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Case Report

# A rare case of benign vulvovaginal leiomyoma: Case report and literature review

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ABSTRACT

*Introduction:* Smooth muscle tumors of the vulva are more difficult to diagnose and are frequently mistaken as Bartholin cysts prior to surgery.
*Case presentation:* A 41-year-old female presented with a left vulvar mass that increased in size compared to the previous year. The patient had normal urination and a regular menstrual cycle. The presentation was not associated with dyspareunia, abnormal bleeding, and signs of infection (e.g., fever, vaginal discharge). The history of any sexually transmitted disease was inconclusive. There was also no family history of malignancy. Physical examination showed a solitary swelling mass, measuring 5 × 2 cm in the left labia majora at the site of the Bartholin gland. The mass was firm in consistency, partially movable, and non-tender with no inguinal lymphadenopathy. Histopathology after surgical removal revealed a benign vulvar leiomyoma.
*Discussion:* Labia majora leiomyoma at the site of the Bartholin gland is rather uncommon. Some cases can develop into atypical leiomyoma or even leiomyosarcoma with local tissue infiltration.
*Conclusion:* If the clinical picture is unusual, it is better to send the patient for ultrasound and MRI to exclude other causes followed by performing wide local surgical excision of the mass to allow proper histopathological and/or immunohistochemistry examination to differentiate between benign and malignant tumors.

## 1. Introduction

Leiomyoma of the vulva is a rare and benign smooth muscle tumor (SMT), first described as "myoma" by Rudolf Virchow in 1854 [1,2]. This tumor can derive from smooth muscle within erectile tissue, the dartos muscle, the erector pili muscle, the round ligament, or blood vessel walls [3]. Morphological features of smooth muscle tumors of the vulva are non-specific, often resembling other soft tissue tumors of the vulva, which can lead to misdiagnosis of these tumors as Bartholin cysts, abscesses, or other benign conditions [3,4]. In this study, a rare case of vulvovaginal leiomyoma in the left labia majora is reported. This report has been written in accordance with the SCARE criteria guidelines for case reports [5].

## 2. Case presentation

A 41-year-old female, gravida 1, para 1, presented with a left-sided vulvar mass that increased in size compared to the previous year. There were no signs of infection such as fever or vaginal discharge, dyspareunia, irregular menstrual cycles, abnormal bleeding, or a history of malignancy in the family. The patient also reported no history of disrupted urination or sexually transmitted diseases. However, the patient was experiencing discomfort at the perineum, especially in sitting and walking.

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## 3. Clinical findings

Physical examination revealed a solitary swelling measuring 5 × 2 cm in the lower part of the left labia majora at the site of the Bartholin gland. The mass was firm in consistency, partially movable, non-tender with no inguinal lymphadenopathy.

## 4. Diagnostic assessment

Ultrasonography showed a solid mass in the posterior vaginal wall, normal uterine size, homogenous myometrium, no focal myometrial lesion, no intrauterine fibroid, and normal ovaries with no cystic or solid lesions, and no pelvic free fluid.

## 5. Therapeutic intervention

As a result, the Gyn-Oncologist at the private Baxshin Hospital decided to remove the tumor surgically. A Foley catheter was firstly introduced in the urethra for protecting the latter. Under spinal anesthesia, a two cm incision was made, and a firm encapsulated mass was successfully excised and sent for histopathological examination. Then, the incision was closed using interrupted sutures. The operation took about 45 minutes.

## 6. Postoperative mass examination

A macroscopic examination of the removed tissue showed a gray mass measuring 3.5 × 3.0 × 1.5cm. On the cut section, the mass was solid and gray-white. Microscopically, it was a well-defined, cellular tumor that was composed of bundles of epithelioid spindle cells showing mild pleomorphic nuclei with prominent nucleoli and very low mitotic activity (1–2 mitosis) per high power field (HPF) without necrosis, suggesting vaginal leiomyoma (Fig. 1). Immunohistochemical studies revealed that tumor cells were strongly positive for smooth muscle actin (SMA) and positive for calponin, which confirmed that the tumor was comprised of smooth muscle tumor cells. There was also no evidence of atypia or necrosis. The tumor was diagnosed as vulvar leiomyoma based on histologic features and immunohistochemistry.

