Adenomyoepithelioma of the Breast

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Adenomyoepithelioma of the breast is a rare lesion characterized by biphasic proliferation of ductal epithelium and myoepithelial cells. A 37-year-old female presented with a non-tender, mobile, smooth mass located in the retroareolar area of her left breast. Ultrasonography revealed a 1.76 × 1.73 × 1.23 cm wider-than-tall tumor with homogeneous hypoechoic echotexture. The tumor margins were smooth with a thin echogenic capsule. The tumor was compressible and had posterior acoustic enhancement. Histopathologic analysis demonstrated a well-circumscribed tumor with mammary ductal epithelium and myoepithelium in the fibrous stroma. Immunohistochemical staining was positive for smooth muscle actin, vimentin, and S-100 protein in the myoepithelial cells. No recurrence or distant metastases was observed during a 14-month follow-up period.

KEY WORDS — adenomyoepithelioma, breast, hypoechoic, myoepithelium, ultrasonography


Introduction

Adenomyoepithelioma of the breast is a rare lesion that was first described by Hamperl in 1970 [1]. Although most tumors are benign, local recurrence and axillary nodal or distant metastases have been reported [2–4]. Thus, complete excision with adequate margins is recommended to minimize the likelihood of recurrence.

Most breast pathologies can be diagnosed by fine needle aspiration biopsy (FNAB) or core needle biopsy [5,6]. However, cytology and core needle biopsy may not detect adenomyoepithelioma [7–9]. Tumor excision is therefore necessary for diagnostic confirmation. Persuading patients to undergo additional surgery following local excision and histologic diagnosis is difficult. Many patients decline to have this procedure. Therefore, accurate preoperative diagnosis is critical to avoiding subsequent surgery. To the author’s knowledge, most adenomyoepitheliomas are analyzed by mammography. A few authors have reported sonographic evaluation of adenomyoepithelioma, but without describing detailed sonographic features [7–10]. This article describes the sonographic features of an adenomyoepithelioma indicative of a benign tumor.

Case Report

A 37-year-old, gravida 2, para 2, woman presented with a 2-month history of a palpable lump with bloody discharge from her left nipple. Physical examination identified a 2 × 1.5 cm non-tender, mobile, smooth mass in the retroareolar area of

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the left breast. No axillary lymphadenopathy was detected. The patient had no family history of breast diseases. Although the patient had not taken sex hormones, she was taking prednisolone (10 mg/day) and hydroxychloroquine sulfate (200 mg/day) for systemic lupus erythematosus.

The patient was examined by the HDI 5000 ultrasound system (ATL Ultrasound, Bothell, WA, USA) using a 5–12 MHz linear-array transducer. Ultrasonographic analysis revealed a 1.76 × 1.73 × 1.23 cm mass in the retroareolar space of the left breast (Fig. 1). The mass had homogeneous hypoechoic echotexture and a wider-than-tall shape with slight lobulation. The tumor margins were smooth and the tumor had a thin echogenic capsule. The tumor showed posterior acoustic enhancement and tumor contour changed when pressure was applied with a probe. No microcalcifications within the mass were demonstrated. A small cystic lesion was next to the tumor.

The patient refused galactography and the tumor was excised as the patient requested. Histopathologic examination indicated a well-circumscribed tumor with mammary ductal epithelium and myoepithelium in the fibrous stroma (Fig. 2). Immunohistochemical analysis demonstrated positive staining for smooth muscle actin, vimentin, and S-100 protein in the myoepithelial cells. Although the tumor was completely excised, secondary surgery to excise a wider margin was recommended. The patient declined this surgery. Nipple discharge disappeared after tumor excision and there was no recurrence or distant metastases during a 14-month follow-up period.

**Discussion**

Myoepithelioma, adenomyoepithelioma, and myoepithelial carcinoma are typical breast lesions originating from myoepithelial cells [3]. Adenomyoepithelioma, a rare lesion characterized by biphasic proliferation of ductal epithelial and myoepithelial cells, can be subclassified into spindle cell, tubular, and lobulated types based on the growth pattern and cell morphology [1,3,11]. The typical histologic appearance of a benign adenomyoepithelioma comprises acinar structures consisting of an inner layer of epithelial cells with eosinophilic cytoplasm, and a dominant peripheral layer of myoepithelial cells (Fig. 2). Myoepithelial cells are easily confirmed immunohistochemically.
via positive staining for smooth muscle actin, vimentin, and S-100 protein [12]. Although most adenomyoepitheliomas are benign, malignant change can occur [11–13]. Malignant change can involve only the epithelial or myoepithelial component. Although malignant transformation of both components can also occur, it is extremely rare [14]. Metastases to the lungs, brain and thyroid can occur in malignant adenomyoepithelioma that are hematogenous rather than lymphatic [4,14,15].

