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Pleomorphic Adenoma of the Accessory Axillary Breast: A Rare Case Report.

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ABSTRACT

Pleomorphic adenoma (PA) is the most common tumor type in the salivary gland. PA is uncommon in the breast tissue. Only 73 cases of PA of the breast have been reported in the world literature. But no case in the accessory axillary breast has been reported so far. It is most commonly seen in postmenopausal women and is characterized by an admixture of epithelial and myoepithelial cells embedded in abundant myxomatous stroma. Its clinical and histologic appearance can be challenging and may lead to a misdiagnosis of invasive carcinoma. We report a case of pleomorphic adenoma (benign mixed tumor) of the accessory axillary breast, which is an extremely rare location for this tumor.

Keywords: Pleomorphic adenoma, Accessory axillary breast, postmenopausal women.

INTRODUCTION

Pleomorphic adenoma (PA) is the most common tumor type in the salivary gland. PA is uncommon in the breast tissue.

CASE REPORT

A 45 year old woman presented with a painless left axillary swelling since 1 year. Examination of left axilla revealed a non-tender, soft and freely mobile swelling placed in the subcutaneous plane [Figure 1]. FNAC was done and the smear showed benign epithelial cells arranged in sheets in the background of abundant chondromyxoid stroma [Figure 2]. Excisional biopsy was performed, and the tumor histologically showed pleomorphic adenoma composed of duct epithelial cells, myoepithelial cells, and a myxochondroid matrix.

DISCUSSION

A benign mixed tumor or pleomorphic adenoma of the breast (PA) is an uncommon neoplasm, accounting for 73 cases in the literature. But to the best of our knowledge none of it has been reported in the axillary breast. It is a circumscribed lesion characterized by a mixture of epithelial and myoepithelial cells embedded in abundant stroma, which may be composed of 1 or more of the following matrices: (1) myxoid, (2) chondroid, or (3) osseous. PA was first reported in 1906 by Lecene who described a case with cartilaginous and osseous metaplasia.[1] Women are more commonly affected (female-male ratio >10:1) than men, and patients can range in age from 23 to 78 years (mean, 61 years). Tumors range from 0.7 to 20 cm in diameter and are most commonly located in the periareolar area or upper outer quadrant.[2]

On palpation, PA is described as being firm to hard and well circumscribed. Dimpling of the overlying skin and nipple retraction are uncommon features that may arouse suspicion of malignancy. On gross examination, PA may also be circumscribed, and it has a homogeneous yellow-white appearance on sectioning. Morphologically, it is composed of a mixture of epithelial, myoepithelial, and stromal elements. In the less circumscribed form, the tumor cells may surround lobules and ducts. Epithelial cells appear cuboidal to columnar with bland cytologic nuclear features and minimal mitotic activity. They are arranged as tubular structures, islands, cords, or sheets and may exhibit apocrine differentiation.[3] The surrounding myoepithelial cells appear polygonal, plasmacytoid, fusiform, or stellate with small nuclei and clear to eosinophilic cytoplasm. They are either admixed with epithelial cells or dispersed throughout the stroma. The stroma may vary from loose and myxoid, to cartilaginous with mildly atypical chondrocytes, to having an osseous matrix.

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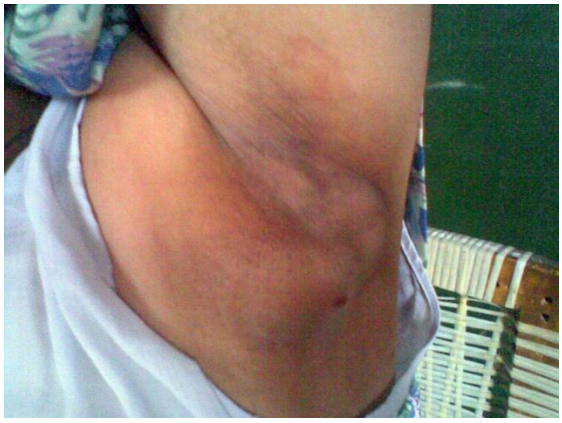

Figure 1: Axillary swelling.

