Adenoid Cystic Carcinoma of the Breast Arising in a Lipoma Mimicking Hamartoma: imaging manifestations and review of the literature

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Adenoid cystic carcinoma of the breast is a rare neoplasm and only a few articles have described the imaging findings of the disease. We report a 52-year-old woman who presented with a palpable mass beneath the left nipple. Ultrasound revealed a well-defined, lobulated, hypoechoic mass. Mammography demonstrated a radiopaque mass in a well-defined fat-containing lesion, suggestive of hamartoma. The patient underwent surgery due to enlargement of the tumor. The final diagnosis disclosed adenoid cystic carcinoma arising in a lipoma. The unique imaging findings have never been reported in literature.

Although adenoid cystic carcinoma (ACC) occurs most frequently in the salivary glands and the upper respiratory tract, it is rare in the breasts [1-5]. Mammary ACC accounts for only 0.1-0.4% of all breast carcinomas [3-5]. Its rarity makes it difficult to establish an accurate preoperative diagnosis [4, 6]. The prognosis of mammary ACC is reportedly excellent, with few instances of local recurrence or distant metastasis [3-8]. The medical literature reported only a few cases with imaging and histopathological diagnoses of ACC of the breast [1-7, 9], showing varied radiologic appearance [1-3, 5, 6, 9]. Herein, we report and discuss the imaging and histopathological findings of a patient with ACC of the breast.

CASE REPORT

A 52-year-old postmenopausal woman came to the outpatient department with the complaint of an enlarging palpable, painless mass in the left breast for 6 months. Physical examination revealed a freely movable, elastic mass in the subareolar region of the left breast, about 6 cm in size, without skin change or nipple discharge. There was no palpable axillary lymphadenopathy.

Ultrasonography (Fig. 1a) showed a well-defined, lobulated mass, measuring about 2.2 cm in diameter in the left central breast. The patient underwent ultrasound-guided fine needle aspiration (FNA) of the lesion and the cytological analysis disclosed fibroadenoma with scanty cellularity. The following mammography (Fig. 1b) demonstrated a focal radiolucent lesion about 6.5 cm in diameter in the left central breast, associated with a well-defined oval solid mass, measuring approximately 2 cm in diameter, lying inside the anterior aspect of the radiolucent lesion, which corresponded to the lesion disclosed on ultrasonography. The initial impression was a ham-
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The patient received surgical excision for the tumor due to enlargement of the mass. The operative finding showed a 6 x 5 x 4 cm yellowish, elastic, soft tumor at the left subareolar region. The gross specimen revealed a yellowish elastic tissue measuring about 6 x 4 x 2.8 cm. After serial dissection, it consisted of a white well-circumscribed tumor measuring 2 x 1.8 x 1 cm in size located close to the section margin (< 1 mm). Histological analysis of the tumor revealed adenoid cystic carcinoma with mixed cribriform, solid, and tubular patterns of growth (Fig. 1c). The immunohistochemical study of the tumor showed that it was negative for Her-2-neu, estrogen receptor and progesterone receptor. The rest of the breast tissue consisted of normal glandular tissue within a fibrofatty stroma.

The patient underwent left modified radical mastectomy and axillary lymph node dissection with prosthesis reconstruction 2 weeks later. The histopathology revealed no evidence of tumor cells within the breast and dissected lymph nodes. The patient did not undergo post-operative radiation and chemotherapy. She was free from local and distal recurrence of the disease at her 1-year follow-up.

