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A Rare Case Of Recurrence Of Pleomorphic Adenoma Of The Breast With Malignant Transformation

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Abstract

PA mainly occurs in the salivary gland, yet it also can be found in skin, larynx lacrimal gland, vulva , lungs, sella turcica ,paranasal sinuses, nasal septum, and kidney. It rarely occurs in breast and malignant transformation is even rarer. We are presenting a case of a 68 year old female who presented with malignant transformation of pleomorphic adenoma of breast.

Keywords: Pleomorphic adenoma, breast.

Introduction

Pleomorphic adenoma (PA) or benign mixed tumor is common tumor of salivary glands. The occurrence of PA located in breast is extremely rare and PA of breast was mostly found in subareolar region in postmenopausal females.[1]The uncommon sites of PA are larynx, paranasal sinuses, palate, and nasal septum. It can also occur in skin where it is known as chondroid syringoma.[2] We are reporting a case of recurrence of pleomorphic adenoma of breast.

Case presentation

A 68 year old post-menopausal female presented to surgery opd with complaint of lump in right breast since 15 days. There was no history of increase in size of lump, no history of nipple discharge and no history of fever.

On local examination, the lump was retroareolar and measured 4X4 cm in size. The lump was firm, mobile and non-tender. Nipple areola complex was normal. No lump was palpable in left breast. No axillary lymph nodes were palpable.

FNAC was done which revealed proliferative breast disease without atypia. A trucut biopsy was done. Sections from biopsy revealed fibrocollagenous tissue lined by benign pseudostratified columnar epithelium and a focus of chondroid tissue. There was no evidence of malignancy.

Lumpectomy was done and sent for histopathological examination. Grossly, the lump was irregular, yellowish brown, firm to hard in consistency and gave gritty sensation on cutting. Total measurement of lump was 5X4X3 cm.

Microscopic examination revealed a well circumscribed mass composed of epithelial elements within a myxoid, chondroid and fibrous stroma. The epithelial element showed branching tubules, solid cords and many non branching tubules lined by cuboidal cells with round to oval nuclei with or without prominent nucleoli. Few tubules were lined by cells with eosinophilic cytoplasm and showed secretions in the lumen. Clear cell change was also seen at places. There was large amount of intervening myxoid connective tissue. At places osseous tissue with bone marrow elements was also seen. The histological features were suggestive of Chondroid syringoma.

The patient presented again after 1&1/2 year with lump in right breast in upper outer quadrant.

On local examination, the lump was 6X5 cm, firm, freely mobile and non tender. Lumpectomy was done and sent for histopathological examination. Grossly the lump was irregular, yellowish brown , firm in consistency and showed whitish firm nodule on cut surface. Total measurement of lump was 7X5X3 cm.

Microscopic examination revealed a poorly circumscribed lobulated tumor mass composed of epithelial tumor cells arranged in cords, nests, tubular and tubulobranching pattern.

The tumor cells were round to oval with round nuclei and scanty to moderate amount of cytoplasm. There was mild anisonucleosis. Areas of hemorrhage and necrosis were also seen. The intervening connective tissue showed hyaline, mucoid, myxoid and vascular areas. Surrounding adipose tissue showed breast acini. The histological features were suggestive of malignant epithelial tumor. Considering previous clinical findings the diagnosis of malignant adnexal tumor and infiltrating ductal carcinoma was given. Immunohistochemistry (IHC) was done for ER, PR and Her2/neu and was negative for all these markers.

Discussion

PA mainly occurs in the salivary gland, yet it also can be found in skin, larynx lacrimal gland, vulva , lungs, sella turcica ,paranasal sinuses, nasal septum, and kidney.[1,2] The breast is a modified sweat gland which is embryologically similar to the ectodermal layer of salivary glands.[1]

Pleomorphic adenoma of the breast (PAB) is a rare neoplasm and occurs commonly in middle aged and elderly postmenopausal patients. The male to female ratio of about 1:10. It usually develops in the retroareolar region of the breast.[3] Most of the cases of PA were reported in women; however, three cases were of men with an age ranging between 23 and 85 years.[4]

It has been suggested that PA probably starts as an intraductal papilloma. The myoepithelial cells of the papilloma are extraordinarily stimulated resulting in formation of the characteristic stromal elements. It has been further suggested that multifocal PA may arise from multiple intraductal papillomas.[7]

PA usually presents as small nodules of a few centimetres in the greatest dimension. Occasionally, PA may be multifocal.[6]

