

**Adenomyoepithelioma Of Breast : Report Of A Rare Case**  
**Reena Kouli<sup>1</sup>, Swagata Dowerah<sup>2</sup>, Luckymoni Duara<sup>3</sup>**  
**Department of Pathology, Assam Medical College<sup>1,3</sup> and Department of Pathology,**  
**Silchar Medical College<sup>2</sup>**

**ABSTRACT**

Benign adenomyoepithelioma of the breast is a rare tumor characterized by biphasic proliferation of both an inner layer of epithelial cells and a prominent peripheral layer of myoepithelial cells. This entity may rarely progresses to a more malignant state or gives rise to metastasis. Accurate diagnosis and close follow up is essential for proper treatment of these tumors. We report a 15 year old girl who presented with large lump in the breast which was clinically mistaken for a giant fibroadenoma of breast. No nodes were palpable. On gross examination, a large lump 11x11x8 cm was noted which was well circumscribed, solid on cut section. On histopathologic examination the diagnosis of adenomyoepithelioma was made which was further supported by immunohistochemistry findings.

**Key words:** adenomyoepithelioma, breast, immunohistochemistry

**Corresponding author address:** Reena Kouli , Department Of Pathology, Assam Medical College Dibrugarh, Assam. Mobile:9435330569

**Conflict of interest:** No

**Case report is Original:** YES/NO

**Whether case report publishes any where?** YES/NO

**INTRODUCTION**

The adenomyoepithelioma is a biphasic tumor, either benign or with low potential of malignancy, which can be found in salivary gland, skin adnexa and lung, but is more frequent in the mammary gland. This entity rarely progresses to a more malignant state or gives rise to metastasis [1]. The classification of the World Health Organization (WHO, 2012) divides the adenomyoepithelioma into a benign type where both the epithelial and myoepithelial component are histologically non-malignant and a form which shows a malignant transformation [2]. Benign adenomyoepithelioma of the breast is a rare tumor characterized by biphasic proliferation of both an inner layer of epithelial cells and a prominent peripheral layer of myoepithelial cells. It was first described by Hamperl [3] in 1970; more than 60 cases have been reported since then.

**CASE REPORT**

A 15 year old girl presented with lump in the right breast which she had first noticed 4 months back. On examination, a mobile, firm, painless lump was palpable which measured around 10x10 cms and occupied most of the breast. Neither axillary nor supraclavicular lymph nodes were palpable. A clinical diagnosis of giant fibroadenoma was made and the lump was excised. FNAC and mammographic findings were not available when the specimen was sent to our department for histopathological examination. On gross examination, a 11x 11 x8 cm mass was seen which was grayish white in colour, well circumscribed, lobulated and solid on cut section with areas of translucency . H & E staining of the sections revealed tubules lined by proliferating myoepithelial cells and luminal epithelial cells. The myoepithelial cells had clear abundant cytoplasm and were peripherally located in the acini, the epithelial cells had eosinophilic cytoplasm and were located centrally. Many of the tubules were cystically dilated and were surrounded by a fibrous stroma. There was no evidence of malignant change. A diagnosis of adenomyoepithelioma was made, tubular type. Immunohistochemistry was performed for EMA, S-100 and SMA. The luminal cells stained positive for EMA whereas S100 and SMA were positive in the myoepithelial cells. Post surgery, the patient is doing well and there is no evidence of recurrence so far.



Fig. 1 : Gross specimen of the tumor showing a large lobulated grey tan tumor mass with solid cut surface.

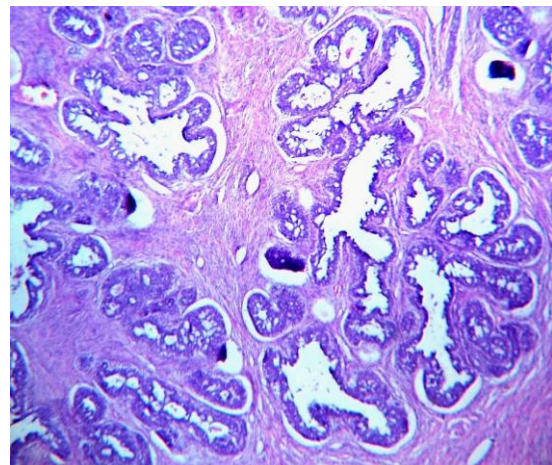


Fig 2 : Photomicrograph showing tubules composed of epithelial and myoepithelial cells surrounded by a fibrotic stroma (H&E, 10X).

