International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Solaja OA et al. Int J Reprod Contracept Obstet Gynecol. 2024 Apr;13(4):1038-1041
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20240809

Case Report

Vulval leiomyoma causing coital difficulty: report of two cases

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Received: 06 February 2024
Accepted: 04 March 2024

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ABSTRACT

Leiomyoma is a smooth muscle benign tumor and it is a common benign lesion of the uterus in women of the reproductive age group with a prevalence of about 30%. Though the prevalence of uterine fibroid was 6.5% in Ile-Ife, and 8.5% of gynecological admissions in Ilesha, Nigeria, vulval leiomyoma is very rare and often misdiagnosed as Bartholin cyst or with other differentials like lipoma, etc. It is usually not considered a differential of vulval masses, but with detailed examination and with the use of investigative tools, the diagnosis can become clearer. This paper presents 2 cases of histologically diagnosed vulval leiomyoma seen at the Obafemi Awolowo University Teaching Hospitals Complex (OAUTHC) between January 2017 and December 2021 with both patients complaining of coital difficulty. During this time, the prevalence of this lesion was calculated to be 0.092% of gynecology admissions and 0.17% of gynecological surgeries done in the hospital.

Keywords: Vulval leiomyoma, Misdiagnosis, Coital difficulty, Histology, Ultrasound scan

INTRODUCTION

Leiomyomas also known as myomas or fibroids are benign tumors of smooth muscles, that can occur anywhere smooth muscles exist in the body, though it is usually discussed in relation to the uterus, it does rarely exist outside the uterus.[1] Leiomyomas are a very common disease of the uterus and its prevalence is about 30% amongst women of the reproductive age group. The prevalence of uterine fibroid was put at 6.5% in this hospital, and it accounted for 8.5% of gynecological admissions in Ilesha, Nigeria.[2-5] Its prevalence could be as high as 70-80% following histology of uteri after hysterectomy and about 51% of uterine leiomyomas can be diagnosed on ultrasound.[6,7] It is rarely seen in other parts of the body like the ovary, fallopian tube, vagina, urethra, urinary bladder, and vulva.[3,8] Wamsteker et al recently classified leiomyoma into 9 classes (i.e., from class 0-8), and this classification was later adopted by FIGO.[9,10] By this classification, vulval leiomyoma belongs in class 8 (i.e., others; specify). We, present these two cases of vulval leiomyoma to add to the body of knowledge available on the subject matter.

CASE REPORT

Case 1

Mrs. A.O., a 43-year-old P1+1 (1 alive), a known human immunodeficiency virus (HIV) positive patient on highly active antiretroviral therapy (HAART), presented with a 5-year history of vulval swelling and 3 years history of

infertility. The swelling was initially about the size of the distal phalanx of her index finger but had grown to the present size of 8 cm by 6 cm pedunculated mass, with the stalk extending from about the 5 o'clock region down to the posterior fourchette. The mass was firm (not cystic) and this had made coitus difficult. A diagnosis of vulval lipoma was entertained though she had been referred to the gynecology unit with a diagnosis of Bartholin's cyst. She had been diagnosed HIV positive and had been on HAART. At the time of presentation at the clinic, she had been on HAART for 11 years and the viral load was undetectable. She had investigations done which were all normal and the abdominopelvic ultrasound did not reveal any uterine mass. She had an excision of the vulval mass under spinal anesthesia and the mass was pale and well-circumscribed. The suspicion of leiomyoma was entertained at surgery. The histology revealed a benign and admixed proliferation of smooth muscle cells and fibro-collagenous elements which are altogether arranged in interlacing fascicles and whirling areas of hyalinization are present with a diagnosis of leiomyoma as seen in Figure 1. Consent to write up this presentation was gotten from the patient.

