Original Research Paper

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Pathology

RECURRENCE OF ADENOMYOEPITHELIOMA IN MALE BREAST-A RARE CASE REPORT

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ABSTRACT

Objectives: To present recurrence of a rare case of adenomyoepithelioma in male breast.
Case Report: A 32 year old male presented with swelling in periphery of left breast since 6 years measuring 0.6 x 0.5 cm. Ultrasonography was suggestive of cystic swelling measuring 7.6 x 3.8mm in size, with colour Doppler showing no internal vascularity. Excisional biopsy of lesion was done .Histology showed biphasic tumor in lobular pattern with inner epithelial and outer myoepithelial cells. There was no evidence of nuclear atypia or mitosis. This case was suspected for adenomyoepithelioma and myoepithelioma for which Immunohistochemisrty for calponin, Anti P63, S100P and CK-HMW was done showing positive results favouring adenomyoepithelioma as final diagnosis. Recurrence of swelling at the same site after 6 months of excision was seen. On histology similar findings were seen.

KEYWORDS :

CASE REPORT:A 32 year old male was presented with swelling in periphery of left breast since 6 years.

It was gradually increasing in size,and not associated with pain,nipple discharge. There is no history of diabetes mellitus, hypertension, tuberculosis or any major surgical event in past.On local examination swelling was present in periphery of left breast measuring 0.8x0.5cm in size.It was soft to firm,mobile, nontender and overlying skin was normal. On sonomammography it shows an indistinct, irregular, hupoechoeic mass measuring 0.9x0.7cm with enhanced peripheral vascularity.

Wide Local excision of lump was done under local anesthesia and was sent for histopathological examination.

We received a fibro fatty tissue mass measuring approximately 0.8x0. 6cm.whole tissue was submitted for processing. Serial section were studied ,it shows a well circumscribed encapsulated tumor , with proliferation of epithelial element arranged in sheets and in tubules , showing dual cell lining inner cuboidal and outer clear myoepithelial cells. Amidst few dilated blood vessels . No evidence of atypia/mitosis. Differential diagnosis of adenomyoepithelioma or myoepithelioma was given.

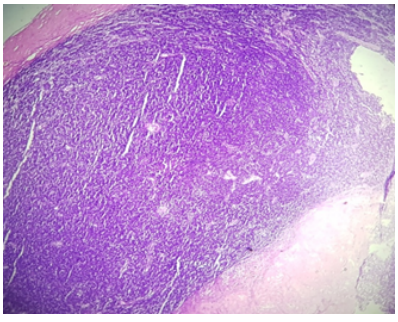

Fig1- (H & E)well circumscribed capsulated tumor(SCANNER VIEW

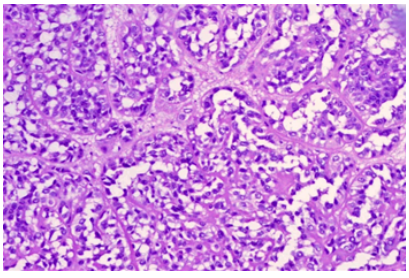

Fig2-(H & E)Tumor cells arranged in sheets and tubules pattern(100X)

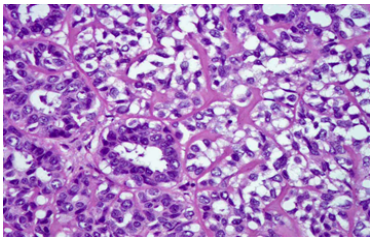

Fig 3-(H & E)tubules showing dual cell lining Inner cuboidal epithelial cells , outer myoepithelial cells with vacuolated cytoplasm.(400X)

Immunohistochemistry was done to confirm the diagnosis,so a panel of markers were done like pancytokeratin,calponin,p63,vimentin,CK HMW,S 100,Ki67,EMA. Tumor shows:

Pancytokeratin :positive in both the elements
Calponin: positive in myoepithelial elements
Anti P63: positive in myoepithelial elements
Vimentin: negative
CK HMW: positive in epithelial component
S 100: positive in myoepithelial elements
Ki67:positive,<1%
EMA: positive in epithelial cells lining the tubule

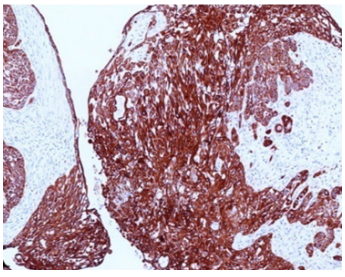

Fig 5-ihc Pancytokeratin Positive For Epithelial And Myoepithelial Components

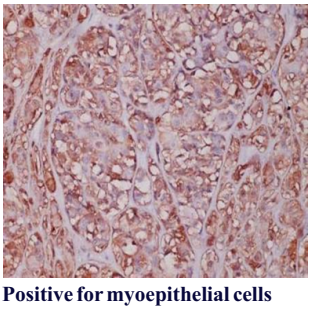

Fig 6-IHC S100 Positive for myoepithelial cells

Diagnosis: Adenomyoepithelioma Of Left Breast

Patient presented with similar swelling on same site 6 months after the removal of previous lump.It was approximately 0.6x0.4cm,mobile non tender. Wide Local Excision of lump was done and tissue was sent for histopathological examination.

