Adenoid Cystic Carcinoma of the Breast associated with Adenomyoepithelioma

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We report her on a case of adenoid cystic carcinoma (ACC) associated with adenomyoepithelioma of the breast in a 73-year-old woman. ACC accounts for about 0.1% of all breast cancers. The patient presented with a large, nodular, rubbery mass that measured 5.5×4.0 cm in the subareolar region of the left breast. Light microscopy revealed various growth patterns of ACC with the adenomyoepithelioma. Immunohistochemically, the epithelial cells in the ACC component reacted strongly for cytokeratin. The myoepithelial cells stained for smooth muscle actin, vimentin and S-100 protein, whereas staining for cytokeratin was weak. The adenomyoepithelioma component showed a similar staining pattern. Reactivity for both estrogen receptor (ER) and progesterone receptor (PR) were negative. There seems to be a spectrum of adenomyoepithelioma-related epithelial and myoepithelial lesions that ranges from low grade to high grade. Therefore, we suggest the present case of ACC arose from an adenomyoepithelioma or there is a close relationship among these combined epithelial and myoepithelial tumors. For both mammary ACC and adenomyoepitheliomas, complete excision with a margin of uninvolved tissue is the recommended treatment. Most of these patients will have an excellent prognosis.


Key Words Adenoid cystic carcinoma, Adenomyoepithelioma, Breast carcinoma, Salivary gland-like tumor

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a rare breast tumor composed of epithelial and myoepithelial cells similar to its salivary counterpart. Although the histologic features and tumor biology in salivary gland have been relatively well documented, breast ACCs are rare and not aggressive lesions. Here a case of ACC associated with adenomyoepithelioma of the breast is presented, along with histologic, cytologic and immunohistochemical features.

CASE REPORT

A 73-year-old woman presented with a left breast lump of unknown duration. Physical examination revealed a huge, tender, solitary mass in the subareolar region of the left breast. Mammography showed a huge lobulated mass with a well-defined contour, but it was partly indistinguishable from

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normal parenchyma and infiltrated to the nipple and adjacent tissue. Ultrasonographically, the mass was solid, partly cystic and necrotic, with a heterogeneous, low echogenicity compared to the parenchyma. Physical examination, ultrasonography and mammography strongly suggested a malignant tumor. The aspiration cytology showed small and large sheets, nests or small aggregates of round to oval cells with scant cytoplasm (Fig 1). In some clusters, the epithelial cells surrounded homogeneous acellular material. A diagnosis of mammary carcinoma with features strongly suggestive of ACC was considered, but we misdiagnosed. A lumpectomy was subsequently performed. Grossly, the tumor is a $5.5 \times 4 \times 3.5$ cm-sized solid to rubbery breast mass (Fig 2). The mass is brown well circumscribed, but adipose tissue is firmly attached. The cut surface is pale soft homogeneous and shows focal cystic spaces. Microscopically, the tumor was composed of cribriform and trabecular-tubular patterns of ACC component admixed with adenomyoepithelioma component. The ACC component consisted of dark basoloid (myoepithelial) cell and slightly larger ductal (epithelial) cells with an eosinophilic cytoplasm. The cribriform structures were punctuated by spaces filled with basement membrane-like material (hyaline bodies) (Fig 3) and mucoid secretory material which stain with Alcian blue and PAS stains.

The adenomyoepithelioma component consisted of irregular glandular structures with small lumina lined by an inner epithelial layer and an outer hyperplastic myoepithelial cell layer (Fig 4).

The epithelial cells were cuboidal and had a moderately abundant eosinophilic cytoplasm; the myoepithelial cells were cuboidal or polyhedral and had some clear cytoplasm. In the H&E stained sections it was not always possible to distinguish clearly between the epithelial and myoepithelial cells. Between the glandular structures there was a cellular fibroblasticstroma. Perineural invasion was not seen. Immunohistochemically, the epithelial cells in the ACC component reacted strongly for cytokeratin. The myoepithelial cells stained for alpha-smooth muscle actin, vimentin and S-100 protein, whereas staining for cytokeratin was weak. The adenomyoepithelioma component showed a similar staining pattern.