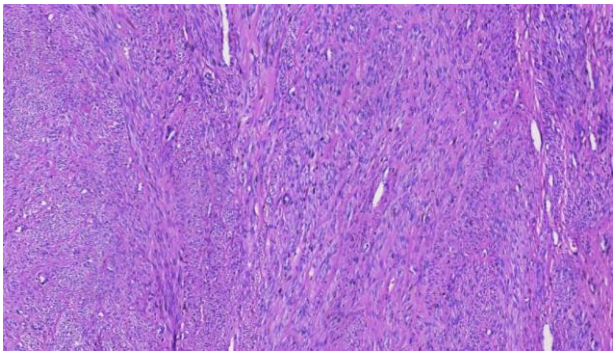
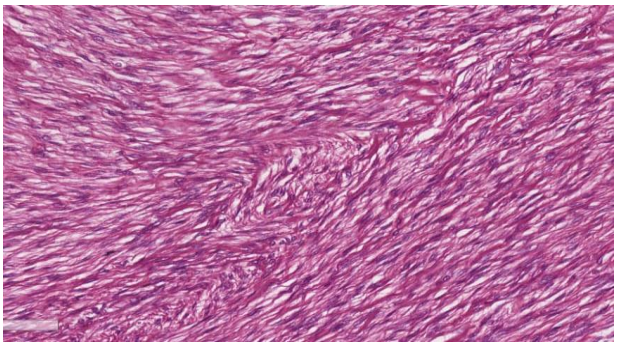


Figure 1: Section shows tissue composed of fascicles and bundles of benign spindle-shaped cells with abundant cytoplasm and plump nuclei.

Case 2

Mrs. A. T., a 37-year-old P5+0 (3 alive) presented with a month's history of right vulval swelling. The swelling measured about 3 cm by 3 cm in its widest diameter at the 9 o'clock position involving the labia minora (Figure 3a-c). There was no associated pain, ulceration, itching, or discharge but there was an increasing discomfort during sexual intercourse, which necessitated her presentation. A diagnosis of suspected Bartholin's cyst was made, and this was also in keeping with her referral from the outpatient department. Physical examination was essentially normal, there was no abdominal/uterine mass palpated. She had blood investigations done which included full blood count, fasting blood glucose, and two hours postprandial and the results were within normal limits. Abdominopelvic ultrasound done did not reveal concurrent uterine fibroid. She was scheduled for marsupialization at the day case theatre. During and after the surgery, the excised mass grossly looked pale, well circumscribed, and whorled and these findings suggested that the diagnosis could be leiomyoma. The sample was sent for histology which revealed benign proliferation of smooth muscle fibers disposed in interlacing fascicles within a fibro-collagenous stroma and a diagnosis of leiomyoma was made as seen in Figure 2. Consent to write up the case was given by the patient.



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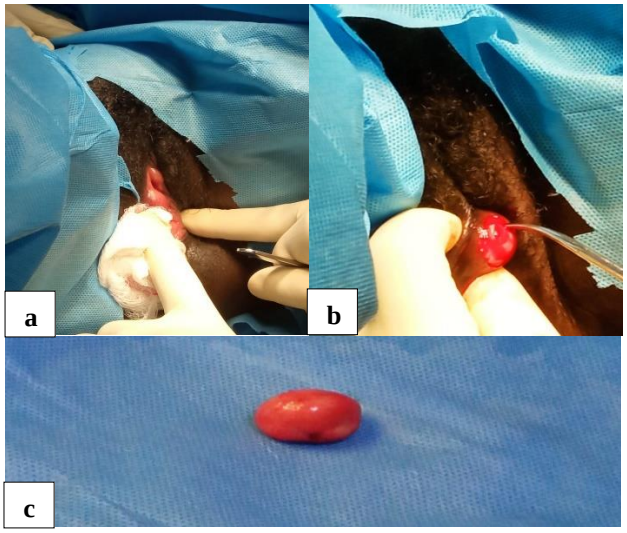

Figure 3: Case 2 (a) the vulval mass, (b) the surgical excision of the mass, and (c) the excised mass sent for histopathology.

