Case report

Phyllodes tumour of the labia minora

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Accepted 21 October 2019

SUMMARY

A 33-year-old woman presented for a preoperative examination prior to an upcoming operative hysteroscopy. During the examination, a firm 1 cm mass in her right labia minora was noted. The mass was excised in the operating room without difficulty. Pathological examination revealed a benign phyllodes tumour of the vulva. Phyllodes tumours are uncommon fibroepithelial tumours primarily found in the breast although rarely may present as a vulvar lesion. Phyllodes tumours of the vulva are rarely reported in the literature, with only 17 previously reported cases. This case represents the first reported case of a phyllodes tumour occurring in the labia minora. While most of these tumours are benign, it is important to keep these and other rare tumours in the differential diagnosis of vulvar masses. Even with benign tumours, continued surveillance for recurrence should be performed.

BACKGROUND

Vulvar lesions are both a common presenting complaint and incidental finding for practicing women's health physicians. These lesions can range from benign cysts to invasive vulvar cancer. Mammary-like tissue can be present in the vulva and was first reported by Hartnug *et al* in 1872.¹ Since then, many benign and malignant lesions similar to breast tissue have been reported in the vulva, such as lactating adenomas, hidradenoma papilliferum, hidradenocarcinoma papilliferum, fibroadenoma, extramammary Paget's disease and phyllodes tumours. Herein, we present a case of a benign appearing labial cyst which was pathologically determined to be consistent with a rare phyllodes tumour. Seventeen previously reported cases of vulvar phyllodes tumours have been reported in the literature^{2–12}; however, our case represents the first reported tumour located in the labia minora. As these tumours are rarely found in the vulva, treatment and prognosis is less established¹³; we discuss the differential diagnosis, treatment and surveillance to be considered for these rare tumours.

CASE PRESENTATION

A 33-year-old nulligravida woman undergoing evaluation for primary infertility was scheduled for hysteroscopic evaluation secondary to a uterine abnormality discovered on hysterosalpingogram and confirmed with saline infusion sonography. On the day of her preoperative appointment, a genitourinary exam was notable for a clearly defined firm, mobile, non-painful, 1 cm mass located in mid-portion of her right labia minora. The patient could

not recall how long the mass had been present but did not feel like it had grown much since she first discovered it; however, she desired its removal.

The labial mass was excised in the operating room at the time of her scheduled operative hysteroscopy. A small linear incision was made on the medial portion of the labia minora and the lesion was easily removed intact. The mass was noted to be a solid, rubbery consistency and without any cystic component. Gross pathological examination demonstrated a 1.5×1.0×0.8 cm, white-grey tumour. Microscopic examination reported a well-circumscribed fibroepithelial lesion with a predominantly leaf-like pattern and hyalinised stroma with no increased stromal cellularity consistent with a benign phyllodes tumour (figure 1).

OUTCOME AND FOLLOW-UP

The patient recovered well from surgery and has had no signs of a persistent or recurring labial mass. She continues in her pursuit of infertility treatment.

DISCUSSION**Clinical and histological features**

Phyllodes tumours are primarily found in the breast although they may rarely present in the vulva as solitary, well-circumscribed, skin-coloured tumours which are freely mobile and non-tender to the touch. Their rate of growth is variable, with some presenting with recent rapid growth and others being present with little growth for many years.^{13 14} The majority of lesions are located in the labia majora but have been described to also occur in the interlabial sulcus, periclitoral and on the mons.¹³ Our case is the first reported case, which we could find, of a phyllodes tumour occurring in the labia minora.

Phyllodes tumour of the vulva and phyllodes tumour of the breast share similar characteristic histological features. Phyllodes tumours are defined by leaf-like projections into glandular lumens consisting of epithelium adjacent to a hypercellular stromal component (known as the cambium layer).¹⁴ Ectopic mammary tissue in the vulva was long thought to represent the caudal remnants of the mammary ridges. However, recent studies are proposing that anogenital mammary-like glands are a normal constituent of the anogenital area.^{13–15}

Differential diagnosis

Clinically, phyllodes tumours closely resemble, and may be mistaken, for other benign lesions such as epidermoid cysts, fibromas, lipomas, fibroadenomas, hidradenomas and syringomas.^{13 14}

