Case Report

Spuriously aggressive features of a lactating adenoma prompting repeated biopsies

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\textbf{Abstract}

We present an atypical presentation of a common pregnancy-related breast mass, a lactating adenoma with imaging and pathologic correlation. The patient presented with a rapidly enlarging left breast mass associated with skin changes and severe pain in the perinatal period. Core biopsies were considered discordant, and the patient went on to surgical excision for the definitive diagnosis of an infarcted lactating adenoma. The symptoms of infarction may obscure the diagnosis of common entities and result in additional evaluation.

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\textbf{Case report}

A 33-year-old woman with a history of a left breast mass discovered during the first trimester of pregnancy had an image-guided biopsy early in the third trimester when the enlarging mass measured 4.5 cm. The biopsy was performed at an outside institution and yielded gestational hyperplasia. Just prior to delivery, the mass rapidly increased in size. A second core biopsy of the, now, 15 cm mass performed after delivery demonstrated gestational and secretory hyperplasia and was deemed discordant. Approximately 2 weeks postpartum, the patient presented to our institution for diagnosis and management of the large left breast mass associated with debilitating pain, erythema, and skin ulceration (Fig. 1). The mass was not adherent to the chest wall, and there was no lymphadenopathy. The right breast appeared normal.

On inspection, the medial half of the left breast was virtually replaced by a large protruding mass. The patient was experiencing severe pain and was unable to lie in a supine position. Ultrasound, performed with the patient in a semi-erect position, demonstrated a circumscribed predominately solid mass of mixed echogenicity extending from the nipple to the medial edge of the breast without associated internal vascularity (Fig. 2). Occluded milk duct abscess or infected galactocele was suspected clinically, and aspiration was requested. An ultrasound-guided aspiration was attempted during which no fluid or blood could be obtained. Surgical exploration of the healed biopsy tract was performed and yielded multiple fragments of tan rubbery tissue with no evidence of an abscess, raising the possibility of an angiosarcoma. The patient was admitted to the hospital for pain management and surgical excision of the mass for definitive diagnosis.

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A wide surgical enucleation was performed, and a 16-cm necrotic, tan, lobulated spongy mass with fibrotic capsule (Fig. 3) was shelled out from adherent surrounding breast tissue. Pathology revealed expanded lobules with closely packed glands as well as infarctive changes (Fig. 4).

Discussion

Discovery of a breast mass during pregnancy or the postpartum period is common, and the majority of these masses are benign. Many predate the pregnancy but were clinically undetected. A third of these masses are unique to pregnancy, while the remaining two-thirds are masses that occur in like aged nongravid patients. A lactating adenoma is one of these unique entities and is also one of the most prevalent breast masses seen during this time [1]. The diagnosis may be obscured by the symptoms of infarction which mimic other entities, both benign and malignant.

Lactating adenomas develop due to the physiologic changes occurring in the breast with the hormonal variations of pregnancy and lactation. The typical presentation is a painless, soft, mobile breast mass detected in the third trimester of pregnancy or in the puerperal breast-feeding period. They may be multiple, bilateral and develop in ectopic breast tissue along the milk line from the axilla to the groin [2]. However, acute infarction can cause significant enlargement and moderate-to-severe pain making difficult the distinction from an infected galactocele, abscess, or a high-grade malignancy with or without cystic degeneration. Approximately 5% of lactating adenomas are reported to undergo infarction [1].

Grossly, lactating adenomas are typically circumscribed, lobulated solid masses. They range from gray-tan to yellow and may be firm or rubbery. Histologically, they are composed of circumscribed aggregates of lobules with secretory hyperplasia and lactational change but often lack a true capsule [3] (Fig. 4).