There are many recurrences reported and few malignancies, based on the behavior of this kind of tumor and on its pseudopod like extension in the adjacent tissue. Based on the histopathological findings (small tumor size, well defined, with reduced nuclear atypia, without obvious tumor necrosis), there are few differential diagnosis of pleomorphic adenoma of the breast, such as: adenocarcinoma with cartilage/osseous metaplasia, stromal sarcoma, phyllodes tumor and fibroadenoma.[5]

Gross examination usually discloses a lobulated,well circumscribed hard nodule), with a gritty cut surface. Histologically, breast PA is similar to its salivary gland counterpart . Its low power appearance is characterised by glandular structures and anastomosing cords of epithelial

cells immersed in myxoid stroma, surrounded by a fibrous pseudocapsule that can be invaded or interrupted by nests of neoplastic cells. The glandular structures show an inner luminal layer of epithelial cells surrounded by an outer layer of myoepithelial cells. Myxoid stroma contains spindle or stellate cells. Cartilaginous and osseous areas are frequently encountered.[6]

On immunohistochemistry chondroid syringoma is positive for alcian blue staining and they did not stain after they were digested with hyaluronidase, were prominent in the matrix among tumour cells for cytokeratin(AE1+AE3),S-100 protein, neuron specific enolase and glial fibrillary acidic protein(GFAP).GFAP is characteristic of malignant chondroid syringoma,39% of cases with this staining positive have metastatic lesions and 22% died of this malignant tumour. Intraluminal cells are CEA positive. The stromal cells are cytokeratin negative and sporadically positive for vimentin. Positive chondroid areas are S-100 protein and vimentin positive.[8]

During FNA the malignant focus may be missed and the aspirates in such a case will be misdiagnosed as benign. An excision biopsy of all lesions hence is the recommended approach.[9]

Metaplastic carcinoma of the breast should be considered in the differential diagnosis of a breast tumor with an abundance of cartilaginous or osseous metaplasia. Usually metaplastic breast carcinoma consists of an anaplastic invasive duct carcinoma present in conjunction with foci of pseudosarcomatous transformation of the epithelial component that yields malignant cartilage or bone formation. In contrast, pleomorphic adenoma of the breast exhibits no evidence of anaplasia within the metaplastic stroma and only mature, benign-appearing cartilaginous or osseous tissue is present.[10]

Another differential diagnosis is intraductal papilloma with myxochondroid stroma. Intraductal papilloma can be distinguished by their lack of proliferatifve myoepithelial elements,which is characteristic of PAB. Extensive surgical excision with clear margin is the single therapeutic option.[11]

K Kusafuka et al examined four cases of matrix producing carcinoma of the breast, histochemically and immunohistochemically, and the matrices of matrix-producing carcinoma were similar to those of normal fibrous cartilage from the immunohistochemical findings of type II and type I collagens, aggrecan, and ChM-I depositions. Aberrant expression of sox9 in metaplastic cells induces such cartilage-specific matrix molecules.[12]

An excision of the tumour with adequate margins is the treatment of choice in cases of PA of breast. Although it is a benign tumour, an inadequate excision can result in recurrence and even multiple recurrences have been described.

Though a margin as less as 3mm has been described as adequate, given the ability of the tumour to have extracapsular extension and pseudopods, N.Leekha et al suggested that a wider margin may be necessary to prevent recurrence.[7]

Conclusion

Pleomorphic adenoma is rare benign tumor of the breast and may be confused with a number of benign and malignant tumors, particularly in the presence of suspicious clinicoradiologic findings. Careful paraffin sections, immunohistochemical studies and special stains should be performed to facilitate diagnosis in especially difficult cases and prevent unnecessarily overaggressive surgery.

References

1. Han Y, Zhang Q, Li SX, Feng L, Zhang L, Li Z, et al. Case Report Pleomorphic adenoma of the breast : a report of two cases and a literature review. Int J Clin Exp Pathol 2016;9(2):2459-2465.
2. Khamechian T, Alizargar J, Mazoochi T. Case Report Reporting a Rare Case of Pleomorphic Adenoma of

the Breast. Hindawi Publishing Corporation Case Reports in Medicine 2014;1–5.