DISCUSSION

ACC has been extensively described in several organs, including salivary gland, cervix, lacrimal gland, lung and larynx. However, it is an uncom-
mon neoplasm of the breast, comprising less than 0.1% of breast carcinomas, and is alleged to have myoepithelial cell differentiation. The average age at presentation is between 50 and 63 years. Rare example afflicting men is on record. The distinctive cytologic traits of ACC in the breast are similar to those occurring in other body sites. Features of ACC identified in our case and previously reported in other studies showed cellular smears with metachromatic spherules surrounded by three-dimensional uniform epithelial cell clusters with scant cytoplasm and bland nuclei. The cytologic features of adenomyoepithelioma are less well characterized and frequently mimic those of different breast lesions. We were unable to distinguish the cytologic features of ACC and adenomyoepithelioma. Despite its characteristic appearance on aspiration cytology, ACC may be misinterpreted, because of its low incidence in the breast. Histologically, ACC shows three different architectural patterns as the salivary analogue: trabecular-tubular, cribriform, and solid. ACC is represented by both true glandular lumina containing basophilic secretions of PAS positive neutral substances and pseudocysts with cylindromatous material contain myxoid stroma or collagen fibers. Two different types of cells line these two types of spaces. One type of cell, basaloid cell, displays centrally located oval nuclei and a thin cytoplasm. These cells line the spaces containing stroma and basement membrane-like material. The second cell type, characterized by round nuclei and eosinophilic cytoplasm, surrounds true glandular lumina. Adenomyoepithelioma is composed of two cell types, myoepithelial and epithelial. The myoepithelial component is composed of cuboidal to spindle shaped cells with round nuclei and clear cytoplasm. This cell type constitutes the outer layer of the tubular structures. The epithelial component is composed of columnar cells with eosinophilic cytoplasm, which line the lumina of the tubular and papillary structures. Immunohistochemically, the epithelial cells in the ACC component reacted strongly for cytokeratin.

Fig 3. ACC showed cribriform patterns and tubular solid patterns with focal cylindromatous areas and basement membrane-like material (hyaline bodies)(H&E stain, ×100).

Fig 4. The adenomyoepithelioma component consisted of irregular glandular structures with small lumina (H&E stain, ×100).
The myoepithelial cells stained for smooth muscle actin, vimentin and S-100 protein. Basaloid cells usually express laminin, fibronectin and type IV collagen, whereas luminal cells express epithelial differentiation. E-cadherin and β-catenin. Either the epithelial, the myoepithelial or both components may become malignant and give rise to a carcinoma while the background lesion retains its adenomyoepitheliomatous appearance. Carcinomas arising in an adenomyoepithelioma have been reported in the literature. A reported case of ACC arising in an adenomyoepithelioma was small and confined to the peripheral aspect of the lesion. In present case, a majority of the tumor was an ACC and focally admixed with adenomyoepithelioma. The histogenesis of carcinoma ex adenomyoepithelioma is unclear. There seems to be a spectrum of adenomyoepithelioma-related epithelial and myoepithelial lesions that ranges from low grade to high grade. Therefore, we suggest a present case of ACC is arising in an adenomyoepithelioma or the existence of a close relationship among these combined epithelial and myoepithelial tumors.

In contrast to the aggressive behavior of salivary gland ACC, mammary ACCs have a better prognosis. For both mammary ACC and adenomyoepitheliomas, complete excision with a margin of uninvolved tissue is the recommended treatment. Ro et al. divided ACC into three groups according to the proportion of solid growth (I, no solid elements; II, < 30% solid; III, > 30% solid). They found that recurrences and metastases were restricted to patients with grade II or III. The present case was categorized to the group II. Recently, Shin and Rosene reported that the solid basaloid features are indicative of a tendency to develop axillary lymph node metastases and suggested that axillary dissection should be performed in patients with these features.

REFERENCES