The mammographic appearance of a lactating adenoma is usually a circumscribed oval mass equal to or lower in density to breast parenchyma. The sensitivity of mammography for mass detection may be decreased in the peripartum patient due to a hormone-driven increase in parenchymal density superimposed on the high tissue density usually found in young women. Ultrasound is therefore the preferred modality when evaluating peripartum patients.
On ultrasound, most lactating adenomas are circumscribed, homogeneously hypoechoic or isoechoic masses with posterior acoustic enhancement displaying increased vascularity. They may also appear as a hyperechoic mass due to inspissated milk or other proteinaceous secretions within the dilated acini. Occasionally, an echogenic “pseudocapsule” may be seen due to a thin layer of compressed stroma around the enlarging neoplasm [4]. Microlobulation may occur late in pregnancy or during lactation. Spuriously aggressive features such as irregular margins, posterior acoustic shadowing, and internal heterogeneity, may occur with infarction making it indistinguishable from malignant entities.

Most lactating adenomas regress spontaneously requiring no additional treatment. If the lesion does not regress and produces severe pain, surgical management may be necessary. Bromocriptine has been used to reduce the size of a lactating adenoma prior to surgical excision [5]. There is no malignant potential although cases of concomitant lactating adenoma and breast cancer have been reported [6,7].

Differential considerations for a rapidly enlarging breast mass during pregnancy or lactation include both benign and malignant entities. While benign masses such as galactoceles, abscess, lactating adenoma, and fibroadenoma are most commonly encountered, other masses including phyllodes tumor, pregnancy-associated breast cancer, and sarcoma must also be considered.

The presence of a fat fluid level within the mass and the lack of associated vascularity favor a galactoceles. An infected galactoceles presenting as a debris-filled hypoechoic mass with prominent posterior acoustic enhancement associated with erythema, warmth, and tenderness may be indistinguishable from an abscess.

The imaging appearances of a lactating adenoma, fibroadenoma, and phyllodes tumor are often indistinguishable. They may all present as oval well circumscribed, slightly hypoechoic parallel masses with mild posterior acoustic enhancement. Lactating adenoma and fibroadenoma may also enlarge and infarct in association with pregnancy or lactation, possibly from vascular insufficiency related to increased metabolic demands during pregnancy [8]. Phyllodes tumors similarly exhibit rapid growth; however, the median age of phyllodes tumor presentation is during the fifth decade, and there is no definitive association with pregnancy or lactation.

Breast cancer and sarcoma typically have suspicious features on ultrasound with indistinct margins, variable echogenicity, and increased vascularity. Pregnancy-associated breast cancer, defined as breast cancer diagnosed during pregnancy or the first postpartum year, most commonly presents as a palpable mass with suspicious imaging findings. Primary angiosarcoma, observed in women during the third to fifth decades, also presents as a palpable mass which may enlarge rapidly and typically has markedly increased vascularity on ultrasound [9]. There are no known risk factors for primary angiosarcoma of the breast, and although 6%—12% of these tumors are diagnosed during or shortly after pregnancy, there is no established link between primary angiosarcoma and hormonal dependency [10].

A newly palpable or enlarging breast mass in a pregnant or lactating patient should be promptly investigated. The value of ultrasound in demonstrating malignant masses in this clinical scenario is well established [11,12], and biopsy of any mass with suspicious features is recommended to exclude malignancy. Fine needle aspiration may yield nondiagnostic results as well as false-positive results secondary to the proliferative changes associated with pregnancy [13]. Core biopsy is definitive and can safely be performed. A result of benign breast tissue with gestational/secretory hyperplasia with or without infarction is concordant with a lactating adenoma.

**Conclusion**

The discovery of a palpable breast mass lasting 2 weeks or longer in a pregnant or lactating patient should prompt an imaging evaluation beginning with ultrasound. Biopsy should be performed on any suspicious mass and should not be delayed until after delivery. Understanding the significant differences in ultrasound appearances between malignant and benign masses is necessary for accurate diagnosis.
physiological changes in the breast during pregnancy and lactation will aid in determining concordance of biopsy results.

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