## 7. Follow-up and recurrence

The patient was discharged without further difficulties, and there was no sign of recurrence during an eight-month follow-up period.

## 8. Discussion

Though leiomyoma is widespread in the uterus, it is uncommon in the vulva, ovaries, urethra, and urinary bladder [6]. Vulvar myoma can occur at any age and is mostly misdiagnosed until surgical excision is done followed by microscopic and/or immunohistochemical examinations are done (Table 1).

Because both vulvar leiomyoma and Bartholin's cyst share some of the same presenting symptoms, such as a painless lump and swelling of the area, vulvar leiomyoma is frequently misdiagnosed and the most common preoperative diagnosis was Bartholin's gland cyst [7]. The current case also clinically diagnosed Bartholin cyst. The direction of the labia minora and the consistency of the cyst can help distinguish between Bartholin's cyst and vulvar leiomyoma. An everted labia minora and soft consistency of the cyst suggest Bartholin's cyst, whereas an inverted labia minora and a hard consistency of the cyst suggest vulvar leiomyoma [8]. Vulval leiomyoma can also be found hidden by another pathology or present in the clitoris [9,10].

The differentiation between benign and malignant forms of smooth muscle tumor of the vulva is a major diagnostic challenge, as many vulval lesions have similar appearances, making it difficult to identify benign from malignant lesions by gross inspection [11,12]. In both cases, spindle-shaped cells are organized in fascicles with interdigitation in herring bone or whorl formations. Large size and infiltrative margins indicate a higher risk of recurrence and possible malignancy [13]. Recurrence was more likely if at least three of the following features were found: (i) five cm in diameter or larger; (ii) had five or more mitotic figures per 10 HPFs; (iii) an infiltrative margin; (iv) moderate to severe cytological atypia. The neoplasm was considered leiomyosarcoma if all features were present [2,14,15]. The fundamental criterion for distinguishing them is mitotic activity, which is a typical leiomyoma is below (3/HPF) [15]. In our case, the tumor cells were strongly positive for smooth muscle actin and calponin. Also, it was five cm in diameter but with low mitotic activity 1–2/HPF.

Leiomyoma of the vulva should not be regarded as safe unless a histopathological examination is done [16]. Out of Nielsen et al. 25 cases of vulval leiomyomas, four and five were found atypical or sarcomas, respectively. One case even died of a leiomyosarcoma of the vulva [17]. Therefore, it is recommended to surgically remove any vulval mass for proper histological and immunological examination [8–10,17–20]. This should be performed after proper ultrasonographic examination or even magnetic resonance imaging to determine preoperative soft tissue invasion [18] and to differentiate between a Bartholin

