Unilateral ectopic breast tissue on vulva in postpartum woman
A case report
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Abstract
Rationale: Ectopic mammary gland tissues occur in about 2% to 6% of women in general population. Vulva is considered a rare site for the ectopic breast tissue.

Patient concerns: We report a rare case of a 27 year-old woman, para 2 and presenting as a vulvar mass in the postpartum period.

Diagnoses: Ectopic mammary tissue in vulva.

Interventions: The mass was removed by wide local excision. Histopathological assessment revealed features of ectopic mammary tissue

Conclusion: The vulvar region is one of the reported sites for ectopic breast tissue in the body. The presence of a rapidly enlarging, well-encapsulated mass in the vulvar region associated with recent delivery or lactation is suggestive of ectopic breast tissue.

Lessons: It is important for clinicians to get a good history and consider ectopic breast tissue on vulva in postpartum women and confirm diagnosis via biopsy with histopathological examination.

Keywords: ectopic breast tissue, extramammary, postpartum, vulva, wide local excision

1. Introduction
Ectopic mammary gland tissues occur in about 2 to 6% of women in general population. It is a residual tissue that persists during embryologic development.[1] Most of the reported cases in the literature tend to occur at the upper end of the embryonic ridge where very few cases were arising from the inferior end of the embryonic mammary ridge.[2] Vulva is considered a rare site for the ectopic breast tissue. Clinically, it gets commonly confused with other benign swellings like lipoma and also malignant neoplastic lesions. Diagnosis is confirmed only after histopathological assessment of tissue.[3]

We describe herein a case of an ectopic breast tissue presenting as a vulvar mass in the postpartum period.

2. Case report
A 27-year-old woman, para 2, referred to the gynecology oncology clinic at King Fahad Medical City, Saudi Arabia on January 2017, with a history of right vulvar swelling for 9 months. She noted the mass first by herself during her last pregnancy at 18 weeks. It was gradually increasing in size especially during the postpartum period with no other symptoms. She had 2 pregnancies, ended by cesarean deliveries for obstetric indications. Her last pregnancy was singleton, uncomplicated during antenatal and she delivered at term. During postpartum period, she breast feed her baby and her menstrual period returned back after 35 days without any significant symptoms.

She had no history of any medical illnesses or allergies. She denied the use of any medications. She has no family history of any malignancies.

Her body mass is 18.8kg/m². Abdominal examination was normal. External genital examination revealed a single ovoid, pedunculated mass coming from the inferior side of the right labia majora which measured about 6 × 4cm. It was soft in consistency, freely mobile without abnormalities of the surrounding skin. There was no palpable inguinal lymphadenopathy. Vaginal and speculum exam revealed no abnormalities and no adnexal mass was detected. Basic blood work up and ultrasound of the pelvis were performed which revealed no abnormalities. At this point we decided to proceed with wide local excision.

Eight weeks postpartum: The patient underwent wide local excision of the mass without any complications and the lesion was sent for histopathology (see Fig. 1). The histopathological assessment revealed features of ectopic mammary tissue. The
The patient was discharged home the day following surgery with follow-up after 4 weeks. She discharged on oral analgesia and advice for wound caring and no restriction for breast feeding. Postoperatively, she developed a wound gapping that was managed conservatively successfully with the help of wound care team. She recovered completely and has gone back to her normal daily activity. Consent for publication of the report and the image was obtained from the patient.

3. Discussion

Ectopic mammary tissue extends along the milk lines, bilaterally from the mid-axillae through the normal breasts and then inferiorly to the medial groins.[4] These tissues disappear soon after its formation. However, breast tissues may occur anywhere along the milk line, most commonly just below the breast. More rarely, in other sites.[5] In the present case, it appeared on vulva of a postpartum woman.

It has been reported that ectopic breast tissues occur more frequently among Japanese women and less frequently among Caucasians.[6,7]

Ectopic breast tissues are susceptible to the same pathological processes affecting the normal breast tissues and frequently respond to hormonal stimulation associated with menstruation, lactation or pregnancy, resulting in engorgement and discomfort.[11] It is well known that during pregnancy, high levels of estrogen, progesterone, and prolactin promote the growth and proliferation of the breast tissues. Similar effects can happen on ectopic breast tissue. Ectopic breast tissue in older women is of more concern because such tissue could undergo neoplastic changes.[15]

Postpartum, elevated levels of prolactin and withdrawal of estrogen and progesterone result in the onset of lactogenesis. The high level of prolactin declines approximately 3 to 4 days after delivery; unless the production remains stimulated by sucking of the baby.[19] These hormonal changes explain the presentation of our case where the vulvar mass was discovered during pregnancy and it was gradually increasing in size especially during the lactation postpartum.

The ectopic breast tissue lesions are usually asymptomatic as seen in our case. However, pain might occur due to a rapid growth of the tissue, in a fashion similar to that of normal situated breast tissue during pregnancy and lactation.[9] They have no typical characteristics, so it may go unnoticed for several years. The only way to confirm its diagnosis is via biopsy with histopathological examination as done in the present case.[10]

Vulvar breasts serve no useful function in the human; on the contrary, they act as the potential source of diseases. Therefore, whenever discovered they should be promptly removed.[13] We followed this recommendation with our case through immediate surgical excision.

4. Conclusion

The vulvar region is one of the reported sites for ectopic breast tissue in the body. The presence of a rapidly enlarging, well-encapsulated mass in the vulvar region associated with recent delivery or lactation is suggestive of ectopic breast tissue. It is important for clinicians to get a good history and consider ectopic breast tissue in the differential diagnosis if a swelling occurs along the milk line.

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