Case Report

Malignant adenomyoepithelioma of the breast with lymph node metastasis: A rare case report with review of literature

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ABSTRACT

Malignant adenomyoepithelioma of the breast is a rare lesion characterized by malignant proliferation of epithelial and myoepithelial cells that show characteristic histologic and immunohistochemical features. Metastases associated with these tumors are usually hematogenous. Axillary lymph node metastases are thought to be unusual. It has been recently suggested that axillary node dissection is not indicated unless clinically palpable. We present a case of a 35-year-old woman, who developed a malignant adenomyoepithelioma with axillary lymph node metastasis that included epithelial and myoepithelial elements.

Key words: Adenomyoepithelioma, breast, lymph node

INTRODUCTION

Adenomyoepithelioma of the breast is a benign neoplasm characterized by biphasic proliferation of epithelial and myoepithelial cells.[1] It resembles adenomyoepithelioma of salivary glands as first described by Hamperl in 1970.[2] Malignant change can occur rarely in one or both cellular components, and it can be either a pure myoepithelial carcinoma or a combined malignant adenomyoepithelioma.[1] Malignant neoplasms arising in adenomyoepitheliomas have been described in many patterns and have been subclassified as undifferentiated,[3] myoepithelial, or epithelial. Around 30 cases of malignant adenomyoepithelioma have been reported in the literature.[1,4,5]

CASE REPORT

A 35-year-old female presented with a complaint of lump in the left breast for 5 months. At the time of presentation, there was an outgrowth invading the nipple since 10 days along with gradual pain. Local examination revealed a firm to hard outgrowth, invading the nipple of left breast [Figure 1a]. Fine needle aspiration cytology revealed benign epithelial cells in sheets and clusters with papillae formation having bare nuclei with foamy macrophages suggestive of intraductal papilloma. She underwent lumpectomy with axillary lymph node dissection and the excised tissue submitted for histopathological examination.

Gross examination of lumpectomy specimen disclosed irregular, firm, and gray nodular mass of breast tissue measuring 5.0 cm × 5.0 cm × 5.0 cm with multiple lymph nodes measuring 3.0 cm × 2.0 cm × 1.0 cm. Microscopic examination shows cells having a small nucleus with occasional nucleolus, clear as well as eosinophilic cytoplasm and occasional mitosis. These cells are arranged in an alveolar pattern having thin to thick fibrous septa with myofibroblast [Figure 1b and c]. Surrounding

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tissue shows intraductal papillary hyperplasia and adenosis. In one of the axillary lymph node, there was a complete replacement of lymphoid tissue by tumor cells [Figure 1d].

The immunohistochemical (IHC) studies were performed with a battery of epithelial and myoepithelial markers. The myoepithelial markers namely smooth muscle actin (SMA), p63, and S-100 highlighted the prominent myoepithelial cells of adenomyoepitheliomatous component [Figure 2c-e]. In the malignant spindle cell, there was an expression of cytokeratin-7 (CK-7) and epithelial membrane antigen (EMA), CK-20 was immunonegative [Figure 2a, b, and f]. On follow-up, the patient was doing well without recurrence since last 6 months.

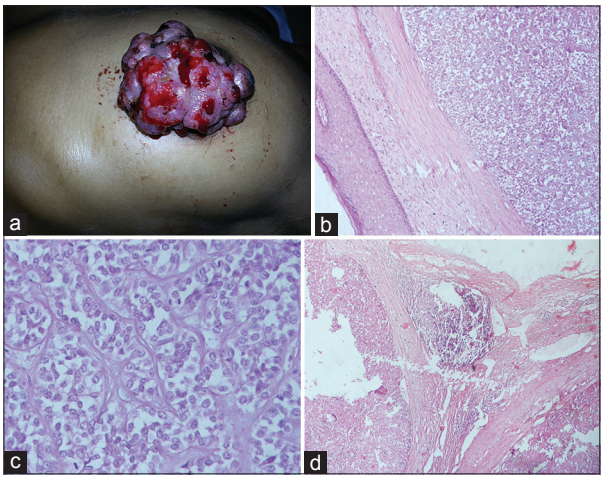

Figure 1: (a) Appearance of lesion invading the nipple, (b) low power (H and E, ×10) view of malignant adenomyoepithelioma, (c) high power (H and E, ×40) view of malignant adenomyoepithelioma, and (d) H and E stained slide showing replacement of lymph node tissue by tumor cells (×4)

DISCUSSION

Myoepithelial cells are a normal component of breast tissue, and their presence in neoplastic lesions has been considered a hallmark of benignity. Recently, however, breast neoplasms have been described that are entirely or partially composed of myoepithelial cells. Neoplasm of purely myoepithelial origin have been called myoepitheliomas and may be benign or malignant in approximately equal proportions.[4,6] Tumors with the bicellular proliferation of epithelial and myoepithelial cells are called adenomyoepithelioma.[4]

Adenomyoepitheliomas of the breast are rare neoplasms first described by Hamperl in 1970 and further classified by Tavassoli in 1991.[6,7] Though most of the adenomyoepitheliomas are benign, either of its two components may become malignant. The evolution of malignant adenomyoepithelioma seems to begin with adenosis, with or without myoepithelial hyperplasia, advances to benign adenomyoepithelioma and proceeds to a possible malignant transformation.[4] Malignant adenomyoepithelioma showed immunoreaction with CK-7, p63, S-100, and EMA; however, CK-20 was negative. IHC, myoepithelial cells stain variably to SMA.

