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Case Report

A Rare Case of Vulvar Fibroid with Cystic Degeneration Mimicking Bartholin Cyst

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Abstract: Vulvar swelling is a common condition in the reproductive age group of women. The most common vulvar swelling is a Bartholin cyst or abscess, which is unilateral. The rarest presentation of vulvar swelling is Leiomyoma, which is like a Bartholin cyst. The common symptom of vulvar Leiomyoma is discomfort during walking, voiding, or coitus, otherwise asymptomatic. Here we present a rare case of vulvar fibroid mimicking a Bartholin cyst as the patient presented with swelling in the vulva, which was gradual in onset and associated with pain. Following which patient was investigated further and managed with surgical excision and marsupialization. Therefore, we should also rule out the rarest form of any condition. Here, we provisionally diagnosed a vulvar leiomyoma as a Bartholin cyst, which was confirmed by Histopathology of excised specimen.

Key Words: Bartholin cyst, Leiomyoma, Labia majora, Spindle cells, Excision

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1. INTRODUCTION

Smooth muscle neoplasms are the most common tumors of the female genital tract. Among these, the most common benign tumor is Uterine leiomyoma [1]. However, Leiomyoma can develop at any site where smooth muscle cells are present such as the vulva, vagina, ovaries, urinary bladder, urethra, round ligaments, uterosacral ligaments, inguinal canal, and retroperitoneum[2]. Vulvar Leiomyoma is a unique presentation in about 0.03% of all gynecologic neoplasms and 0.07% of all vulvar tumors[3]. The most common site is the labia majora. Vulvar leiomyomas are often encountered around 40-50 years of life. These tumors arise from smooth muscle within erectile tissue, wall of blood vessels, and round ligament. Also, very rarely arise from dartos muscle, arrector pilli muscle cells in Bartholin gland[4]. These tumors usually occur as a single, well-circumscribed mass seen originating from the anterior midline wall and occasionally arise from the anterior and lateral vaginal walls. It often presents as spindle-shaped cells, but other histological variants like epithelioid tumors are also seen. Clinical symptoms include painless mass, pain, itching, and erythema. The clinical diagnosis of vaginal Leiomyoma necessitates a high index of doubt because the tumor could easily be misdiagnosed for cystocele, urethrocele, Skene duct abscess, Gartner duct cysts, urethral diverticulum, vaginal cysts, Bartholin gland cysts, or vaginal malignancy[5]. Transperineal ultrasonography is used to diagnose vulval Leiomyoma. A benign and malignant variant can be differentiated with the help of magnetic resonance imaging. Surgical excision is the treatment of choice in all smooth muscle tumors of the vulva. The exterior surface of the specimen was ragged and tan-colored. The cross-section of the specimen was solid with a "whorled" appearance[6]. Histopathology usually confirms the diagnosis. Histological examination revealed a well-circumscribed tumor composed of intersecting fascicles of uniform spindle-shaped cells with elongated ovoid nuclei and fairly abundant eosinophilic cytoplasm. Three principal histological variants of vulvar leiomyomas have been recognized: spindled, epithelioid, and myxoid or myxohyaline[7]. This spindled pattern is a relatively common type with a fascicular proliferation of spindle-shaped cells with oval to elongated nuclei and richly eosinophilic cytoplasm[7]. When immunohistochemically stains are done, vulvar leiomyomas stain positive either for estrogen or progesterone receptors and rarely both. Thus, treatment with receptor modulators in adjuvant to surgery may be beneficial[8]. Vulvar Leiomyoma is often misdiagnosed as a Bartholin cyst due to its site of occurrence and the lesion's consistency. Here a case of 49 years old woman presented with vulvar swelling which was unilateral in the region of labia majora, and had complaints of swelling, which was gradual in onset with pain. The patient also had a history of similar complaints and was managed surgically. The patient was diagnosed with Bartholin cyst due to its similar history in the past, its location of origin at present, its complaints, and the consistency of swelling. Following this patient underwent surgical excision, and the specimen was sent for Histopathology, where it was diagnosed as vulvar leiomyoma. The patient was followed up carefully for a few months as it reoccurred. Although it's an uncommon condition of vulvar swelling, Leiomyoma should always be in differential diagnosis until its proved to prevent unnecessary investigation and treatment of the disease. This article discusses the pathology, diagnosis, differential diagnosis, investigation, and treatment of vulvar Leiomyoma.

