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Research Paper

Atypical vulvar and vaginal mass revealing an angiofibroma: a literature review of two cases.

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Abstract
Angiofibroma is a rare benign mesenchymal tumor. In women, it occurs mainly in the vulvovaginal region. Its clinical presentation is misleading and similar to several other vulvar tumors of different cellular origins. Its histological and immunohistochemical characteristics help to distinguish it from other tumors.

We present the case of two patients who consulted for the management of a vulvar mass in one case and a vaginal mass in the other, for whom the diagnosis of angiofibroma was made on the excised specimen and confirmed by immunohistochemical study.

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I. Introduction

Vulvovaginal mesenchymal tumors are rare but represent a broad spectrum of lesions, including aggressive angiomyxoma (AAM), angiomyofibroblastoma (AMFB), cellular angiofibroma (CAF), and related entities. Cellular angiofibroma is a benign tumor specific to the soft tissues of the lower genital tract. An accurate diagnosis of these tumors is important because of the differences in prognosis.
In this literature review, article we report two cases of masses of the female genital tract in different locations, the excision of which revealed an angiofibroma after immunohistochemical study.

Medical history of Patients

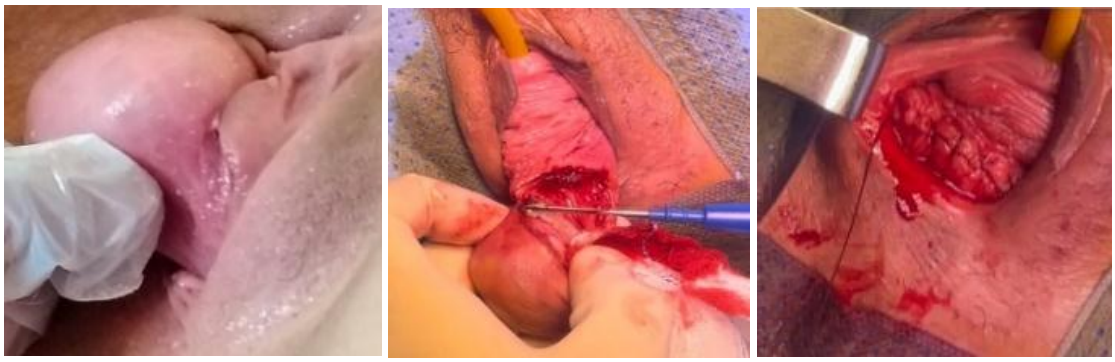
Case 1

The patient was a 40-year-old primiparous woman who consulted for the management of a vaginal swelling that had appeared 2 years ago.

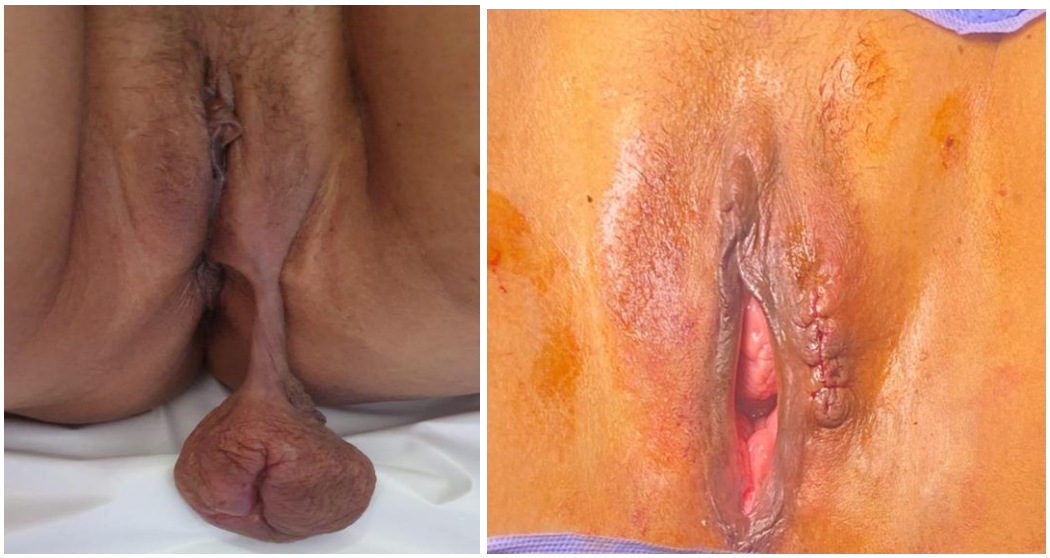
Pelvic examination revealed a pink swelling prolapsing from the vagina, well defined, pink, about 4 cm long, hard, painless, appearing to detach from the anterolateral vaginal wall and near the urethral meatus; the cervix and the rest of the vaginal mucosa were macroscopically healthy.

