a sleeve-like pattern, were distributed equally throughout the tumor (Fig. 2). Adnexal structures, such as sebaceous and eccrine sweat glands, were entrapped within the mass. Non-encapsulated pushing borders were visualized. Only very rare mitoses were present, and there was no necrosis (Fig. 2).

4. Immunohistochemistry

An immunohistochemical study was performed using the following monoclonal antibodies for CD117, CD68, S100 protein, CD31, CD34, CD3, CD20, vimentin, desmin, smooth muscle actin and muscle-specific actin.

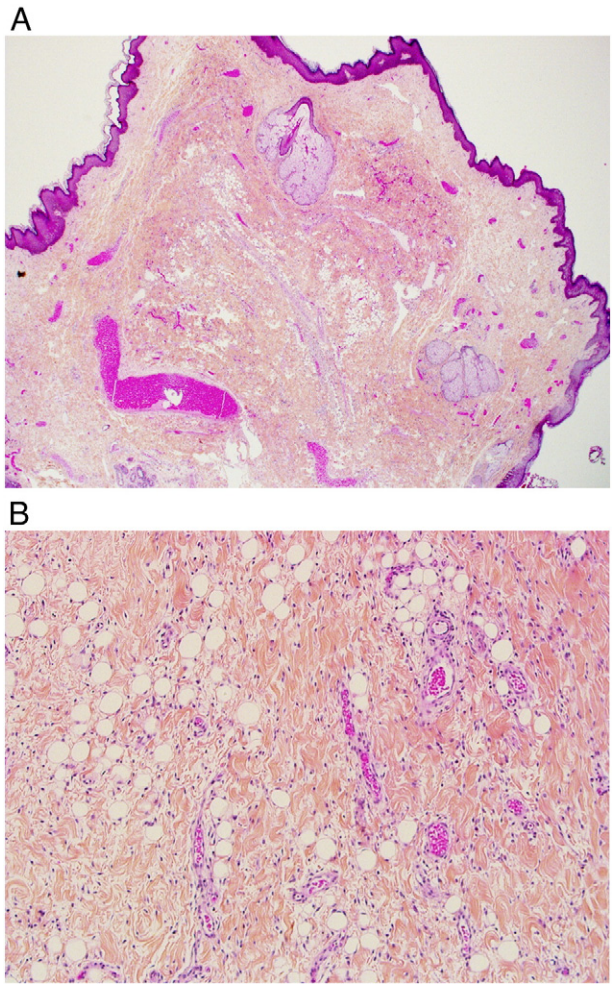

Fig. 1 Lipomatous angiomyofibroblastoma. (A) Polypoid mucocutaneous dermal lesion with loose fibromyxoid stroma. Adnexial structures are entrapped at the periphery. (B) Scattered mature adipose tissue within the tumor's stroma.

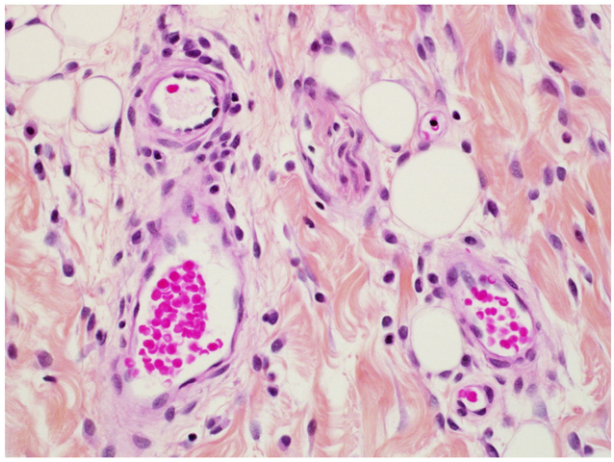

Fig. 2 Lipomatous angiomyofibroblastoma. Network of small to medium sized dilated vessels surrounded by condensed spindle cells (sleeve-like pattern). Absence of mitose and necrosis.

The spindle cells were reactive for both desmin and vimentin and did not express CD34, smooth muscle actin, S100 protein and muscle-specific actin. Dispersed and centrally located mature adipocytes expressed S100 protein. CD117 and CD68 markers stained a few dispersed mast cells and histiocytes, respectively.

5. Discussion

LAMF is a rare benign mesenchymal neoplasm that generally arises as a superficial mass within the female genital tract [1,2]. To our knowledge, there have been nine cases of LAMF reported in the literature (Table 1). Four of those cases (case 6, 7, 8, and 9) were described as AMF with a major component of adipose tissue [3,4]. The age of diagnosis ranged from 23 to 50 years with an average of 38 and a median of 39 years. Among reported symptoms, four patients described a painless mass or lesion. All LAMFs occurred in the vulva. Size varied from 1.1 to 11 cm with an average of 5.5 cm. In six cases, mature adipocytes made up 30% to 80% of the mass; this information was not available for other cases. This raises the question of using a threshold in an AMF as a lipomatous variant. Hiruki et al. [3] were the first to introduce the adipose tissue as an integral component of the tumor. This opens the perspective that vulvar stem cells differentiate toward mesenchymal cells as in AMF as well as toward lipomatous cell as in LAMF.

All ten cases of LAMF are histologically similar as they are described as having both hypercellular and hypocellular areas composed of spindle and epithelioid cells set in a loose fibromyxoid background. Vascular networks surrounded by condensed spindle cells were distributed throughout the tumor. Mature adipocytes comprised a significant amount of the