

CASE REPORT

ECTOPIC BREAST TISSUE OF THE VULVA

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Ectopic mammary gland tissue in the vulva is an exceptionally rare disease. We present a case of a 62-year-old woman with a left vulvar mass of 30 years duration that progressively increased in size. The patient reported having pressure and discomfort, especially during movement. Surgical excision was performed, and a histopathological examination revealed a well-differentiated ectopic breast. We also review other cases of vulvar ectopic breast to further comprehend the characteristics of this rare disease. Clinicians and pathologists should always consider it as a differential diagnosis when presented with a vulvar mass.

Key words: ectopic, breast tissue, vulva.

Introduction

The incidence rate of ectopic mammary gland is about 1–3%, and it can occur in men or women, more commonly in women [1]. Ectopic mammary gland tissue in the vulva is rare, and ectopic mammary gland tissue in the lactation period under the clitoral prepuce is extremely rare. To date, no relevant case report has been presented by Chinese scholars. The present study aimed to report a case of ectopic breast tissue under the prepuce of the clitoris during lactation, who was admitted to Sichuan Maternal and Child Health Hospital (Chengdu, China).

Case report

A 62-year-old woman was admitted to Sichuan Maternal and Child Health Hospital with a left vulvar mass of more than 30 years that progressively increased in size. Current medical history: a mass of left vulva was found with size of about that of a bean, as well as being soft, without tenderness, redness, and swelling, more than 30 years before admission to our hospital. No attention was paid, and no treatment was given. Over the past 10 years, the size of the left vulvar mass gradually increased, with the size of about that of a quail egg, as well as being

soft, with smooth surface, clear boundary, and without tenderness. Once she was admitted to our hospital, “vulvar lump” was identified. The symptoms were summarized as follows: a 4 × 4 × 3 cm lump on the left labia minora close to the vaginal mouth, which was painless, soft, and dark. The patient was tired, and discouraged, with general appetite, and no difference in stool and urine. Past history: lumbar disc herniation for more than 3 years, and she denied history of hepatitis, tuberculosis, other infectious diseases, drug addiction, and being allergic to drugs or food. She also denied any history of blood transfusion or blood products; history of vaccination was unknown, and there was no other important medical history. Menstrual history: the first menstrual period occurred at the age of 13 years, menstruation period was 5 days, period between menstruations was 30 days, with regular menstrual cycle, red colour, moderate amount, and no dysmenorrhoea; menopause occurred at the age of 48 years.

Marriage and childbearing history: she married at the age of 23 years, with average physical fitness, 4 pregnancies, 2 abortions, 2 spontaneous births, including one male and one female, who were all healthy. Family history: her parents had both died, and there was no family history of hereditary diseases.

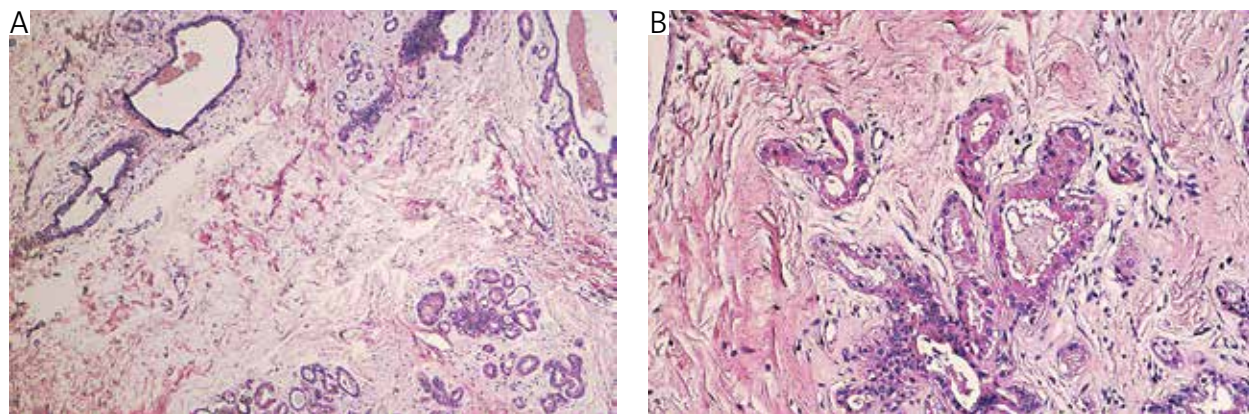


Fig. 1. The gland is covered by two layers of epithelium, the inner layer is a single columnar epithelium with apical secretion, the outer layer surrounds the myoepithelial cells, and the interstitium surrounds loose or dense fibrous tissue: A) HE 40 \times ; B) HE 100 \times