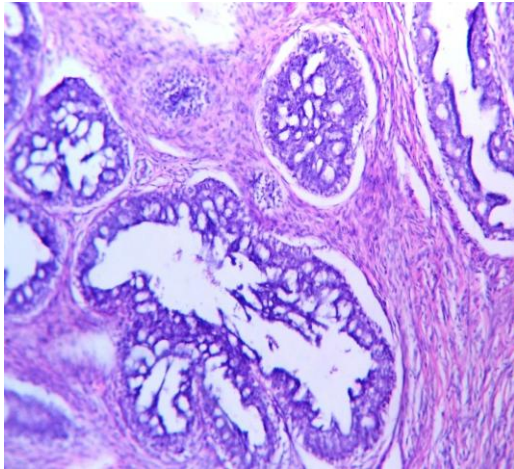


Fig. 3 : Higher magnification showing tubules lined by inner epithelial cell layer and outer myoepithelial cells (H&E 40X)

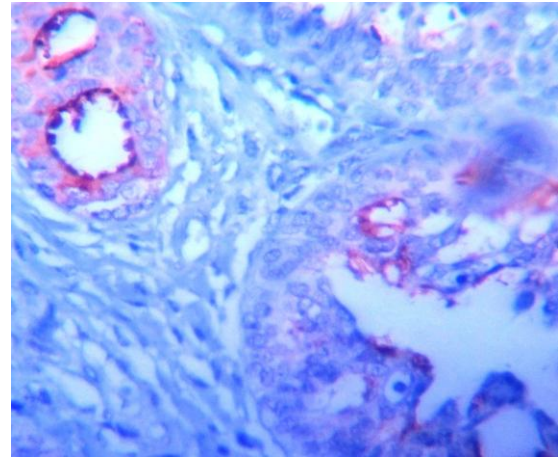


Fig. 4a IHC for EMA showing positive staining of the epithelial cells

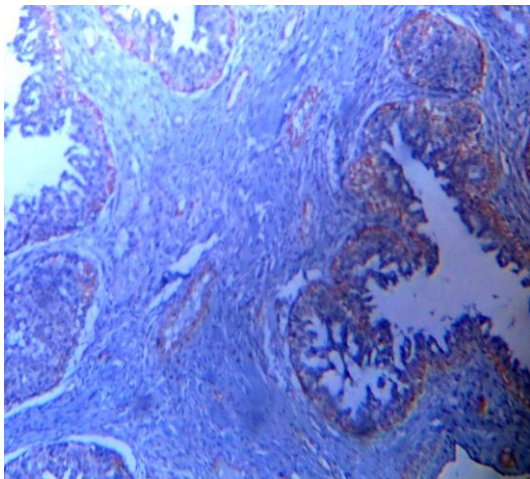


Fig. 4b showing positive staining for SMA in myoepithelial cells

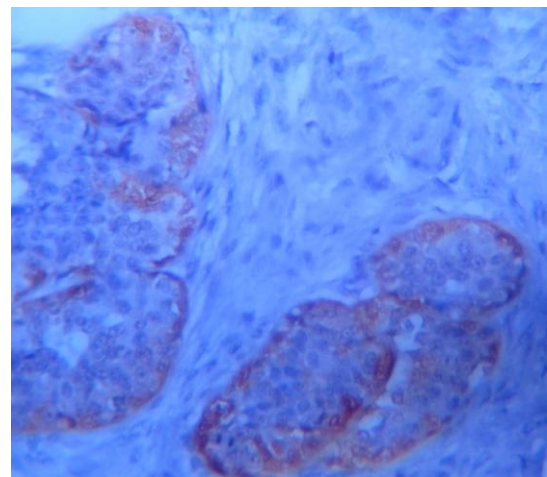


Fig. 4c showing positive S 100 staining of the myoepithelial cells

## **DISCUSSION**

Adenomyoepithelioma, a biphasic neoplastic proliferation of luminal and myoepithelial cells, was first described by Hamperl [3] in 1970. . As Hamperl noted, this tumor may display a heterogeneous pattern because of the variable proliferation of epithelial and myoepithelial cells. The tumor has a biphasic nature, composed of cuboidal to columnar, epithelial-lined tubules surrounded by myoepithelial cells [4]. A spectrum of histologic patterns, however, has been observed among various examples of these tumors and even in different areas of

individual tumors[5]. These variations were based on the distribution of proliferating glandular and myoepithelial cells, the extent of spindle or polygonal configuration of myoepithelial cells, the prominence of papillary component, and the degree of fibrosis[5].

So far, other than case reports, only 4 comprehensive series studies [1,4,5,6] have been reported. Patient age ranged from 22 to 92 years (mean age, 59 years) in one series of study[4]. In our case however, the patient was only 15 years old. The size of adenomyoepithelioma ranges from 0.3 cm to 7 cm, with an average size of 2.5 cm[1,4]. The tumors have been described as round to lobulated, well circumscribed or discrete, and firm[1,5]. Our tumor was quite large in size measuring 11x11x8 cm. Tavassoli[1] proposed a classification system of myoepithelial lesions of the breast. These lesions were divided into: myoepitheliosis, adenomyoepithelioma and malignant myoepithelioma. Tavassoli[1] classified adenomyoepitheliomas as spindle cell, lobulated, and tubular (or adenosis) types with carcinoma arising in adenomyoepithelioma. The most common pattern is the tubular type with features characterized by proliferation of glandular cells and surrounding myoepithelial cells of abundant clear cytoplasm, as was seen in our case.