2. CASE REPORT

A 49 years old parous and sterilized woman presented to the hospital with a complaint of swelling in the genital region for five months. The swelling presented with slow progression over the period, with the complaint of pain over the swelling for 5 days (figure 3). There were no complaints of lower abdominal pain, discharge, fever, dysuria, or weight loss. A swelling that measured 7 × 4 cm was present in the right labial area during local examination with no warmth, tenderness, or redness. The provision diagnosis was Bartholin cyst due to its site of occurrence and mass consistency.

3. MEDICAL AND SURGICAL HISTORY

The patient also had a history of surgery for the right Bartholin cyst. Therefore, she was advised to go for surgical management. Therefore, preoperative evaluation for excision was carried out. As a result, the patient was diagnosed as hypothyroid and started on Thyronorm 75mcg.

3.1. Family History

No relevant history in her family.

3.2. Observation

The patient was admitted for observation and to evaluate the cause and further line of management.

3.3. Special Test and Investigations

The patient was evaluated preoperatively with routine basic blood investigations like complete blood count, random blood sugar, thyroid function test, and ultrasound abdomen (Figures 1 and 2). All investigations were found to be within normal limits.

3.4. Diagnosis

Provisionally patient was diagnosed with a Bartholin cyst. The final diagnosis will be based on the Histopathology of the lesion.

3.5. Intervention/Treatment Plan

They were planned to proceed with surgical excision and biopsy. Hence the patient is taken up for excision under anesthesia. The incision made over the swelling at the mucocutaneous junction showed a fleshy mass measuring 6× 4 cm. The mass was removed in toto and sent for a histopathological examination.

Fig 1: USG whole abdomen

Fig 2: USG whole abdomen

Fig 3: Preoperative image of vulvar leiomyoma

Fig 4: Excised Mass of Vulvar Fibroid

The Postoperatively patient had no complications. The patient was discharged on postoperative day 2. The gross specimen of grey-white soft tissue mass measures 6 × 4 × 2.5cm with irregular margin and variable in consistency (figure 4). Microscopic examination revealed irregular margins showing a spindle cell neoplasm arranged in a whorl pattern. The nuclei are spindle-shaped without pleomorphism and atypia. Cystic degeneration was noted in peripheral areas (figure 5). Special stain compatible with Leiomyoma. The confirmatory diagnosis was benign vulvar Leiomyoma.

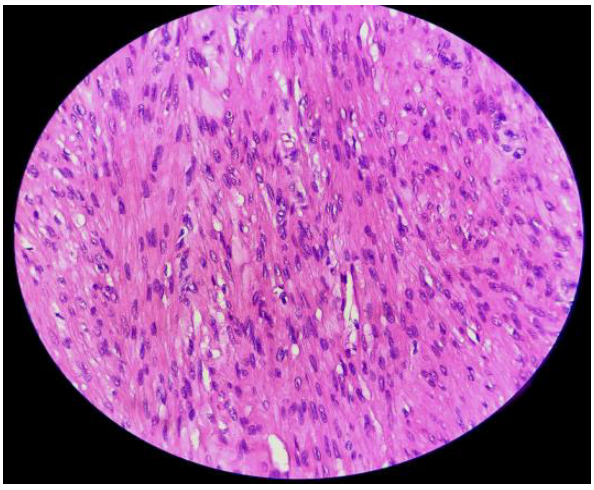

Fig 5: Showing microscopic feature of the specimen showing spindle cell neoplasm arranged in whorls

3.6. Follow Up

The patient was reviewed after two weeks and found healthy.

