Adenomyoepithelioma Arising in Axillary Breast Tissue- A Diagnostic Rarity: case Report with Literature Review

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ABSTRACT

Adenomyoepithelioma is an uncommon, benign tumor of biphasic nature which arises from myoepithelial and epithelial cells. It has been recognized mainly in breast tissue, along with skin adnexa, lungs and salivary glands. The typical histologic appearance consists of acinar structures composed of an inner layer of epithelial cells with eosinophilic cytoplasm and a prominent peripheral layer of myoepithelial cells with clear cytoplasm. Prognosis of patients with AME is usually good, but it has a potential for local recurrence and rarely, malignant transformation with distant metastases to lung, brain, and liver. We present the rare case of a 42 year old female with left sided axillary lump that was diagnosed as adenomyoepithelioma arising from accessory breast tissue in the axilla. This of interest not only because of the rarity of the lesion, but also due to the peculiarity of its origin from accessory mammary gland tissue of axillary location.

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**Introduction**

Adenomyoepithelioma (AME) is an uncommon, diagnostically challenging tumor which demonstrates dual differentiation into luminal cells and myoepithelial cells. It was first described in breast by Hamperl in 1970. While AMEs have been reported in primary breast tissue, their location in axillary breast tissue is a diagnostic rarity as well as curiosity. Although AMEs are benign in majority of cases, sporadic malignant AMEs with distant metastases have also been reported. Recognition of this unique entity, its accurate diagnosis, and the knowledge of its expected behavior are important in guiding the most appropriate patient management.

**Case Report**

A 42 year old female presented in the rural outreach OPD of our hospital with a 3x3 cm sized painless, mobile lump in the left axilla for 1.5 years. There was no tenderness, erythema, warmth or discharge associated with the mass. Bilateral breasts and contralateral axilla revealed no abnormality on clinical examination. Family history was unremarkable. Routine hematological parameters, chest X-ray and ultrasound of abdomen revealed no abnormality. Local excision was done and the lump was submitted to the pathology department. Grossly, a 2x1.5x1 cm sized, rounded firm mass with irregular outer surface and greyish-brown cut surface was seen (Fig 1). Histopathological examination revealed a well-circumscribed tumor with pushing margins composed of closely packed ducts lined by a bilayer of inner cuboidal to columnar cells and outer polygonal cells with clear cytoplasm in a fibrous stroma (Fig 2). Adjacent foci revealed presence of mammary gland tissue showing adenosis. Deep and lateral resection margins were free of tumor. Rest of the tumor sample was sent for IHC to another center, and it was positive for S100, SMA, pan-Cytokeratin (AE1/AE3), estrogen receptor (ER) and progesterone receptor (PR). Based on the histopathology and IHC, the tumor was diagnosed as Adenomyoepithelioma of left axillary breast tissue. Postoperatively, her recovery was uneventful. She has been on follow up for the past 7 months with no evidence of tumor recurrence till date.

**Discussion**

Adenomyoepithelioma is an uncommon, benign tumor which arises from myoepithelial and epithelial cells. It has been recognized mainly in breast tissue, along with skin adnexa, lungs and salivary glands. In skin, it is known as apocrine mixed tumor and in salivary glands as epithelial-myoeipithelial carcinoma. AME has a biphasic nature and the typical histologic appearance consists of acinar structures composed of an inner layer of epithelial cells with eosinophilic cytoplasm and a prominent peripheral layer of myoepithelial cells with clear cytoplasm. Epithelial cells are cuboidal to columnar and tend to have hyperchromatic nuclei with dense eosinophilic cytoplasm, when compared with myoepithelial cells.

AME diagnosed in axillary lump in the present case can be speculated to be arising from skin adnexa, axillary breast tissue or as a metastatic lesion from breast, salivary glands or lung. But the presence of adjacent foci of uninvolved breast tissue along with positivity for hormone receptors...
and failure in detection of any primary source in bilateral breasts, lungs or salivary glands clinched the diagnosis in favor of AME arising from accessory breast tissue of axillary location.

The histogenesis of this tumor is still obscure. All cases have been sporadic and no familial aggregation has been observed. Kiaer et al reported a case of sequential changes from adenomyoepithelial adenosis into benign AME which eventually became low grade malignant AME during the course of 18 years. From this observation, Choi et al proposed that adenomyoepithelioma was derived from a long-standing underlying breast disease such as adenosis and fibroadenoma. Tavassoli has described three variants of AMEs- tubular variant (proliferation of rounded tubules, as well as unusually prominent and hyperplastic myoepithelial cells), spindle cell variant (predominantly spindled myoepithelial cell proliferation admixed with a few columnar epithelium-lined tubules) and lobular variant (solid nests of myoepithelial cells proliferating around compressed tubules). Combinations of growth patterns sometimes exist within the same tumor.

The interplay between epithelial and myoepithelial cell elements is highlighted by immunohistochemical staining with antibodies specific for these 2 components. The cytoplasm of epithelial cells uniformly reacts with antibodies to cytokeratins and the luminal surfaces of the glandular cells are positive for the epithelial membrane antigen. Luminal cells also possess receptors for ovarian steroid hormones (ER and PR). The myoepithelial component is highlighted by p63, smooth muscle myosin heavy chains, CK5, CD10, calponin, actin, and S100.

Because of the rarity and varied nature of AME’s, they can be confused with other neoplasms. The differentiation of intraductal papilloma with prominent myoepithelial cells from AMEs can be made based on architecture, pattern, and degree of myoepithelial proliferation. Myxochondroid matrices produced by the myoepithelial cells may also be noted, as seen in pleomorphic adenomas. Clear cell carcinoma may also mimic AMEs, but that may be differentiated by the presence of both epithelial and myoepithelial cell types, and confirmed with immunohistochemistry. Some areas of an AME may resemble adenoid cystic carcinoma, but that has infiltrative borders, a characteristic cribriform architecture with smaller, more hyperchromatic, and basaloïd appearing myoepithelial cells than AME. In case of small biopsy, the sampled tissue may even be mistaken for invasive carcinoma. The presence of regularly spaced, rounded or ovoid glands; unidirectional streaming of the glands; and prominent clear cell or spindle cell myoepithelium are some morphologic clues to the diagnosis of AME.

Prognosis of patients with AME is usually good, but it has a potential for local recurrence, especially in the tubular and lobulated variants. Total surgical excision with an adequate margin of resection is therefore recommended. Malignant transformation has also been reported along with distant metastases to lung, brain, and liver. Carcinomas may arise from ductal epithelial cells, myoepithelial cells, or both. Atypical features include infiltrative margins, increased mitotic activity, cytologic atypia with nuclear pleomorphism, prominent nucleoli, hyperchromasia, and necrosis.

**Conclusion**

This unique case of adenomyoepithelioma arising from axillary breast tissue is of interest not only for its rarity, but also due to the peculiarity of its origin from accessory mammary gland tissue of axillary location. Its diagnosis and optimal therapy are problematic issues for the clinicians due to the possibility for local recurrence and rare chance of metastatic spread. Recognition of biphasic cellular elements and the overall tumor architecture in combination with immunohistochemistry are important when diagnosing AMEs. Any case with significant cytologic atypia, rapid growth, infiltrating margins, necrosis, and brisk mitotic rates raises the likelihood of the lesion being malignant which is associated with the potential of metastasis. Hence, the histopathologist should duly report any atypical features and recommend complete excision with adequate margins so as to decrease any possibility of recurrence and metastasis in future.

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**References**


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