A pelvic MRI was requested to characterize the mass and rule out any malignancy. It described an atypical pedunculated vaginal mass whose insertion zone appeared to be in the anterior wall of the upper third of the vagina, measuring 04x3 cm; a biopsy of the mass with an anatomopathological study concluded that the vaginal mucosa was normal. The decision was made to surgically remove the mass for therapeutic and diagnostic purposes.

She underwent vaginal excision of the mass; histopathology revealed a nodular, poorly cellular formation lined with surface epithelium, with numerous vascular structures surrounded by smooth muscle fibers and loose fibroblasts tissue; the immunohistochemical study showed desmin to be negative and CD34 to be positive, confirming the diagnosis of angiofibroma.



Case 2
The patient was a 61-year-old multiparous woman with no known comorbidities who consulted a specialist for the treatment of a chronic vulvar growth that had been progressively increasing in size over the years.
On examination, a mass was found in the lower 1/3 of the labia majora, with a soft pedicle and the same coloring as the vulva, measuring approximately 5 cm and painless on palpation.
She underwent an excision of the mass with the closure of the margins and the specimen was sent for histological study.



The macroscopic appearance is that of a polypoid formation ulcerated on the surface, lined by skin tissue measuring 8x6x4 cm with a whitish appearance when cut.
Anatomopathological examination revealed fusocelullar proliferation on a myxoid background, the nature of which is to be determined by immunohistochemical study. The immunohistochemical study revealed the positivity of calponin and the negativity of desmin, AML, CD34 and progesterone receptors.

II. Discussion

Vulvovaginal mesenchymal tumors are essentially benign tumors, as described in 2000 by M. Nucci; these tumors share clinical, macroscopic, microscopic, and immunohistochemical characteristics, making it quite difficult to distinguish them from one another in some cases.[1]

It is found in women aged between 22 years and 77 years, with a median age of around 47 years [2]. In women, the labia majora are the most frequent site, followed by the vagina and perineum.[3]

They most often present as a well-limited, painless, slow-growing mass that has been present for weeks or years prior to diagnosis. Clinically, cellular angiofibroma is often mistaken for Bartholin's cyst, labial or submucosal cyst[4]. Diagnosis usually follows a complete local excision of the clinically benign mass.[5]

Histologically, the cellular angiofibroma is well-circumscribed and has two main components: spindle cells and prominent blood vessels. Mitotic activity is low and there is no cytonuclear atypia or tumor necrosis.[1]

This lesion expresses hormone receptors and in approximately 50% of cases, CD34. It does not usually express desmin or smooth muscle actin (SMA)[5] ;[1] . It is important to note that smooth muscle markers (particularly desmin) are usually negative, unlike other vulvovaginal mesenchymal tumors.

Several tumors need to be considered in the differential diagnosis of cellular angiofibroma. The first is invasive angiomyxoma (AAM): a large malignant tumor (generally >5 cm in diameter) with a tendency to recur after local excision; the second is angiomyofibroblastoma; immunohistochemistry
and FISH analysis could be used in such conditons[6]. Other differential diagnoses include leiomyoma, spindle cell lipoma, and mammary-type myofibroblastoma (MFB). [5]
Surgical removal of the lesion with tumor-free margins appears to be the most appropriate management for these lesions. This therapeutic procedure also helps to correctly diagnose the patient.[1]

III. Conclusion:

Cellular angiofibroma is a rare vulvar tumor that must be considered in the differential diagnosis of vulvar tumors. The immunohistochemical study confirms the diagnosis and eliminates differential diagnoses with prognosis and treatment.

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