Based on the rarity of adenomyoepithelioma and the varied and hypercellular nature of such tumors, they can be confused with the cytologic features of a number of benign and malignant breast lesions [7–9,16]. High mitotic activity and cytologic atypia identified in both epithelial and myoepithelial cells can aid in differentiating malignant and benign tumors. Core needle biopsy is potentially a more accurate diagnostic modality than FNAB. However, core needle biopsy can generate conflicting results [9]. When a benign adenomyoepithelioma is diagnosed by FNAB or core needle biopsy, total excision of the lesion with a margin of uninvolved breast tissue is indicated.

Clinically, adenomyoepithelioma commonly presents as palpable, non-tender, solid, well-circumscribed or lobulated, firm or hard masses [3]. Tavassoli reported an average size of 2.5 cm for 27 tumors, most of which were centrally located [3]. Rosen reported an average size of 1.5 cm for 18 tumors that were principally peripherally located [11]. Such tumors have reportedly remained palpable for over a year prior to excision in some
cases. However, most patients experience recent onset. Rapid enlargement of a mass is extremely suggestive of malignant change [7]. Bloody nipple discharge has not been reported in the literature as a symptom of adenomyoepithelioma. However, our patient presented with bloody nipple discharge and a clinically palpable lump. In patients with nipple discharge, mammography, breast ultrasonography and galactography can help to establish the diagnosis [17]. The sensitivity of mammography is reduced in women with radiographically dense breasts [18,19], therefore, ultrasonography is commonly employed by physicians to diagnose breast lesions in Chinese women as most have smaller breasts and denser breast tissue than Western women [20,21]. Some authors recommend galactography as the diagnostic procedure of choice for nipple discharge [22,23]. However, others argue that galactography has little role in diagnosing nipple discharge and surgical biopsy should be offered in the presence of a palpable tumor or if diagnostic breast imaging is positive [24–26]. Sonographic examination of the present case showed that the tumor connected with a lactiferous duct (Fig. 1). Although histologic examination did not reveal the connection between tumor and lactiferous duct, Singh Gill et al reported a well-circumscribed adenomyoepithelioma extending into the lactiferous ducts [9]. In the present case, bloody nipple discharge vanished after tumor excision and did not reoccur during the follow-up period, suggesting that adenomyoepitheliomas can present with nipple discharge.

Imaging findings for breast adenomyoepithelioma are typically nonspecific. Mammography and sonography of malignant adenomyoepithelioma show features indicative of malignancy. A benign adenomyoepithelioma produces mammographic findings indicative of a benign lesion. Sonographic features vary from a solid mass to a cystic lesion [7–10]. Solid adenomyoepithelioma can present with sonographic features suggestive of a malignant tumor. Tukel et al described a solid adenomyoepithelioma with hypoechoic echotexture, irregular contours and spicular extensions [10]. This mass had microcalcifications within the lesions producing minimal acoustic attenuation. Howlett et al described the sonographic features of three patients with adenomyoepithelioma [7]. Sonography of the benign adenomyoepithelioma demonstrated a circumscribed and homogeneous hypoechoic solid nodule. Sonographic findings for the malignant adenomyoepithelioma were a hypoechoic nodule with ill-defined margins and retrotumor acoustic shadowing. The third mass was an infiltrating ductal carcinoma arising in a benign adenomyoepithelioma. Sonography indicated a hypoechoic mass with ill-defined margins. Papaevangelou et al characterized the sonographic features of cystic adenomyoepithelioma [8], which are typically well-circumscribed cysts with a hypoechoic, lobulated solid lesion at the posterior wall of the cyst. The sonographic characteristics in the present case indicated a benign tumor, i.e. well-circumscribed, smooth margins, thin echogenic capsule, slightly lobulated contour, and homogeneous hypoechoic internal echotexture [27].

Recurrence can happen several years after surgery in cases of benign and malignant adenomyoepithelioma. An initial incomplete or narrow excision margin for a benign adenomyoepithelioma may predispose patients to local recurrence. Therefore, complete local excision should be performed. If the excision margin is narrow or incomplete, re-excision to obtain suitable margins is recommended. Long-term follow-up is suggested. In cases of recurrence, a wide excision of the lesion is recommended. When carcinoma is diagnosed histologically, the lesion should be treated as a carcinoma by mastectomy and axillary dissection.

References
