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Vulvar leiomyoma mimicking Bartholin's gland cyst: A case report

Saeed Baradwan [a,*], Hassan M. Latifah [a], Haneen Al-Maghrabi [b], Abdulmonem M. Almutawa [b]

[a] Department of Obstetrics and Gynecology, King Faisal Specialist Hospital and Research Center, Jeddah, Saudi Arabia
[b] Department of Pathology and Laboratory Medicine, King Faisal Specialist Hospital and Research Centre, Jeddah, Saudi Arabia

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ABSTRACT

Vulvar leiomyomas are rare benign tumors originating from smooth muscle cells of the vulvar tissue. This report concerns a 31-year-old woman who presented with a painless vulvar mass measuring 5 × 4 cm. The mass was clinically diagnosed as a Bartholin cyst due to its non-tender and soft nature, and as a result the patient underwent wide local excision of the vulvar mass. Histopathology confirmed it to be benign vulvar leiomyoma. This case emphasizes the importance of thorough clinical evaluation and accurate histopathological examination in identifying vulvar masses. This report underscores the diagnostic difficulties associated with rare vulvar lesions and stresses the necessity of a comprehensive approach to their evaluation and treatment.

1. Introduction

Extrauterine leiomyomas, originating from any organ system that contains smooth muscle, are exceedingly rare [1]. Among these, leiomyoma of the vulva is exceptional, accounting for a mere 0.03 % of all gynecological tumors and 0.07 % of all vulvar neoplasms [2]. Vulvar leiomyomas usually affect females of reproductive age [3]. The similarity in symptoms between vulvar leiomyoma and Bartholin's cyst, such as a painless lump and swelling of the area, often leads to misdiagnosis, with the more common preoperative diagnosis being Bartholin's gland cyst [4]. Another diagnostic challenge with vulvar masses is the differentiation between benign and malignant types, as most have a similar appearance. Due to the low incidence of the tumor, there are no established evidence-based diagnostic algorithms or published recommendations for treatment, highlighting the need for further research in this area. Excision of vulvar leiomyomas remains the only viable surgical option for a complete cure [5].

We present a case that initially appeared as a painless vulvar mass, clinically diagnosed as a Bartholin cyst due to its non-tender and soft nature. However, the subsequent surgical exploration revealed a surprising histopathological diagnosis of vulvar leiomyoma, underscoring the unexpected nature of this case.

2. Case Presentation

A 31-year-old woman, para 3, with previous vaginal deliveries, presented with a history of right vulvar swelling for four months. She first noted the mass herself and observed a gradual progression in size over that period. There was no history of discharge, pain, fever, weight loss, or bowel or bladder complaints. She was experiencing discomfort at the perineum, mainly in sitting and walking. She denied any history of sexual trauma, dyspareunia, or other vulvar pathology. She attained menarche at the age of 11 years, and her menstrual cycles were regular with normal flow. During her previous pregnancy one year earlier, she was diagnosed with human immunodeficiency virus (HIV). Since then, she had been followed by an infectious disease physician and was on antiviral medication. Past surgical history was notable for lipoma excision on her left side. She was a non-smoker with an unremarkable social and family history.

Her body mass index was 28.6kg/m2. Abdominal examination was normal. External genital examination revealed a solitary swelling measuring 5 × 4 cm in the lower part of the left labia majora at the site of the Bartholin gland. The mass was soft in consistency, non-tender, and freely mobile without abnormalities of the surrounding skin. There was no palpable inguinal lymphadenopathy. A vaginal and speculum exam revealed no abnormalities, and no adnexal mass was detected. A basic blood workup and pelvis ultrasound were performed, and no abnormalities were observed. The differential diagnoses were Bartholin cyst, lipoma, sarcoma, and aggressive angiomyxoma. At this point, it was decided to proceed with wide local excision.

The patient underwent wide local excision of the mass without any complications, and the lesion was sent for histopathology. Gross pathology examination revealed a well-circumscribed mass measuring 4 cm in its greatest dimension. The outer surface was smooth and intact.

* Corresponding author.
E-mail address: dr.saeed_bardwan@yahoo.com (S. Baradwan).