Auxiliary examination: on 29 January 2023, colour Doppler ultrasound performed in our hospital showed that the uterine body was in a posterior position with a anteroposterior diameter of about 2.1 cm, the uterine wall was detected with punctate echo, the echo of the remaining muscle layer was uniform, the uterine cavity line was thin and centred, and the anteroposterior diameter of the cervix was about 1.9 cm. No obvious abnormal blood flow signal was found by colour Doppler flow imaging (CDFI). **Attachment:** there was no obvious abnormal echo in the bilateral attachment area. No obvious liquid anechoic area was found in the pelvic cavity.

Gross pathological examination: “vulvar mass”, a piece of non-plastic tissue with skin flap, with a size of 2.2 \times 1.5 \times 1.3 cm, the size of skin flap area was 2.0 \times 1.5 cm, and the section was grey-brown and solid.

Pathological results: “vulva”, a benign lesion, which was consistent with ectopic breast tissue

Discussion

The incidence rate of ectopic breast is about 1–3%, which can occur in both men and women, especially in women. It is generally located in the armpit, abdominal wall, groin, or limbs [1]. Ectopic breast tissue of the vulva, as reported in the present study, is extremely rare. During human embryonic development, there are several distributed primordial mammary glands. With the development of these primordial mammary glands, they disappear, leaving only one pair of mature mammary glands on the left and right sides of the chest. The breast primordium is rarely extended to the vulva, the breast primordium remaining in the vulva can be single or multiple, and it can be limited to one or both sides [2]. In the present patient, the labia majora of the vulva could be found, and there was breast lobule and duct structure. Immunohistochemical testing of oestrogen (ER) and progesterone (PR) indicated their

positivity, which could be representative of abnormal breast tissue. A previous study [3] showed that up-regulation of levels of ER, PR, and prolactin and a decrease of the androgen level could induce hyperplasia of breast tissue. The present patient was in the post-partum lactation period. The ectopic breast tissue begins to secrete under the influence of lactation stimulation. The vulvar breast tissue rapidly expands in size, making the duct dilate and become cystic. However, because there is no nipple to discharge milk, the milk accumulates and forms a vulvar cyst. Ectopic breast tissues are classified as supernumerary breast tissue and accessory breast tissue. Accessory breast tissue was found in the present case. During lactation, the gland changes, and pathological changes of breast tissue may occur, including breast cyst, mastitis, breast fibroadenoma, and even lobular cancerization. Therefore, there is plasma cell mastitis in the case of ectopic breast (Fig. 1). There are currently 2 hypotheses about the occurrence of ectopic breast tissue in the vulva. One hypothesis is the breast line theory [4]. It is accepted that the ectopic breast tissue originates from the original embryonic breast line, and the degeneration and replacement of the breast line occur at the 5th and 6th weeks of the embryo's life to form the abnormal ectopic breast. It also proliferates with the stimulation of placental hormones during pregnancy. The second hypothesis holds that this is a mammary gland (e.g. a genital gland), and it has the potential to evolve into benign lesions similar to fibroadenoma or malignant lesions (e.g. invasive adenocarcinoma) [5]. The best treatment for both diseases is surgical resection. The patient was surgically resected, and the surgical efficacy was satisfactory, but whether it recurs in the future remains to be further investigated. In conclusion, ectopic breast has the potential for malignant transformation and should be removed surgically. Incision consistent with the skin texture can be selected for intradermal suture to achieve the effect of perineal beauty.

Conclusions

The cyst should be completely removed intraoperatively to avoid increasing the surgical difficulty arising from peeling. It is suggested that the cystic cavity be closed layer by layer to avoid leaving a dead cavity and forming a subcutaneous haematoma.

The authors declare no conflict of interest.

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