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RECURRENT LEIOMYOMA OF THE VULVAR: A CASE REPORT

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ABSTRACT

We present a rare case of recurrent leiomyoma of the vulvar in a 25-year-old P2 who had the primary surgery at a private clinic. The swelling, which was misdiagnosed as Bartholin cyst was discovered at excision to be a firm mass. She noticed a similar swelling on the same site four months after the excision. The histopathology of both lesions were the same.

INTRODUCTION

The vulvar is an uncommon site for leiomyoma. [1, 2] It is responsible for 0.07% of all leiomyomas.[3] Vulvar leiomyoma usually presents as a mass and frequently misdiagnosed as Bartholin gland cyst.[4] Recurrence of vulvar leiomyoma is indeed rare. We present a case of a 25-year-old P2 who developed recurrent vulvar leiomyoma four months after excision.

CASE REPORT

She is a 25-year-old P2 woman who was referred from a private clinic with a one-year history of recurrent vulvar mass. Her last childbirth was 2 years prior to presentation. She noticed the swelling 4 months after she had excision of a similar albeit smaller mass. It grew rapidly over 8 months to its present size. The primary excision was done under local anaesthesia in the private clinic as the mass was misdiagnosed as a Bartholin's gland cyst. There was history of dragging sensation and pain when she sat for long. She also complained of difficult and painful coitus. There was no associated pruritus, no abnormal vaginal discharge or bleeding. There was no associated swelling in other parts of the body. She had no family history of similar swelling. Attached to her referral letter was a histopathology report which

showed typical-type leiomyoma with hyaline degeneration reported in December 2017.

On examination, she was found to be a young lady with a stable general condition. Abdominal examination was unremarkable. Vulvar examination showed a mass measuring 6cm x 5cm on the medial aspect of the right labia minora. The mass had no differential warmth and non-tender. It was firm in most areas except at its lower part where it was cystic. It was not attached to the overlying skin and freely mobile in all directions. Other aspects of the vaginal examination were unremarkable. Transvaginal probe on the swelling showed an irregular solid mass with a cystic component and a colour score of 2 (Figure 1a & b). Her full blood count, urinalysis and renal function test were within normal limits. She had blood grouped, typed and saved. She was counselled and she consented to excisional biopsy. The surgery was performed under spinal anaesthesia (Figure 1c & d). A 5cm incision was made on the mucocutaneous junction. The mass was enucleated with ease (Figure 1e). Haemostasis was secured using diathermy. The deep space was obliterated and skin apposed. Blood loss was estimated at 1L. The mass excised was encapsulated with nodular surface, measuring 8cm x6 x4cm. The cut section showed whorl pattern with fibroid nodules (Figure 1f). She had several bouts of vomiting on the second post-operative day which was controlled with promethazine. She had no complaints on follow up one week after surgery (Figure 1g) and the histopathology report was consistent with typical-type leiomyoma (Figure 1i). She was counselled on the need for follow-up to monitor for recurrence.