4. DISCUSSION

The most common benign tumor is Uterine Leiomyoma in reproductive life, but it can rarely occur in the vulva also. So far, around 160 vulvar fibroids have been reported[9]. Vulvar Leiomyoma arises from vulvar erectile smooth muscles or transmigrated cells through round ligament[10]. The mean age of occurrence of the tumor varies from 30 to 50 years of age. The average size of the tumor ranges from 0.5 to 15cm and usually occurs as solitary lesions[11]. Vulvar leiomyomas are usually asymptomatic but occasionally present with discomfort due to swelling. Patients usually present with difficulty in walking, sitting, or having coitus due to an increase in the size of the tumor. The mass gradually causes pain due to peripheral nerve stimulation by huge leiomyoma[12]. Imaging techniques should be performed to confirm the presence, site of occurrence, features, and size of the Leiomyoma if suspected with patient history or local examination. Ultrasound is commonly used to diagnose vulvar leiomyomas because of its affordable, high accessibility, and most reliable tool for uterine and extrauterine leiomyomas[13]. In the initial evaluation, vulvar leiomyomas are often diagnosed as Bartholin cysts or abscesses mistakenly. The confirmatory diagnosis will be obtained only with a histopathologic examination of the specimen. The most common changes in vulval Leiomyoma are degenerative changes[14]. Our case had cystic degenerative changes in Histopathology. Due to the high recurrence rate, Close follow-up is required. The treatment of choice is Excision along with a few surrounding normal tissue [15]. The macroscopic features of cystic consistency make diagnosing Leiomyoma and Bartholin cysts difficult. (Table 1)Always reconsider the diagnosis if labia minora is inverted, variable, or firm, with the consistency of the tumor and intact hymen[16]. The final diagnosis should be made only with histopathologic examination. Vulvar Leiomyoma is usually a painless mass that increases in size gradually and is usually mistaken as a Bartholin cyst or abscess. The usual presentation of Bartholin cyst is tender swelling with a history of fever and other inflammatory signs.

5. INVESTIGATIONS

Transperineal ultrasound, pelvic CT scan, and Magnetic Resonance Imaging are other investigations to confirm the diagnosis[18]. For vulvar smooth muscle tumors, the treatment of choice is always Surgical excision, with histopathological confirmation as a must to rule out whether it is benign or malignant. Long-term follows up is advised in all cases.

Table 1:Comparison of Vulvar Fibroid with Bartholin Cyst

Features	Vulvar fibroid	Bartholin cyst
Occurrence	40-50 years	In reproductive age groups
Pathology	Benign smooth muscle neoplasm	Infective etiology
Clinical presentation	Discomfort predominantly. Pain only if it gets infected	Predominantly painful.With all signs of infection
Location	Labia majora commonly.	Same
Investigation	Histopathology and ultrasound	Clinical examination
Treatment	Does not respond to antibiotics. Surgical excision is the treatment of choice	Antibiotics-respond well If it fails, excision and marsupialization
Image for comparison	 Fig 3 : Preoperative image of vulvar leiomyoma	 Fig 6: Picture of Bartholin cyst[19] Source: Clinical Pathology of Bartholin's Glands: A Review of the Literature in current urology journal. doi: 10.1159/000365683

6. CONCLUSION

Uterine Leiomyoma is the most common, but extrauterine Leiomyoma, like vulva, is an extremely rare condition often misinterpreted as a Bartholin cyst. Uncommon sites of Leiomyoma include the urinary bladder, urethra, ovaries, and vulva. Differential diagnoses are Bartholin cysts, lipoma, leiomyosarcoma, and elephantiasis. So when a reproductive age group presents with unilateral swelling in the vulvar region with firm or variable consistency, Leiomyoma should be kept as a differential diagnosis. Ultrasound or MRI may help in distinguishing the Leiomyoma from the Bartholin cyst. Mass excision should be the treatment so as not to miss any malignancy. Only Histopathology is used to confirm the diagnosis. Long-term follow-up is a must post-surgical excision.

7. AUTHORS CONTRIBUTION STATEMENT

Dr.Logeswari. BM Manuscript writing and Critical analysis and Dr.Mahalakshmi-Data collection.

8. CONFLICT OF INTEREST

Conflict of interest declared none.

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