DISCUSSION

The vulva is a region of the external female genitalia that is very essential to the well-being of the female sex. It is an area that is bound by the mons pubis anteriorly, perineal body posteriorly, genital crural folds laterally, and hymen medially. It is an important part of female sexuality and the female genital tract, and it houses the labia majora, the clitoris, the vestibule, the labia minora, and the hymen, as such, any lesion or pathology that affects the area will affect the sexual activity of the individual.[1,11]

Thus far, about 160 cases of vulval fibroids have been reported in English literature, many of which were case

reports of this very rare benign smooth muscle tumor.[2,12] Vulval myomas account for 0.03% of all gynecologic neoplasms and 0.07% of all vulval tumors as revealed by literature, in another review, vulval leiomyoma accounts for 4.2% of the 95 patients with cutaneous leiomyomas.[1,2,12,13] There were only 2 histologically diagnosed vulval fibroids between 2017 and 2021 in this hospital. There were 2,171 gynecological admissions and 1,155 gynecological surgeries (both major and minor) during this period putting their prevalence at 0.092% and 0.17% respectively.

In a review by Sun et al, the age range of patients that developed vulval leiomyoma was between 15–73 years, but more commonly between the 4th and 7th decade of life.[1,2] The patients in this review were within the 4th and 5th decade of life. The black race has also been associated with the occurrence of the swelling, though recurrence has been reported but family history has not been linked as a risk factor.[1,14,15] The pathophysiology of vulval leiomyoma has been linked with the smooth muscle of the erectile tissue around the vulva, or from vascular walls or due to transmigration through the round ligament, making the clitoris a common site of development of vulval fibroid.[1,16]

Patients usually present with solitary, painless vulval mass that usually grows slowly, though may become associated with some pain as it increases in size. The size ranges between 0.5 -15 cm, the longer the duration of the mass before presentation to the hospital, the bigger the mass as seen in this series.[17] There could be associated difficulty in walking, sitting, and discomfort during sexual intercourse.[1] One of the patients had presented with complaints of inability to get pregnant and with a history confirming inadequate sexual activity as a plausible causative factor. Both patients actually had discomfort/pain during sexual intercourse and this could have led to marital disharmony.

Vulval leiomyomas are usually painless, solitary, and well-circumscribed mass, making the diagnosis of a Bartholin's cyst a close differential but findings of an inverted labia minora and a mass firm in consistency support the diagnosis of vulval leiomyoma in variance to Bartholin's cyst where the labia minora is everted and swelling is cystic.[1,2,8,15,17] Other differentials include benign and malignant entities such as; atypical leiomyoma, leiomyosarcoma, dermatofibrosarcoma, fibroma, ectopic breast tissue, and lipoma.[1,16] The diagnosis of lipoma was made in the first case presented, considering the presentation and examination of the mass until the gross appearance of the mass after excision and histology findings revealed otherwise.

Transperineal ultrasound has been useful in making diagnoses once vulval leiomyoma or another lesion is suspected. Magnetic resonance imaging (MRI) has been used as a potent diagnostic tool but it's expensive. It is used to differentiate such lesions and especially between benign and malignant lesions, computerized tomography scan can also aid in diagnosis.[1,8,12,15,18] The gross appearance of the mass after excision in these patients gave an inclination towards leiomyoma.

The treatment of choice is the excision of the tumor with some of the surrounding normal tissue reducing the rate of recurrence. The histopathological diagnosis of the excised mass is compulsory to confirm the diagnosis. There are reports of the use of selective oestrogen receptor modulators in patients that presented with recurrent mass after re-excision.[1,2,8,14,17]

CONCLUSION

Vulval leiomyomas are very rare tumors and the gynecologist needs a high index of suspicion from the history, coupled with a painstaking careful examination of the mass before the suspicion could be entertained. The histopathology of the excised mass cannot be overemphasized to confirm the diagnosis and counsel patients appropriately. Surgical excision is the mainstay of treatment and it is also important to follow-up these patients.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Solaja OA, Fasubaa OB, Ajenifuja OK, Adepiti CA, Adedeji NO, Adefidipe AA, et al. Vulval leiomyoma causing coital difficulty: report of two cases. Int J Reprod Contracept Obstet Gynecol 2024;13:1038-41.