Adenomyoepithelioma of the breast: A rare case from the Mankweng Hospital breast oncology clinic, Limpopo Province, South Africa

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Adenomyoepithelioma of the breast is a rare benign breast neoplasm. It is characterised by the proliferation of both epithelial and myoepithelial cells. Although it is benign, adenomyoepithelioma has a potential for recurrences and malignant transformations. Here, we report a case of a 69-year-old woman with a breast adenomyoepithelioma that had been resected twice before her admission to Mankweng Hospital. It was a fast-growing tumour involving the retro-areolar area and left inner quadrant of the breast. Applied treatment included wide-simple mastectomy with sentinel axillary lymph nodes biopsy.

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Adenomyoepitheliomas of the breast are rare benign breast neoplasms. They are characterised by proliferations of both epithelial and myoepithelial cells. Epithelial cells resemble those normally covering the mammary ducts. Myoepithelial cells include specialised epithelial cells with a myoid differentiation. They contain normal components of the lobular units of the breast tissue lying between the epithelial layer and basement membrane of ducts along with acini.[1]

Although adenomyoepitheliomas are benign neoplasms, they have a potential for recurrences and malignant transformations. The malignant adenomyoepithelioma is difficult to differentiate from other benign diseases such as intraductal papillomas, tubular adenomas, or sclerosing adenosis.[2]

Case presentation

A 69-year-old, postmenopausal woman presented to hospital with an 18-month history of a tumour in the left breast. She reported being previously treated with a local excision of the breast mass 16 months ago at her community hospital when the mass was smaller, but a few months later the painful mass recurred and was growing rapidly. The patient had repeat resection of the recurrent tumour at the same local hospital 7 months before her admission to our institution, as the tumour continued to grow. There was no family history of breast cancer. The physical examination revealed breast asymmetry, a mobile and irregular breast mass measuring 14 cm × 9 cm in the retro-areolar and left inner quadrant region with an old 3 cm scar (Fig. 1). The axillary lymph nodes were not palpable. Mammogram revealed left breast high-density irregular mass with indistinct margin posteriorly. The size of the mass was 12.9 cm × 5.7 cm on a craniocaudal view, extend from the retro-areolar region and the left inner quadrant. The tumor was associated with scattered fine microcalcifications. There was no focal or diffuse skin thickening. The tumour was a BIRADS-5 breast tumour. The histology of an excisional biopsy revealed morphological features of adenomyoepithelioma with the neoplasm noted to extend to resection margins. Immunohistochemistry revealed that the tumour was positive for P63 and CD10, highlighting the presence of myoepithelial cells, and it was also CK7-positive, indicating the presence of luminal epithelial cells. Simple mastectomy with a sentinel lymph node biopsy was done successfully without any complications. Histological features of mastectomy specimens were in keeping with adenomyoepithelioma, showing all resection margins free of tumour and a reactive follicular hyperplasia in axillary lymph nodes.

Discussion

Neoplasms of pure myoepithelial or mixed epithelial origin have been described in the salivary glands but are rare in the breast.[3] The first case of breast adenomyoepithelioma was described by Hamperl[2] in 1970. Since then, about 150 cases have been reported in the literature.[4] Most patients with adenomyoepithelioma are elderly,[1,3] although it has also been reported in younger patients. The specified

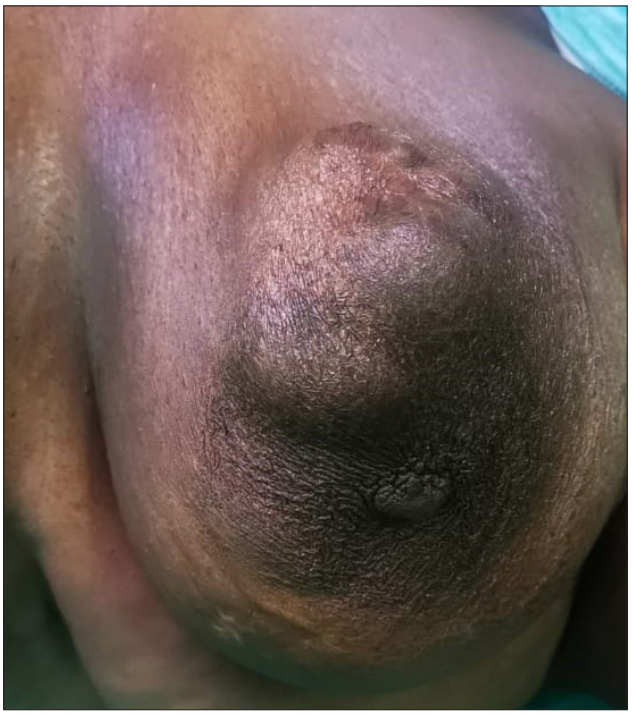

Fig. 1. Location of the mass in the retro-areolar area.

age ranges from 22 - 92 years, with a mean onset age of ~60 years.[2] Tumours appear usually as a solitary, nodular palpable mass. Mastalgia and nipple discharges are reported rarely.[1,2,5] Our patient presented with a nodular mass associated with pain. Adenomyoepitheliomas have been classified histologically as tubular, lobulated, or spindle-cell types of growth patterns.[5] The most common microscopic type is the tubular type. Spectrums of histological patterns depend on noticed relative amounts of proliferating glandular and myoepithelial cells.[6] Immunohistochemical investigations of adenomyoepitheliomas reveal epithelial cells that stain positive to cytokeratin (CK), and myoepithelial cells that are stained with antibodies to smooth-muscle actin, smooth-muscle myosin, calponin, p63, desmin, and S-100.[4,7] Indeed, we found the presence of both epithelial and myoepithelial cells in our case as indicated by cells that were positive for CK, and P63 and CD10, respectively.

The radiological features of breast adenomyoepithelioma are not well described. Predominant ultrasound and mammogram findings demonstrate an irregular mass with suspicious imaging findings.[1,4] In our case, investigations showed the irregular mass and fine microcalcifications, BIRADS-5 highly suggestive of malignancy.

Like other adenomyoepitheliomas described in the literature, recurrence may be associated with incomplete resection of the original tumour. Reported recurrences appear as soon as 4 months and as late as 23 years after primary tumors excisions.[4] Our patient had the recurrence twice within 18 months of presentation. The recommended surgical treatment of adenomyoepithelioma involves wide-local excisions with confirmed negative resection margins. Simple mastectomies or extended resections are advocated for cases with incomplete margin excisions.[2] We offered our patient the simple mastectomy with a sentinel lymph node biopsy. Surgical margins were free of tumour cells with no malignant deposits to axillary lymph nodes. The patient is continuing with follow-ups at the Mankweng breast oncology clinic to assess the outcomes.

Conclusion

Adenomyoepitheliomas of the breast have a risk of recurrence and are best treated with wide-local excisions obtaining clear surgical margins. Close follow-up is essential to assess recurrences and/or malignant transformations.

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