We received single fibro fatty tissue measuring 0.7x0.5cm.Serial section of tumor studied shows similar histopathological findings as previous one. The diagnosis of Adenomyoepithelioma of left breast was given.

DISCUSSION

INTRODUCTION: Adenomyoepithelioma (AME), a biphasic neoplastic proliferation of luminal and myoepithelial cells, first described by Hamperl in 1970. Papillary architecture is seen in most tumors, and therefore, AME is considered to be a variant of intraductal papilloma. Striking morphologic heterogeneity, such as papillary configuration or multinodularity, especially in a limited biopsy sample, may not be appreciated, which may lead to erroneous diagnoses of carcinoma. Although most AMEs have been benign, sporadic malignant AMEs with distant metastases have also been reported. Recognition of this entity, accurate diagnosis, and knowledge of the expected behavior are important in guiding the most appropriate patient management.

CLINICAL FEATURES: Patient age ranged from 22 to 92 years (mean age, 59 years) .Nearly all of the affected patients have been women, although rare cases have been reported in men. Patients usually present with a solitary, palpable nodule, and the duration of symptoms varies from several weeks to several months. The tumors are usually located in a peripheral portion of the breast, although the lesions have been found centrally or near the areola.

ULTRASONOGRAPHY: On mammography, AME appears as a round or lobulated, dense, mostly circumscribed mass, sometimes with partially indistinct margins. Calcifications and cystic appearance are not typical findings.

MICROSCOPIC PATHOLOGY

The AME tumor has a biphasic nature, composed of cuboidal to columnar, epithelial-lined tubules surrounded by myoepithelial cells.A spectrum of histologic patterns, however, has been observed.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.

Three variants of AMEs were described by Tavassoli.

The first variant is the tubular pattern, which is characterized by a balanced proliferation of rounded tubules, as well as unusually prominent and hyperplastic myoepithelial cells.

The second variant is the spindle cell type, which is composed of a predominantly spindled myoepithelial cell proliferation admixed with a few columnar, epithelial-lined tubules.

Finally, the third variant exhibits a lobular pattern composed of solid nests of myoepithelial cells proliferating around compressed tubules; the solid nests of tumor are then surrounded by fibrous connective tissue septa of varying thicknesses.

Most AMEs have papillary configuration and, therefore, have been considered a variant of intraductal papilloma or a morphologic evolution from intraductal papilloma .Myoepithelial cells forming nests or sheets frequently display a spindle to myoid appearance at places with clear cytoplasm. This solid proliferation may displace, compress, or obliterate the epithelial gland, resulting in a zone nearly devoid of glands These areas, if pronounced, may lead to a differential diagnosis of myoepithelioma. The myoid areas may exhibit myoepithelial cells with pink to amphophilic cytoplasm or a plasmacytoid appearance with dense, hyaline-like, glassy eosinophilic cytoplasm and eccentric nuclei.Myxochondroid matrices produced by the myoepithelial cells may also be noted, as seen in pleomorphic adenomas .Dense, collagenous, hyaline-like matrix materials can be seen in thick basement membranes. Epithelial cells tend to have hyperchromatic nuclei and dense eosinophilic to amphophilic cytoplasm, when compared with myoepithelial cells.Apocrine metaplasia of epithelial cell component, as well as squamous and sebaceous metaplasia may be variably present. Atypical features, including increased mitotic activity, cytologic atypia with nuclear pleomorphism, prominent nucleoli, hyperchromasia, and necrosis, if observed, are usually associated with cases that either recurred or had a malignant clinical outcome.

IMMUNOHISTOCHEMISTRY:

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, such as cytokeratin AE1/3, CAM 5.2, or CK7.6. The luminal surfaces of the glandular cells are positive for the epithelial membrane antigen.Polygonal and spindle myoepithelial cells are not reactive to epithelial membrane antigen and are often only subtly reactive or weakly reactive to cytokeratin AE1/3.6,The myoepithelial component is highlighted by p63, smooth muscle myosin heavy chains, CK5, CD10, calponin, actin, and S100.McLaren et al reported that p63 produced the best results with consistent, intense nuclear staining, whereas some studies have reported discontinuous staining patterns.The distribution and intensity of staining for anti-actin antibodies is heterogeneous, more conspicuous in spindle cells than in clear polygonal cells. However, no reactivity to actin is seen in epithelial cells.Smooth muscle myosin–heavy chain is the most sensitive marker, which is easier to interpret than smooth muscle actin and muscle-specific actin because of its low crossreactivity with myofibroblasts.Calponin is also highly sensitive for detecting myoepithelial cells with cytoplasmic staining, but calponin myofibroblasts can be detected in up to 74% of breast proliferations by light, patchy staining patterns.S100 was expressed in virtually all of the myoepithelial cells, with variable intensity and uniformity of reactivity. The myoepithelial-specific antibodies display various cross-reactivity patterns and variable protein expression, especially in the neoplastic myoepithelial cells compared with their normal counterparts. Therefore, a panel-based approach of 2 or more markers should be used to minimize the chance of failure in detecting the myoepithelial cells.Proliferative indices of Ki-67 immunostaining are present in both compartments of the tumor but may be higher in the myoepithelial cells than it is in the ductal cells.Immunostain for estrogen is either negative or weakly positive in a patchy pattern. Progesterone receptor and ERBB2 (formerly Her2/neu) have, however, been consistently reported to be negative in all the published studies.