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ORIGINAL ARTICLE

Malignant Adenomyoepithelial Tumour of the Breast — a Rare Diagnosis — Case Report

V Kishore Reddy Bojja[1] · Sandip Bipte[1] · Aditya Vijay Reelkar[1] · Gaurav Patel[1]

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Abstract
Breast adenomyoepithelioma is an unusual tumour characterized by a biphasic proliferation of epithelial and myoepithelial cells. Most of the breast adenomyoepitheliomas are considered to be benign and characterized by propensity for local recurrence. Malignant change can occur rarely in one or both cellular components. We here present a case of a 70-year-old previously healthy female who initially presented with a painless breast lump. The patient underwent wide local excision in view of suspicion of malignancy and sent for frozen section regarding the diagnosis and margins which surprisingly came as adenomyoepithelioma. Final histopathology came as low-grade malignant adenomyoepithelioma. The patient shows no sign of tumour recurrence in the follow up.

Keywords Adenomyoepithelioma · Breast · Epithelial · Myoepithelial Cells

Introduction

Myoepithelial cells are a normal component of breast tissue. Adenomyoepithelioma is a rare benign breast tissue tumour with proliferation of glandular and myoepithelial cells. It was first described by Hamperl in 1970 [1]. Majority of adenomyoepitheliomas are benign. Few cases of adenomyoepitheliomas are reported to have local recurrences, metastasis, and malignant transformation [2]. Malignant adenomyoepithelioma is of interest not only for its rarity but also for the peculiar aspects of the malignant component that in some areas shows morphological and immunophenotypical similarities to basal-like breast carcinoma. This neoplasm also exhibits a spectrum of morphology and display of biphasic appearance in different areas of the tumour, thus making it diagnostically challenging by core biopsy due to its heterogeneity [3].

Case Summary

A 70-year-old female presented with lump in the right breast for 3 months. Lump was mobile, painless, size of 5 × 3 cm in outer lower quadrant of right breast with firm consistency, and no skin involvement. Neither axillary nor supraclavicular lymph nodes were palpable. Craniocaudal mammography showed a 4.7 cm well-defined nodule with calcification in right breast with a BIRADS score IVC. Breast ultrasound was performed showing a rounded, hypoechoic solid lesion with ill-defined margins in the right inner-inferior quadrant (Fig. 1). On further evaluation with core needle biopsy, there were no features of invasive breast carcinoma. In view of suspicious imaging features, she underwent wide excision of the lump and sent for frozen section which came as adenomyoepithelioma.

Final histopathological analysis showed the 5 × 3.5 cm lesion with multinodular surface, infiltrating margins, and biphasic aspects due to the presence of a double population of cells (epithelial and myoepithelial) suggestive of malignant adenomyoepithelial tumour (Fig. 2). Tumour had low mitotic activity < 3/10 (low grade), no evidence of lymphovascular and perineural invasion (Fig. 3). All the resected

✉ V Kishore Reddy Bojja
kishorereddy.bojja021@gmail.com

Sandip Bipte
sbipte@ymail.com

Aditya Vijay Reelkar
aditya.reelkar@gmail.com

Gaurav Patel
gauravdp49@gmail.com

[1] Dept. of Surgical Oncology, Bombay Hospital and Medical Sciences, New Marine Lines, Mumbai 400020, India

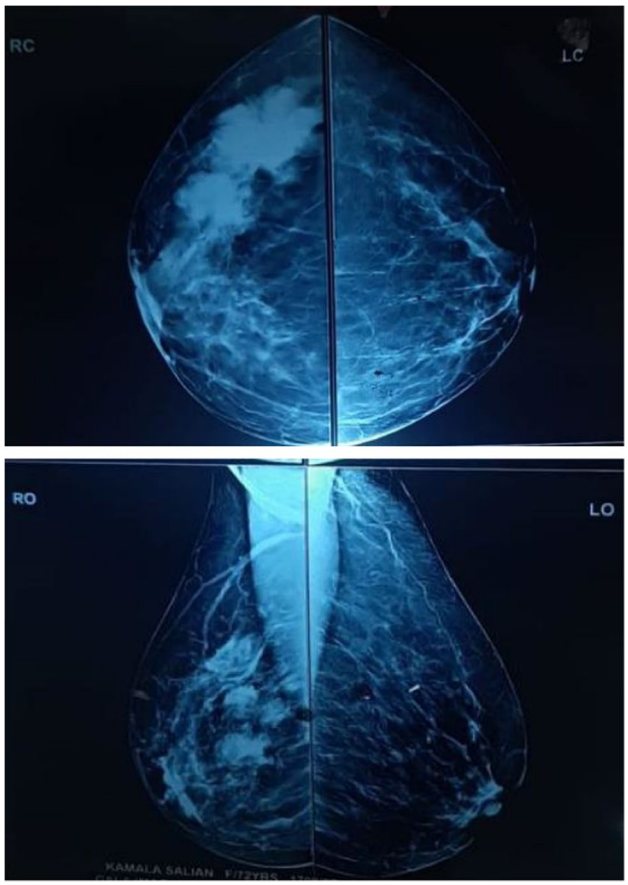

Fig. 1 Mammography showing heterogeneous lesion with irregular borders in right breast with skin and axilla appearing normal

margins are free of the tumour. An immunohistochemical study was then carried out which showed positive for cytokeratin 8/18, vimentin, cytokeratin 5/6, P63, and SMA.

During the follow up, patient had no sign of locoregional recurrence or distant metastasis.

