Bilateral adenomyoepithelioma of the breast presenting with breast abscess in a lactating woman: A case report

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ABSTRACT

The term adenomyoepithelioma has been applied to a broad range of biphasic lesions composed of epithelial and myoepithelial cells. They show diverse morphologic patterns due to the admixture of the two components which may lead to a diagnosis of malignancy. We present a case of bilateral adenomyoepithelioma in a lactating woman who had a concomitant breast abscess. A 25-year-old lady presented with bilateral breast lumps since 2 years, with acute pain. With a clinical diagnosis of an abscess, superimposed on fibrocystic disease, drainage of abscess, and lumpectomy was performed. The clinicopathological features of this entity are discussed.

INTRODUCTION

Adenomyoepithelioma is a proliferation of both epithelial and myoepithelial elements. It is a rare neoplasm first described by Hamperl in 1970 [1]. The term has been used to describe many lesions that are part of a spectrum of changes with myoepithelial prominence occurring in the breast and in other sites. Other lesions in this class include adenoid cystic carcinoma, pleomorphic adenoma, myoepitheliosis, adenomyoepithelial adenosis, and malignant myoepithelioma [2].

CASE REPORT

A 25-year-old lactating woman presented with bilateral lumps in breast present since 2 years. In addition, she complained of mastalgia of 5 days duration along with fever. She was 5 months post-partum and was breastfeeding. The lump on the right side was associated with pain and redness of the skin. On examination, both the breasts appeared normal in size with reddish discoloration of the right lower quadrant. No discharge or nipple retraction was noted. On the left side, the mass was palpable in left upper quadrant measured 5 cm × 5 cm, was freely mobile and firm in consistency. The right side mass involved most of the breast, with a soft to firm consistency and cystic areas peripherally. Ultrasonography revealed multiloculated hypoechoic collection with an adjacent circumscribed complex echogenic mass, BIRADS category 4A. As she was a lactating woman, presenting acutely with superadded infection, bilateral lumpectomy with drainage of abscess was performed.

Grossly, the specimen consisted of 2 lumps from the right side and one left sided lump. The right sided lumps were well encapsulated and weighed 134 g totally. The larger mass measured 7 cm × 5 cm × 3 cm and the smaller one measured 4 cm × 4 cm × 3 cm. The left-sided mass weighed 59 g and measured 7 cm × 5 cm × 2 cm. The cut section of all masses showed a well-encapsulated lesion with central grey white areas and peripheral dilated cysts, filled with inspissated secretions [Figure 1].

Histologically, sections showed a well delineated lesion with a pseudocapsule and was composed of acini, cell nests and cords of cells with predominantly clear cytoplasm, oval nuclei and prominent nucleoli, forming the center of the tumor [Figure 2]. In the surrounding areas, there was a proliferation of spindle myoepithelial cells in sheets and fascicles [Figure 3]. The right-sided mass in addition, had areas of fat necrosis with infiltration by foamy histiocytes, occasional foreign body giant cells and neutrophils in an edematous stroma with proliferating capillaries lined by plump endothelial cells. Immunohistochemistry delineated the epithelial elements (stained by Cytokeratin AE1/AE3) [Figure 4], surrounded by a proliferation of myoepithelial...
Myoepithelial cells are normally present in the breast and are located between the luminal cells and the basal lamina of the mammary duct and lobular system. The most common lesion affecting these cells is sclerosing adenosis. Tumors derived from myoepithelial cells have been reported in skin, salivary glands, breast and lungs [2].

Adenomyoepithelioma usually presents as a solitary palpable nodule ranging from 1 cm to 7 cm in size. They are usually circumscribed but may have lobulated, somewhat irregular, pushing borders. Central necrosis and dystrophic calcifications have been reported in very large tumors. Their biphasic nature consists of cuboidal to columnar epithelial-lined tubules, often with apocrine change, that are surrounded by a layer of myoepithelial cells, which usually have prominent clear cytoplasm. The myoepithelial component may be evenly distributed in the background of the tubular proliferation with patchy areas of more solid nests. However, the myoepithelial component may predominate, take on a spindled appearance,
and the background matrix may assume the a bluish hue due to the presence of the myxoid/hyaline material. However, minimal pleomorphism and low mitotic rate (usually <3 mitotic figures per 10 high power fields) are seen in both elements [2].

A myriad of epithelial-myoepithelial proliferations occur within the breast and hence, need to be recognized and differentiated from adenomyoepithelioma. Adenoid cystic carcinoma may be excluded by the absence of the two types of mucins and the presence of apocrine epithelium. The appearance of pleomorphic adenoma may overlap with that of adenomyoepithelioma. A hyaline matrix with chondroid areas should favor the former diagnosis, while a more distinct encapsulation and definite epithelial tubules are usually seen in the latter. Further, various other lesions like papilloma and nipple adenoma that may contain prominent myoepithelial cells can add to diagnostic confusion [2].

Adenomyoepithelioma may be differentiated from a clear cell carcinoma by the presence of both epithelial and myoepithelial cell types, with immunohistochemical stains. Malignant transformation of only one cellular component, epithelial, has more often been described than myoepithelial. However, only adenomyoepitheliomas in which both the epithelial and myoepithelial cells were malignant have been reported previously [3]. Because of the heterogeneity of adenomyoepithelioma, the diagnosis should be made only on those lesions that do not fit into traditional categories. The presence of an expansion of a myoepithelial component on hematoxylin and eosin and the multinodular or papillary configuration is typical for adenomyoepithelioma. Immunostains can be used in support of the diagnosis although they should not be viewed in isolation. In our case, the histology was characteristic of adenomyoepithelioma. Both the components showed features of benignity. Hence, a diagnosis of benign adenomyoepithelioma was given.

Like the borderline phyllodes tumor, adenomyoepithelioma has been associated with local recurrence without possessing the true metastatic capability. Total surgical excision with an adequate margin of uninvolved breast tissue is therefore recommended.

Those lesions associated with more aggressive histologic features, including solid proliferations of malignant-appearing myoepithelial cells, lobulated, pushing borders or frank invasion, cytologic atypia, and increased proliferative rate tend to fall within Tavassoli’s definition of the “solid” pattern adenomyoepithelioma. The malignant type of adenomyoepithelioma can show nodal and distant metastases [2-6].

The imaging features of adenomyoepithelioma are nonspecific. Benign lesions commonly manifest as relatively circumscribed masses on mammograms and as hypoechoic masses on ultrasound (US) images while malignant tumors may show poorly defined margins and architectural distortion on mammograms and posterior shadowing on US images [7,8].

CONCLUSION

Adenomyoepithelioma is a rare benign tumor of the breast, which has to be differentiated from various benign and malignant myoepithelial lesions occurring in the same. This case is reported because of the unique bilateral presentation associated with lactation and lactational changes.

REFERENCES