Figure 1. The imaging and pathological features of the tumor for our case. a. Ultrasonography revealed a well-defined, gently lobulated, hypoechoic mass (arrows), measuring 2.2 cm in diameter, in the left subareolar region, with parallel orientation and posterior enhancement. b. Mammogram of mediolateral oblique projection of left breast shows a 6.5 cm well-defined, radiolucent lesion located in the central breast (arrows) and a 2 cm, well-defined, radiopaque nodule (arrowheads) in the anterior aspect of the radiolucent lesion. c. Low power photomicrograph shows the classical cribriform pattern of adenoid cystic carcinoma (arrows), with mucin within the pseudocysts (Hematoxylin-eosin stain, magnification x40).
DISCUSSION

Adenoid cystic carcinoma (ACC) of the breast is very rare although its cellular origin in the breast remains unclear, it may derive from the ductal epithelium or myoepithelium [1]. The prognosis is more favorable than other forms of invasive breast cancer because lymph node and distant metastasis are less common [3-8]. The most common clinical feature of ACC is a painful, palpable mass [4, 6]. ACC tends to occur more commonly in the subareolar region but nipple discharge is uncommon [4, 7].

There are few articles mentioning about the ultrasonography of mammary ACC. Ultrasonography of breast ACC most commonly demonstrates as an irregular or polylobulated mass with somewhat inhomogeneous hypoechogenicity [2, 9]. Sometimes it appears as a well-defined, oval or lobulated mass [6, 9]. In our patient, the tumor showed a well-delineated margin, mild lobulated configuration, mild inhomogeneous hypoechogenicity, and posterior enhancement on ultrasound. On mammography, ACC can appear as an irregular or circumscribed, oval to lobulated mass [2, 6, 9], or a focal asymmetry with microcalcifications [1]. Blanco et al. [5] reported an ACC arising in a fibroadenoma, which appeared as a well-defined oval mass on mammography and ultrasound. For our patient, mammography of the lesion showed a well-delineated radiopaque mass within a radiolucent lesion, leading to the initial misdiagnosis as a breast hamartoma. However, when analyzing the imaging findings of our case retrospectively, there is only a single solid part lying eccentrically in the radiolucent mass. This finding is not really typical for hamartoma. The typical mammographic manifestations for a hamartoma are a well-defined radiolucent mass surrounded by a thin radiopaque pseudocapsule and containing multiple round to oval radiopaque masses of varying size related to presence of fibrous and glandular elements [3]. According to the imaging and pathological findings, we believed that the solid part corresponded to the ACC, which was surrounded by lipomatous tissue. We could not find any report showing an ACC within a fat-containing lesion when reviewing the literature. On the corresponding ultrasound, we failed to recognize the fatty component, which is probably due to the relatively isoechoic presentation of the fatty component making it difficult to be identified on ultrasound.

Though the diagnosis of ACC can be made by FNA cytology [8], the carcinoma in our case was missed by FNA cytology most likely due to insufficient tissue sampling. Large core biopsy or open surgical biopsy is more reliable than FNA for tissue diagnosis [10].

Although mastectomy or lumpectomy followed by irradiation is sufficient to attain adequate local control for ACC of the breast, there is little consensus in the literature regarding the role of adjuvant systemic therapy for these patients [4, 6, 7]. However, ACC of the breast is a rare neoplasm and has rare local recurrence or distant metastasis [4, 6, 7]. Therefore, since the tumor of our patient showed negative estrogen/progesterone receptor and negative axillary lymph nodes, it was reasonable for our patient not to receive any adjuvant systemic treatment.

In conclusion, we present a case with unique imaging findings of adenoid cystic carcinoma of the breast lying within a lipoma, thus leading to misdiagnosis of a hamartoma mammographically. Awareness of the rare imaging manifestation of this tumor will help prevent misdiagnosis in the future.

REFERENCES

因源自於脂肪瘤內而形似缺陷瘤之乳房腺狀囊狀癌：影像表現及文獻回顧

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乳房腺狀囊狀癌是一種罕見之乳房腫瘤，目前只有少數文獻報告討論其影像表現。我們報告一位 52 歲女性病人，以可觸摸到之左側乳頭下腫塊為臨床表徵。超音波表現為一個邊緣清楚，分葉狀，低迴音之腫塊。乳房攝影呈現一個含有脂肪成分、放射透光狀腫塊，其型態表現有可能為缺陷瘤。由於腫塊變大，故病患接受手術，開刀結果為一源於脂肪瘤內之乳房腺狀囊狀癌。此一特殊影像表現形式尚未於文獻報告提出過。