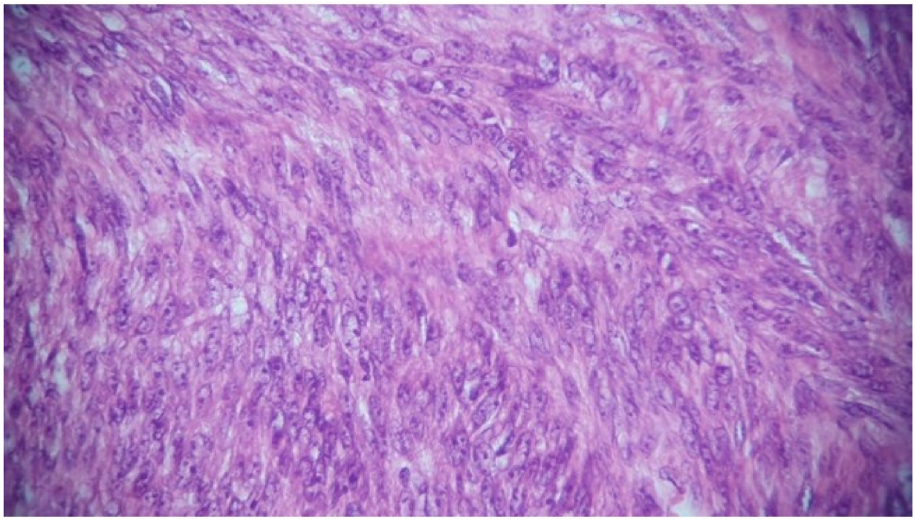

**Fig. 1.** Microscopic examination revealed a well-defined cellular tumor composed of bundles of epithelioid spindle cells showing mild pleomorphic nuclei with prominent nucleoli, diagnosed with vulvovaginal leiomyoma.

**Table 1**
A summary of some studies that reported vulvar leiomyoma.

| Reference | Age in years | Diagnosis before surgery | Investigations before surgery | Treatment | Months of follow-up | Recurrence |
|---|---|---|---|---|---|---|
| Aguilera Martínez et al. [23] | 44 | NR | NR | Surgery | NR | NR |
| Al Azzam et al. [24] | 15 | Bartholinitis | None | Surgery | NR | NR |
| Ammouri et al. [21] | 30 | NR | U/S | Surgery | 24 | No |
| Aneiros et al. [25] | 26 | NR | NR | Surgery | NR | NR |
| Chang et al. [8] | 50 | Bartholin gland cyst | None | Failed oral cephalexin followed by surgery | 2 | No |
| Celik et al. [26] | 73 | None | None | Surgery | NR | NR |
| Fontinele et al. [18] | 14 | Benign mass | U/S | Surgery | NR | NR |
| Francis et al. [27] | 56 | Bartholin gland carcinoma | None | Surgery | NR | NR |
| Guven et al. [28] | 67 | Fibrous histiocytoma or vulval carcinoma | U/S, punch biopsy | Surgery | NR | NR |
| Heller et al. [19] | 30s | Bartholin gland cyst | None | Surgery | NR | NR |
| Hopkins-Luna et al. [29] | 45 | Bartholin gland abscess | None | Failed antibiotics followed by surgery | NR | NR |
| Jang et al. [4] | 45 | Bartholin gland cyst or abscess | U/S | Surgery | 10 | No |
| Kajiwara et al. [30] | 29 | Probable leiomyoma | U/S and FNAC | Surgery | NR | No |
| Katenkamp & Stiller [31] | 71 | None | None | Surgery | NR | NR |
| Khandeparkar et al. [32] | 38 | Spindle cell lipoma or leiomyoma | FNAC | Surgery | 6 | No |
| Kim et al. [33] | 35 | None | U/S, MRI | Surgery | NR | NR |
| Koc et al. [34] | 47 | Bartholin gland mass | MRI | Surgery | 12 | No |
| Kothandaraman et al. [20],[a] | 63 | Malignant peripheral nerve sheath tumor | Wedge biopsy, U/S, contrast CT | Surgery | 8 | NR |
| Kumar et al. [35] | 42 | Ulcerative leiomyoma | Biopsy | Surgery | NR | No |
| Kurdi et al. [2] | 46 | Bartholin gland cyst | None | Surgery | NR | NR |
| Kurdoglu et al. [36] | 39 | Leiomyoma | None | Surgery including laparotomy | NR | NR |
| Nemoto et al. [37] | 40 | Bartholin gland abscess | CT, barium enema, IVP | Surgery | 18 | No |
| Neri et al. [12] | 41 | Bartholin gland cyst or abscess | None | Surgery | NR | NR |
| Ngo & Haertsch [13] | 27 | NR | MRI | Surgery | 30 | No |
| Nielsen et al. [17] | 52 | NR | NR | NR | 189 | No |
| | 34 | NR | NR | NR | 180 | No |
| | 47 | NR | NR | NR | 54 | No |
| | 26 | NR | NR | NR | 41 | No |
| | 47 | NR | NR | NR | 24 | No |
| | 19 | NR | NR | NR | 15 | No |
| | 20 | NR | NR | NR | 120 | Yes |
| | 17 | NR | NR | NR | 11 | No |
| | 45 | NR | NR | NR | 8 | No |
| | 43 | NR | NR | NR | 8 | No |
| | 45 | NR | NR | NR | 5 | No |
| | 41 | NR | NR | NR | 1 | No |
| | 24 | NR | NR | NR | NR | NR |
| | 17 | NR | NR | NR | NR | NR |
| | 42 | NR | NR | NR | NR | NR |
| | 40 | NR | NR | NR | NR | NR |
| Pandey et al. [7] | 20 | Bartholin gland cyst or abscess | None | Antibiotics followed by excision of the mass | NR | NR |
| Pitukkijronnakorn et al. [38] | 25 | Bartholin gland cyst | None | Surgery | NR | NR |
| Reyad et al. [11] | 41 | NR | None | Surgery | NR | NR |
| Siegle & Cartmell [39] | 30 | NR | NR | NR | NR | NR |
| Sloboda & Molnar [40] | NR | NR | NR | Surgery | NR | NR |
| Sultana & Humayun [1] | 45 | Bartholin gland cyst | None | Surgery | NR | NR |
| Taraschi et al. [9,10] | 39 | Bartholin gland cyst | Gestational U/S | Bartholin gland cystectomy followed by excision for the mass | NR | NR |
| Tavares et al. [41] | 32 | Bartholin gland cyst | None | Surgery | 12 | No |
| Topolovec et al. [15] | 26 | Malignant Bartholin gland tumor | NR | Surgery | NR | NR |
| Youssef et al. [3] | 39 | None | U/S, tumor markers | Surgery | NR | NR |
| Zhao et al. [42] | 30 | Bartholin gland cyst | None | Surgery | 14 | No |
| Zhou et al. [43] | 29 | Bartholin gland cyst | None | Surgery | 29 | No |