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To cite: Fujii DT, Korzen CA, Levine TC, *et al*. *BMJ Case Rep* 2019;**12**:e229917. doi:10.1136/bcr-2019-229917

Figure 1 (A) The lesion is well-circumscribed with characteristic leaf-like projections and associated heterotopic breast parenchyma (arrows) (H&E, original magnification 2×). (B) Beneath the epithelial-lined clefts is a layer of increased cellularity known as the cambium layer (arrow). Normal appearing breast parenchyma can also be seen (right) (H&E, original magnification 10×). (C) Beneath the epithelial-lined clefts is a layer of increased cellularity known as the cambium layer (arrow) (H&E, original magnification 40×). (D) The clefts are lined by epithelial cells (H&E, original magnification 40×).

Therefore, proper diagnosis of a phyllodes tumour requires excision with pathological tissue examination. Epidermoid cysts are common non-tender and mobile cutaneous lesions that are distinguished by keratinaceous debris. Fibromas are firm, smooth masses that primarily contain collagen with scattered fibroblasts. Lipomas are typically softer when palpated and have a similar appearance to normal adipose tissue on histology.

Additionally, phyllodes tumours can be difficult to distinguish from other biphasic tumours, due to overlapping histological features. Fibroadenomas may rarely share the characteristic leaf-like architecture of the phyllodes tumour³; however, the stromal tissue in a fibroadenoma is typically paucicellular and lacks cellular condensation adjacent to the epithelium.¹⁴ Papillary hidradenomas originate from apocrine sweat glands and share a similar complex papillary growth pattern with phyllodes tumours though they exhibit a distinct double layer epithelial lining and lack a prominent stromal component. Chondroid syringomas arise from the eccrine sweat glands and are identified by areas of sebaceous and squamous differentiation.³ Given the stromal component of phyllodes tumours, they must also be discriminated from other soft tissue tumours that may present on the vulva, such as fibromatosis, neurofibromas, nodular fasciitis, angiomyxomas and angiomyofibroblastomas, although most of these tumours can be excluded if there is an epithelial component.²

Treatment and prognosis

The mainstay of treatment is excision with clear margins, although this may be difficult to accomplish while still preserving an acceptable cosmetic outcome. Given the location of this tumour in the labia minora, a wide excision with a typical 1 cm margin was not possible in order to have an acceptable cosmetic outcome. No other primary treatment modalities have been reported.¹³ The prognosis of a phyllodes tumour is difficult to predict because of its rare occurrence in the vulva. Close follow-up and surveillance

is recommended. If there is concern for recurrence then biopsy at minimum or re-excision is probably warranted.

There is no current classification or grading system for vulvar phyllodes tumours.³ Of the 17 reported cases, only 5 (29%) have been reported to be malignant,^{4 12} 1 reported as borderline⁵ and the remainder reported as benign or not reported. Only three (18%) recurrences of phyllodes tumours have been reported.^{6 10 12} This is comparable to a local recurrence rate of 8% and 21%–36% in the breast following wide local excision for benign and borderline or malignant tumours, respectively.¹⁶ Given the rarity of this tumour in the vulva, true recurrence and malignancy rates may not be accurate.

Herein, we describe a rare phyllodes vulvar tumour, which prior had not been described to arise in the labia minora. In conclusion, despite the reported rarity of this tumour, phyllodes tumours should be considered in the differential diagnosis of vulvar masses. The mainstay of diagnosis and treatment is surgical excision with continued surveillance for recurrence.

Learning points

- ▶ Phyllodes tumour can occur in the vulva and should be considered in the differential diagnosis of a vulvar mass.
- ▶ Vulvar masses should be at a minimum biopsied or removed to determine a definitive diagnosis.
- ▶ Surveillance for recurrence should be performed given the rarity of this tumour with unknown recurrence potential.

Contributors DTF, CAK and RJH saw the patient, performed the procedure, collected data, wrote and edited the manuscript. TCL helped gather pathological images, wrote and edited the manuscript. All authors meet criteria for authorship.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Disclaimer The views expressed in this manuscript are those of the authors and do not reflect the official policy or position of the Department of the Army, Department of Defense or the United States Government.

Competing interests None declared.

Patient consent for publication Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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