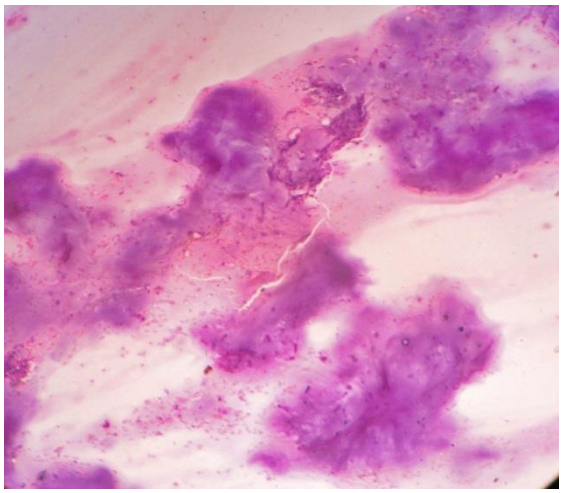

Figure 2: FNAC smear showing epithelial cells arranged in sheets in the background of abundant chondromyxoid stroma.

By immunohistochemistry, the epithelial cells are strongly positive for cytokeratin, carcinoembryonic antigen, and epithelial membrane antigen, and they are occasionally positive for vimentin and glial fibrillary acidic protein.[4] Myoepithelial cells are strongly positive for vimentin, muscle-specific actin, and cytokeratin. Cytokeratin and glial fibrillary acidic protein staining may be more intense in the myoepithelial cells than in the epithelial cells, which are also periodic acid-Schiff positive and diastase sensitive. S100 protein is variably expressed by all PA cell types but is typically more intense in myoepithelial cells.[4] Estrogen and progesterone receptor staining is unpredictable, with variable positivity in some cases.[2]

The cell of origin of PA has been the subject of numerous articles. Its predominant periareolar location has led to speculation that PAs may arise in conjunction with intraductal papillomas or adenomyoepitheliomas.[3] The comparatively larger ducts and abundant myoepithelial cells typically present in this region may explain this phenomenon and the possible role of the myoepithelial cell in the histogenesis of these benign lesions.[2] Despite the association and similar composite cell types, their histologic appearance remains somewhat distinct. Intraductal papillomas form distinct papillary structures, and adenomyoepitheliomas show a predominant proliferation of polygonal myoepithelial cells with clear cytoplasm, typically arranged in nests, which may surround gland-forming epithelial cells. Additionally, the presence of chondroid and osseous metaplasia in intraductal papillomas and adenomyoepitheliomas is extremely rare.

Other authors believe that the actual cell of origin of PA is a pluripotent stem cell, which accounts for the transition seen between epithelial and mesenchymal areas in the typical PA.[5] It is possible that the myoepithelial cell may actually represent that pluripotent cell, capable of differentiating along both epithelial (cytokeratin positivity) and stromal (vimentin positivity) lines, with the additional capacity of osseous and chondroid metaplasia.[4]

The benign nature of PA is clearly exemplified by the absence of metastases in all reported cases. Treatment therefore consists of wide excision with clear margins. Multifocal primary tumors and inappropriate resection by simple enucleation may increase the risk of tumor recurrence. Despite this risk, radical surgery is not indicated for the treatment of this lesion. Unfortunately, 30% of the reported cases in the literature were misdiagnosed as carcinoma and treated by mastectomy. Misdiagnoses are often the result of either suspicious mammographic clinical findings or the misinterpretation of frozen sections or fine-needle aspirates.[1,2,6,7]

CONCLUSION

Pathologists and clinicians need to be aware of this uncommon lesion, its myriad and somewhat deceptive clinical and mammographic features, and its variable histologic appearance, which may mimic malignancy. Immunohistochemical studies and special stains should serve as adjunctive tools to facilitate diagnosis in especially difficult cases. Because of the age of the patients, the risk of recurrence, and the possibility of multifocal disease (deceptively hidden in intraductal papillomatosis), a close clinical follow-up is warranted once a diagnosis of PA is made.

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