**Abbreviations:** NR (not reported); FNAC (fine needle aspiration cytology); U/S (ultrasonography); CT (computerized tomography); IVP (intravenous pyelogram).
[a] This case was thought to be a malignant recurrence of neurofibroma that was removed four years before the new presentation.

gland cyst and a leiomyoma [7]. Counseling the patient preoperatively about the risk of recurrence should also be done [18].

The causes of vulvar leiomyoma are still unknown, but estrogens and progesterone are believed to play a role in tumor proliferation, given that fibroids rarely arise before menarche and frequently disappear after menopause [6,21]. It is also important to highlight that Tavassoli and Norris observed it is unlikely that the tumor will change in its growth or invasion during pregnancy [22].

Although this is not the first case of vulval leiomyoma, we performed an extensive literature search to further investigate the prevalence,

diagnostic options, and treatment of this uncommon neoplasm. It is, however, a limitation that our review was not performed systematically.

## 9. Conclusion

Labia majora leiomyoma at the site of the Bartholin gland is rather an uncommon neoplasm that should not be ignored the clinical practice. The tumor may be found a leiomyosarcoma and lead to the death of the patient. Accordingly, careful examination to exclude other differential diagnoses such as Bartholin gland cyst should be done. An unusual clinical picture should warrant sending the patient for ultrasound and/or MRI to exclude malignancy, followed by surgical excision of the mass with a safety margin of the surrounding normal tissue to allow for proper histopathological and immunohistochemistry examinations. Although recurrence is not common, counseling of the patient should be done preoperatively.

## Ethical approval

Approval is not necessary for a case report in our locality.

## Sources of funding

No source to be stated.

## Registration of research studies

According to the previous recommendation, registration is not required for the case report.

## Guarantor

Jeza M.Abdul Aziz is the Guarantor of submission.

## Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

## Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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## Ethical approval

None.

## Authors' contribution

JMA and NTH contributed to the study conceptualization. JMA, NTH, and AMM reviewed the literature and wrote the manuscript. PAM, NGM, HAK, and KST managed and followed the patient up. All authors edited and approved the final version of the manuscript.

## Research registration

None.

## Provenance and peer review

Not commissioned, externally peer-reviewed.

## Declaration of competing interest

There is no conflict to be declared.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103720.

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