Malignant adenomyoepithelioma has the potential for distant metastases. This typically occurs in lesions larger than 2 cm[4] and in those with high-grade malignant component.[1] These tumors have been reported in women, ranging in age from 26 to 76 years with tumor sizes varying from 1 to 15 cm. The review of 12 published cases of malignant adenomyoepitheliomas showed distant metastases in four cases with spread to lung and brain, evidence suggesting hematogenous spread being more common as compared to lymphatic spread.[4]

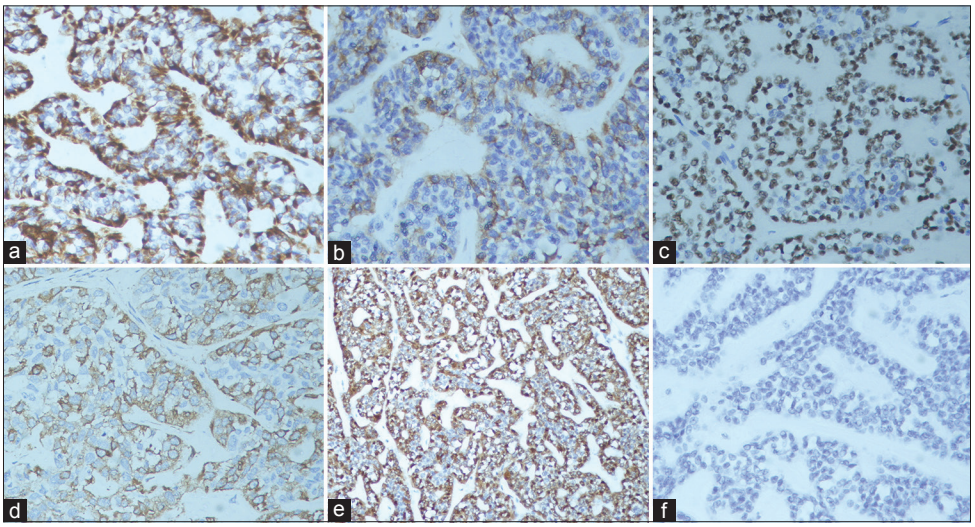

Figure 2: (a) Immunohistochemistry stain for cytokeratin-7 positivity, (b) immunohistochemistry stain for epithelial membrane antigen positivity, (c) p63 positive tumor cells on immunohistochemistry stain, (d) immunohistochemistry stain for smooth muscle actin positivity, (e) immunohistochemistry stain for S-100 positivity, and (f) tumor cells are immunonegative for cytokeratin-20

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 is clinically involved lymph nodes.[1] However, metastases to axillary nodes have been reported in two previous cases of malignant adenomyoepithelioma,[8,9] in addition, to the current where metastasis is also reported.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Hayes MM. Adenomyoepithelioma of the breast: A review stressing its propensity for malignant transformation. J Clin Pathol 2011;64:477-84.
2. Hamperl H. The myothelia (myoepithelial cells). Normal state; regressive changes; hyperplasia; tumors. Curr Top Pathol 1970;53:161-220.
3. Michal M, Baumruk L, Burger J, Manhalová M. Adenomyoepithelioma of the breast with undifferentiated carcinoma component. Histopathology 1994;24:274-6.
4. 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;124:632-6.
5. Choi SY, Kim JS, Kim SJ, Kim YJ, Kim L, Young UC. Malignant adenomyoepithelioma of the breast presenting as a large mass that grew slowly without metastasis. J Breast Cancer 2009;12:219-22.
6. Nadelman CM, Leslie KO, Fishbein MC. "Benign," metastasizing adenomyoepithelioma of the breast: A report of 2 cases. Arch Pathol Lab Med 2006;130:1349-53.
7. Tavassoli FA. Myoepithelial lesions of the breast. Myoepitheliosis, adenomyoepithelioma, and myoepithelial carcinoma. Am J Surg Pathol 1991;15:554-68.
8. Chen PC, Chen CK, Nicastri AD, Wait RB. Myoepithelial carcinoma of the breast with distant metastasis and accompanied by adenomyoepitheliomas. Histopathology 1994;24:543-8.
9. Attili VS, Saini K, Lakshmaiah KC, Batra U, Malathi M, Ramachandra C. Malignant adenomyoepithelioma of the breast. Indian J Surg 2007;69:14-6.