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Serial sectioning of the specimen revealed no areas of necrosis or hemorrhage. The specimen was left for overnight fixation in 10 % neutral buffered formalin. Histopathology examination with hematoxylin and eosin staining (H&E) revealed benign mesenchymal tumor of a smooth muscle origin (Fig. 1A). It comprised monotonous spindle cells with indistinct borders arranged in intersecting fascicles. Mitotic figures were rare, with less than 5/10 high power fields (HPF). Only the focal area of the lesion showed myxoid cellular changes (Fig. 1B). The tumor had a strong and diffuse positivity for desmin and h-caldesmon (Fig. 1C), and there was also focal and patchy positivity for smooth muscle actin (SMA) (Fig. 1D), estrogen receptors (ER) (Fig. 1E), and progesterone receptors (PR) (Fig. 1F). Although the tumor exhibited morphological characteristics consistent with a leiomyoma, smooth muscle tumors are rare in these regions. Given the patient's immunodeficiency disorder, in situ hybridization (ISH) for Epstein-Barr encoding RNA (EBER) was performed to rule out Epstein-Barr virus (EBV)-associated smooth muscle tumors (EBV-SMTs). The test result was negative, leading to the final diagnosis of leiomyoma.

The patient was discharged home the day following surgery with a follow-up after four weeks. She was discharged on oral analgesia and advice for wound care. She recovered uneventfully and completely and went back to her normal daily activity.

3. Discussion

Leiomyomas are benign soft-tissue neoplasms of mesenchymal origin, typically found in the uterus [6]. Although rare, vulvar leiomyomas are most commonly located in the labia majora and clitoris [7]. Despite their rarity, understanding their morphological and epidemiological characteristics remains a challenge [8]. While the causes of