The interplay between epithelial and myoepithelial cell elements is highlighted by immunohistochemical staining with antibodies specific for these 2 components. Adenomyoepithelioma shows strong positivity for keratins CAM5.2 and for EMA in the epithelial component, whereas the myoepithelial cells express several specific markers, including muscle-specific actin and myosin, calponin, p63, CD10 and S-100 protein[7]. Our case showed positivity for EMA in the epithelial component and S 100 and actin in the myoepithelial cells.

The exact etiology of breast adenomyoepithelioma is as yet uncertain. All cases have been sporadic and no familial aggregation has been observed. Kiaer et al [9] reported a case of sequential changes from adenomyoepithelial adenosis into adenomyoepithelioma which eventually became low grade malignant adenomyoepithelioma during the course of 18 years. From this observation, Choi et al. [10] proposed that adenomyoepithelioma was derived from a myoepithelial, long-standing, underlying breast disease such as adenosis and fibroadenoma. The association of adenomyoepithelioma with adenosis has been documented[11].

Most adenomyoepitheliomas can be treated by local excision, but local recurrences have occurred 8 months to 5 years after initial excision[1,6,8]. In the Tavassoli[1] study, most tumors with recurrence were of the tubular type of lesions, extending into, and blending with, the adjacent normal ducts. In our case, there was no recurrence 10 months post surgery.

Differential diagnosis of adenomyoepithelioma includes benign lesions that contain prominent myoepithelial cells and invasive mammary carcinomas. The benign lesions considered in the differential diagnosis of these tumors include tubular adenoma, sclerosing adenosis, fibroadenoma, and pleomorphic adenoma. Tubular adenoma, sclerosing adenosis, and fibroadenoma have less prominent proliferative features compared with adenomyoepithelioma. Pleomorphic adenoma usually has prominent areas of chondroid and osseous differentiation. The prognosis of patients with adenomyoepithelioma of breast is usually good. Failure to achieve a free resection margin may result in local recurrence or

rarely malignant transformation. Therefore the need for correct diagnosis and follow up cannot be overemphasized in these patients.

### **CONCLUSION**

Adenomyoepithelioma of breast is a rare tumor of epithelial and myoepithelial elements which should be considered in the differential diagnosis of solid breast lumps. This case is presented not only for its rarity but also for the very young age at which the tumor presented as well as its large size, which is unusual for a benign adenomyoepithelioma. Further studies will be necessary to understand the true nature of these tumors and their pathogenesis. Accurate diagnosis and close follow up is essential for proper treatment of these tumors.

### **REFERENCES**

1. Tavassoli FA: Myoepithelial lesions of the breast. Myoepitheliosis, adenomyoepithelioma, and myoepithelial carcinoma. *Am J Surg Pathol* 1991, 15(6):554-568. PubMed Abstract | Publisher Full Text
2. Lakhani SR, Ellis IO, Schnitt SJ, Tan PH, van de Vijver MJ: *WHO Classification of Tumors of the Breast*. 4th edition. WHO Press; 2012
3. Hamperl H: The myoepithelia (myoepithelial cells): normal state; regressive changes; hyperplasia; tumors. *Curr Top Pathol*; 1970; 53: 161–220.
4. McLaren BK, Smith J, Schuyler PA, Dupont WD, Page DL. Adenomyoepithelioma: clinical, histologic, and immunohistologic evaluation of a series of related lesions. *Am J Surg Pathol*. 2005;29(10):1294–1299. [CrossRef] [Medline]
5. Rosen PP. Adenomyoepithelioma of the breast. *Hum Pathol*. 1987;18(12):1232-1237. [CrossRef] [Medline]
6. Loose JH, Patchefsky AS, Hollander IJ, et al. Adenomyoepithelioma of the breast: a spectrum of biologic behavior. *Am J Surg Pathol*. 1992;16(9):868–876. [CrossRef] [Medline]
7. Barbareschi M, Pecciarini L, Cangi MG, Macrì E, Rizzo A, Viale G et al: p63, a p53 homologue, is a selective marker of myoepithelial cells of the human breast. *Am J Surg Pathol*; 2001; 25: 1054-1060.
8. Young RH, Clement PB. Adenomyoepithelioma of the breast: a report of three cases and review of the literature. *Am J Clin Pathol*. 1988;89(3):308–314. [Medline]
9. Kiaer H, Nielsen B, Paulsen S, Sorensen I, Dyreborg U, Blichert-Toft M: Adenomyoepithelial adenosis and low-grade malignant adenomyoepithelioma of the breast. *Virchows Arch A Pathol Anat Histopathol*; 1984; 405: 55-67.
10. Choi JS, Bae JY, Jung WH: Adenomyoepithelioma of the breast—its diagnostic problems and histogenesis. *Yonsei Med J*; 1996; 37: 284-289.
11. Ahmed AA, Heller DS: Malignant Adenomyoepithelioma of the Breast With Malignant Proliferation of Epithelial and Myoepithelial Elements A Case Report and Review of the Literature. *Arch Pathol Lab Med*; 2000; 124: 632–636.