Discussion

Myoepithelial cells are normally present in the breast and may appear spindle shaped or as large ovoid cells, sometimes with a clear cytoplasm. Neoplasms of pure myoepithelial or mixed epithelial and myoepithelial origin are described in the salivary glands but are very rare in the breast. The myoepithelial lesions are divided into three different categories: (1) myoepitheliosis, (2) adenomyoepithelioma, and (3) myoepithelialcarcinoma [2]. Tumours with bicellular proliferation of both epithelial and myoepithelial cells are called adenomyoepitheliomas [1, 4]. Though most of the adenomyoepitheliomas are benign, either of its two components may become malignant. Majority of the cases of adenomyoepithelioma are reported in the fifth to sixth decade of life. The exact aetiology

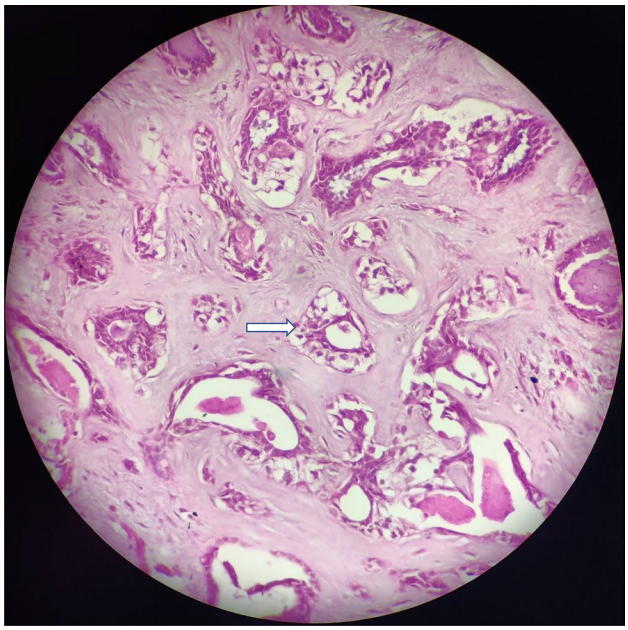

Fig. 2 Showing tumour tissue (arrow) containing biphasic cells (inner epithelial, outer myoepithelial). Myoepithelial cells having abundant cytoplasm

of adenomyoepithelioma is still obscure. The adenomyoepithelioma presents as solid well-delineated rounded nodules varying from 0.3 to 7 cm, with an average size of 2.5 cm, firm, whitish yellowish nodules. Adenomyoepitheliomas are classified as tubular, lobulated, or spindle subtypes [2]. The most common pattern is tubular type. Malignant adenomyoepithelioma has the potential for distant metastases. Morphological features of malignancy that could predict the potential for

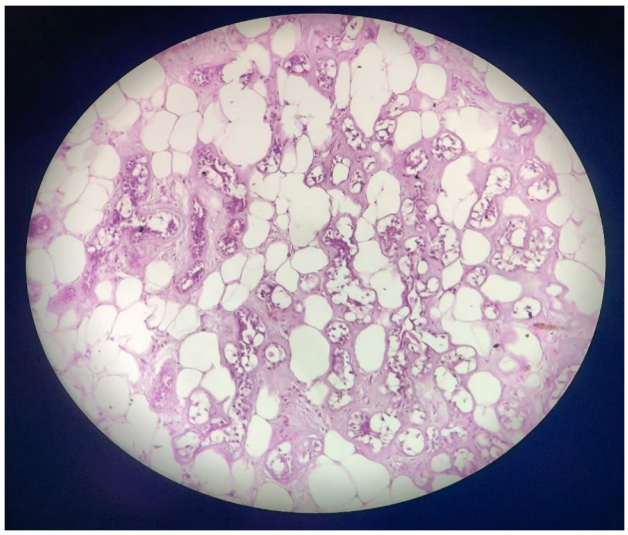

Fig. 3 Microscopic examination revealed invasive component of epithelial cells

local recurrence and/or metastasis are not well-established [5]. These typically occur in lesions larger than 2 cm and in those with high-grade malignant component [6–8]. Malignant predictors include a high mitotic rate, cytologic atypia, and infiltrative peripheral borders [9]. Axillary lymph node involvement is thought to be unusual, hence a recent review article has suggested that axillary node dissection is not indicated unless there are clinically involved lymph nodes [6, 9]. Distant metastases were described in 8 (32%) out of 25 cases reviewed by two authors [10, 11], involving lungs, brain, and soft tissues. On Immunohistochemistry, these tumours are immunoreactive for various markers like SMA, SMMH, p63, 34βE12, CK5, CK14, S-100, and caldesmin [12]. Currently, surgery remains the mainstay of treatment for malignant adenomyoepithelial lesions. A wide local excision with adequate margins is recommended. Both benign and malignant adenomyoepitheliomas are prone to local recurrence. Because these tumours are so rare and only around 30 cases have been described in the literature [13], available data concerning the prognosis of these tumours is scarce. So, wide excision of the lesion with adequate margins is a sufficient treatment.

Conclusion

Breast malignant adenomyoepithelioma is a rare tumour which should be considered in the differential diagnosis of other solid breast lesions. Only a few cases have been reported in the literature and establishing the diagnosis, determining the optimal therapy, and predicting the outcome are problematic issues due to the rarity of this disease. The adenomyoepithelioma appears to have hematogenous rather than lymphatic spread and usually occurs in primary tumour's ≥ 1.6 cm in size. The use of axillary sampling/axillary dissection in the treatment of the tumour without axillary node involvement needs to be addressed in the future.

Declarations

Conflict of Interest The authors declare no competing interests.

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