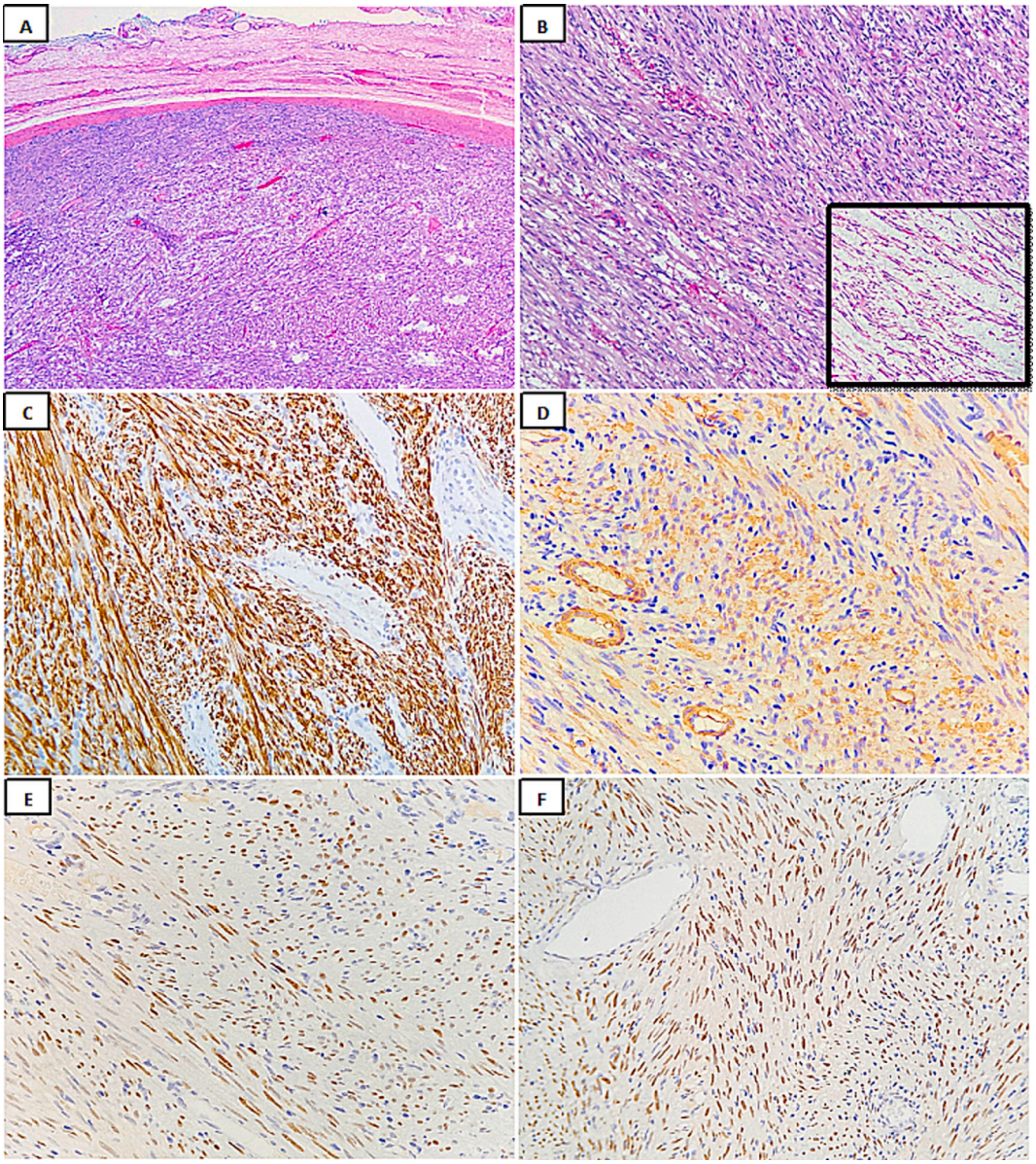

Fig. 1. Histopathology examination was done using hematoxylin and eosin stain (H&E) and immunohistochemistry stains. (A): Microscopic evaluation of the lesion consisted of a single encapsulated and well-demarcated smooth muscle lesion. Note the fibrous capsule rim at the periphery of the lesion (H&E; 4×). (B): The lesion is composed of proliferation of fascicles of spindle cells with cigar-shaped nuclei, scant cytoplasm without increases in mitotic activity and nuclear atypia (H&E; 10×). Focal microscopic area showed hypocellular tumor cells separated by myxoid stroma without mitotic activity, atypia, and necrosis (insert H&E; 40×). (C): Immunohistochemical staining showed strongly positive h-caldesmon in smooth muscle cells (40×). (D): Patchy positive smooth muscle staining for SMA (40×). (E): ER and (F): PR with nuclear positive staining in tumor cells (40×).

vulvar leiomyomas are not fully understood, it is believed that estrogens and progesterone may contribute to tumor proliferation, as fibroids rarely appear before menarche and often resolve after menopause [6]. Additionally, during pregnancy, it is unlikely for the tumor to undergo changes in growth or invasion, as observed by Tavassoli and Norris [9].

The differential diagnosis of a vulvar mass includes soft-tissue sarcoma, Bartholin cyst, fibroma, lymphangioma, and neurogenic tumor. According to the literature review, vulvar leiomyomas can range in size from 0.5 to 15 cm [10]. The present case involved a painless mass in the lower part of the left vulvar region, near the Bartholin gland, which grew to 5 × 4 cm over four months, leading to difficulty in sitting and walking.

Using magnetic resonance imaging and transvaginal ultrasound, it is possible to diagnose vulvar leiomyoma and distinguish it from leiomyosarcoma cm [2]. The diagnosis of a Bartholin cyst in the case reported here was conclusively made based on the patient's medical history and a thorough clinical examination.

Leiomyoma of the vulva should not be assumed to be benign without a histological study. Nielsen et al. found that out of 25 cases of vulvar leiomyomas, 4 were atypical and 5 were sarcomas [11]. This reiterates the need for a histological study, as in the present case. The most reliable way to confirm the diagnosis is through a wide local excision followed by a histopathological examination.

Histologically, vulvar leiomyomas closely resemble leiomyomas found in the uterus. Complete removal of both the leiomyomas and their capsule is crucial to minimize the risk of recurrence. Kothandaraman et al. documented a case of vulvar leiomyoma recurrence four years following primary excision [1]. It is imperative to convey to the patient the potential for recurrence and emphasize the necessity of regular, long-term follow-up.

The primary constraint of the present report is that no pre-operative radiological imaging was done in this case. The clinical findings and location were consistent with a Bartholin gland cyst, and so the patient's management proceeded to wide local surgery. It was a surprise that histopathology subsequently revealed a vulvar leiomyoma.

In conclusion, vulvar leiomyoma tumors are often mistaken for Bartholin's cysts, making diagnosis difficult. The most effective diagnostic procedure is excisional biopsy. Follow-up appointments are essential after treatment.

Contributors

Saeed Baradwan contributed to conception of the case report, acquiring and interpreting the data, drafting the manuscript and undertaking the literature review.

Hassan M Latifah contributed to patient care, undertaking the literature review and revising the article critically for important intellectual content.

Haneen Al-Maghrabi and Abdulmonem M Almutawa contributed to patient care and to drafting the manuscript, and revised and submitted the figures.

All authors approved the final manuscript.

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Patient consent

Written informed consent was obtained from the patient for publication of this case report and the accompanying histopathology images.

Provenance and peer review

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Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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