3. Odunfa A, Bamiro A, Adekoya A.Case Report Pleomorphic adenoma of the breast in a young woman. JIAFP 2015;2(2)
4. Kelten C, Boyaci C, Trabulus DC, Sirin S. Case report Benign mixed tumour of the breast and breast skin, two cases with diagnostic difficulties.BMJ 2015;1–4. doi:10.1136/bcr-2015-210906
5. Radu I, Petcu I, Pănuţă A, Scripcariu D, Buna- M, Bilavschi K, et al. Pleomorphic adenoma of the breast. Arch Clin Cases 2016;3(4):144–8.
6. Pia-Foschini M, Reis-Filho JS, Eusebi V, Lakhani SR. Salivary gland-like tumours of the breast: surgical and molecular pathology. J Clin Pathol 2003;56:497–506.
7. Leekha N, Muralee M, Mathews A, Preethi T, et al. Pleomorphic Adenoma of Breast-A Case Report and Review of Literature. Indian J Surg Oncol (June 2014) 5(2):152–154.
8. Yogi V, Singh OP, Tiwari V. Malignant Chondroid Syringoma : A Case Report with Review of Literature . RRJMHS2014;3(2): 29-32.
9. Kundu R, Punia RS, Handa U, Dalal U. Chondroid syringoma : Cytomorphology of four cases and review of literature. Archives of Cytology and Histopathology Research 2016;1(2):63–7.
10. Balance W, Ro J, El-Naggar A, Grignon D, Ayala A, et al. Pleomorphic Adenoma (benign mixed tumor) of the Breast.An Immunohistochemical, Flow Cytometric, and Ultrastructural Study and Review of the Literature. Am J Clin Pathol 1990;93:795–801.
11. Sunumu O, Report C, Pleomorfik M, Nadir A, Olgu B. Pleomorphic Adenoma of the Breast : A Rare Case Report. Kafkas J Med Sci 2016;6(3):199–201.
12. Kusafuka K, Muramatsu K, Kasami M, Kuriki K, Hirobe K, Hayashi I, et al. Cartilaginous features in matrix-producing carcinoma of the breast : four cases report with histochemical and immunohistochemical analysis of matrix molecules. Modern Pathology 2008;1282–92.

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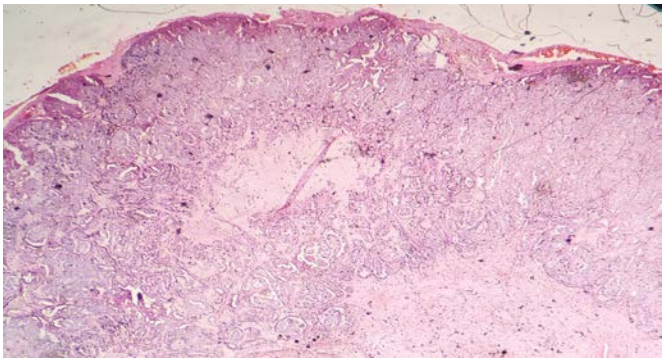

Figure 1: Microphotograph H & E stain 4x -Ttumor mass composed of epithelial tumor cells arranged in cords, nests, tubular and tubulobranching pattern.

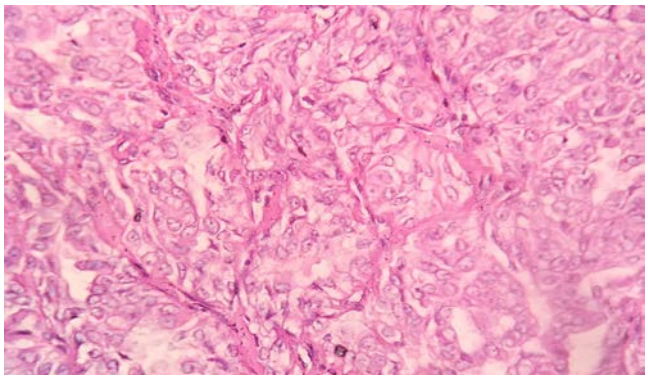

Figure 2: Microphotograph H& E 40x - The tumor cells were round to oval with round nuclei and scanty to moderate amount of cytoplasm. There was mild anisonucleosis.

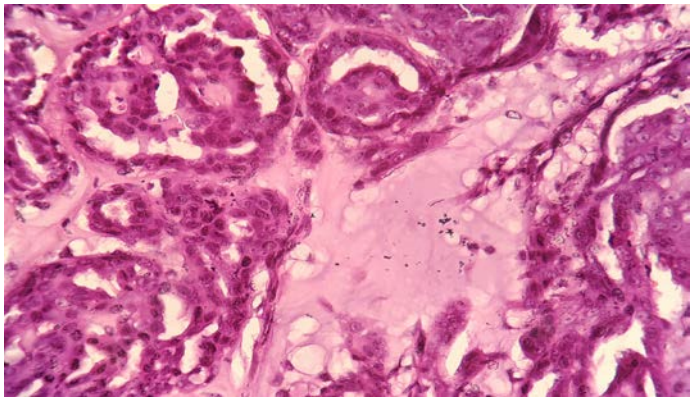

Figure 3: Microphotograph H & E stain 40x – Intervening myxoid stroma seen.