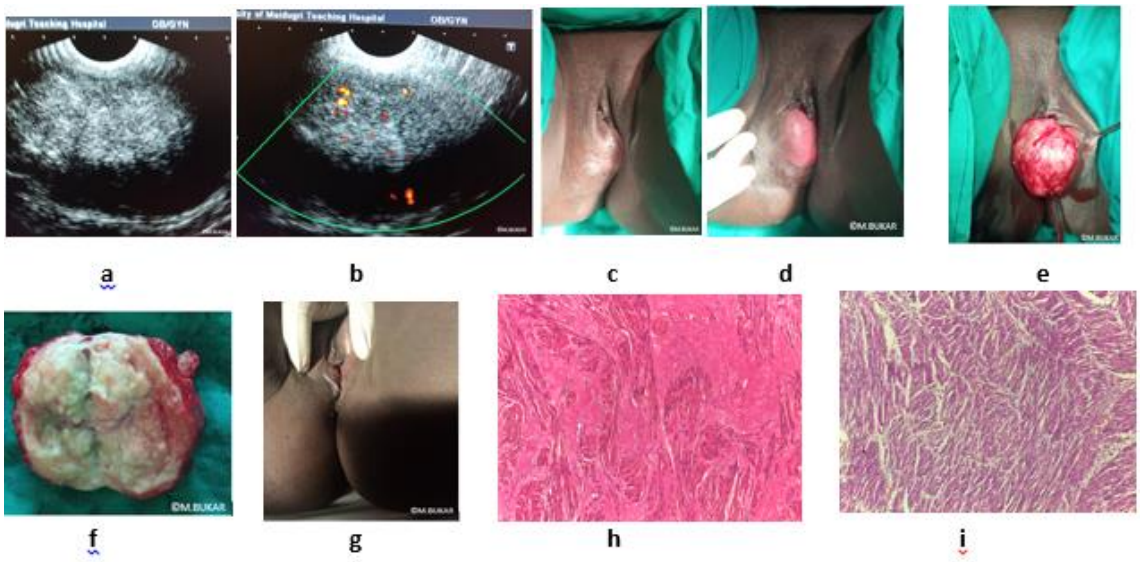

Figure 1: Recurrent Leiomyoma of the Vulvar

A trans-vaginal probe was placed on the swelling to obtain images a & b. Note the irregular solid mass, the cystic area beneath it and the colour score of 2 in b. Image after draping (c), showing the inner aspect of the mass (d) and after partial enucleation (e). Note the cut surface (f) and the vulvar a week after excision (g). Figure 1h is the photomicrograph of the primary surgery in December 2017 and figure 1i is the photomicrograph of the specimen after recurrence in December 2018.

DISCUSSION

Vulvar Leiomyoma is commonly misdiagnosed as Bartholin gland cyst/abscess[4] as in the index case. This underscores the need for meticulous examination paying attention to its consistency and location. It is a rare condition responsible for 0.07% of all leiomyomas.[3] Only few cases of recurrence have been reported. When leiomyoma recurs, it does so several years after excision making it unpredictable.[1] This patient however had recurrence just 4 months after excision probably because the excision was performed under local anaesthesia. An unexpected finding in a private hospital by a general practitioner may lead to incomplete excision and early recurrence as seen in the index case.[6] It has been reported that lesions greater than 5cm tend to recur years after primary excision and that incomplete excision is the most important predictor of recurrence.[6] The mass after the primary excision measured 6 x 4 x 2.5cm.

General examination findings are usually unremarkable. The diagnosis of recurrence was not difficult as the histopathology report of the first excision was available. Even at that the consistency of the mass was not the typical feel of leiomyoma. Trans-perineal sonography is useful in diagnosing and differentiating Bartholin gland pathology from leiomyoma of the vulva.[1] In this case a transvaginal probe was applied on the swelling to obtain images which showed both solid and cystic areas. T2- weighted Magnetic Resonance imaging can differentiate leiomyoma from leiomyosarcoma.[1] Histopathology reports of both the first and second surgery showed typical/spindle-type leiomyoma, which is the commonest type reported in literature.[7] Both histopathology reports did not reveal any features of malignancy. The proposed diagnostic criteria for leiomyosarcoma of the vulva are: tumour greater than 5cm in its greatest dimension, infiltrative margin, more than 5 mitotic figures per 10hpf and presence of moderate to severe atypia. Presence of at least three of these criteria gives the diagnosis of leiomyosarcoma. When two of the criteria are met, it is atypical leiomyoma, presence of a criterion confirms a benignity.[8] Additionally, coagulative necrosis with the aforementioned criteria makes the diagnosis of leiomyosarcoma most likely.[9]

Reports have shown that vulva leiomyoma may stain positive for Estrogen and Progesterone receptors, making receptor modulators adjuvant therapy to surgery.[4,7] Vulvar leiomyoma having strong Estrogen receptor positivity can recur. This patient who had recurrence just 4 months after may benefit from adjuvant therapy but the lack of immunohistochemistry in our centre for determining the ER makes it difficult to prescribe such medications.

The main stay of treatment is excision [5] which was done under local anaesthesia during the first surgery when it was misdiagnosed as Bartholin cyst. Inquiry from the primary surgeon indicated that the blood loss was insignificant. Though the mass was enucleated with ease during the second excision, the blood loss amounted to about 1L. This may be explained by the size and adhesions from previous surgery. It could also be akin to vulvar myomectomy without tourniquet. Unlike in uterine fibroid where there are protocols for preventing primary haemorrhage during myomectomy, there is no specific method in the literature for preventing such during vulvar leiomyoma excision.

The patient was seen a week after surgery with no complaints. The need for regular follow-up to detect at the earliest any recurrence was emphasized to her. It is important to differentiate between Bartholin cyst which is cystic and a leiomyoma which is firm but may be soft when it has undergone degenerative change like in the index case. Everted labia minora in the

former and inverted labia minora in the latter is an important distinguishing feature while trans-perineal ultrasound is helpful in differentiating the two.

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