Mucoepidermoid Carcinoma of the Breast Presenting as Abscess-like Pus Discharging Mass; A Rare Tumor with Unusual Presentation

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INTRODUCTION

Mucoepidermoid carcinoma (MEC) is a common tumor arising from major salivary glands, although it has been reported in other sites as well including lacrimal gland, thyroid gland, thymus, bronchial tree, esophagus, ear, skin adnexa, mandible and pancreas. MEC occurring in the breast is very rare and accounts for only 0.2 to 0.3% of all the primary breast tumors. To date, only 44 cases of MEC cases of breast have been reported in the literature including one case from the Indian sub-continent. Herein, we report a case of primary MEC of the breast in a 73-years-old woman with unusual presentation as abscess like pus discharging mass with a review of the literature.

CASE PRESENTATION

A 73-years-old woman presented with the chief complaints of lump in the right breast for 7 months, associated with surface ulceration, pain and foul-smelling pus discharge for 2 months. On local examination, a hard lump with a solid cystic mass measuring 4x4cm was noted in the lower outer quadrant. The overlying skin showed ulceration containing foul-smelling pus discharge. Nipple discharge was also noted. No axillary lymphadenopathy was noted on the right side. The left breast and axilla were unremarkable. The clinical diagnosis made was carcinoma right breast cT4bN0M0.

Mammography of the right breast showed a large well-defined opacity measuring 44.9x44mm in the lower outer quadrant with BIRADs score 5, suggesting malignant etiology. The left breast was normal. Ultrasonography showed a large well-defined oval-shaped hyperechoic lesion showing vascularity with peripheral hypodense collection with echoes at
lower outer quadrant measuring 43.2x28.9mm in size, a likely neoplastic mass lesion. The patient was a known hypertensive on anti-hypertensive medications for 5 years. No family history of breast cancer was found.

In view of clinical presentation of abscess-like mass with ulceration and discharge, a decision to do incision and drainage along with biopsy was made by the surgeon. Later, modified radical mastectomy was done at a private hospital and pathology revealed a mucopidermoid carcinoma, high grade and cT4N0Mx stage. No further chemotherapy or radiotherapy was given. At present, the patient is doing well without any signs of recurrence or metastatic disease. Microscopically, the sections showed a tumor comprising nests, sheets and cords of epidermoid cells along with intermediate cells and multiple cystic spaces filled with abundant mucoid material lined by mucinous epithelium with peripheral basaloid cells (Figure 1).

Scattered mitotic figures, area of necrosis and foci of perineural invasion were noted. Focal cellular pleomorphism was seen in the epidermoid cells. PAS with diastase and Alcian blue stained the mucinous secretions in cystic spaces. No definite keratinisation or keratin pearl formation was seen. No lymphovascular emboli was found. Additionally, inflammatory cells comprising lymphocytes and plasma cells were also noted.

Immunohistochemistry (IHC) showed mammaglobin, and GCDFP15 (cytoplasmic), mCEA and EMA (membranous) positivity mainly in mucinous lining epithelial cells. GATA3 showed diffuse nuclear positivity throughout the tumor in all the cells (Figure 2). P63 highlighted the basaloid cells. High molecular weight cytokeratin (HMWCK), CK5/6, CK14 and CK8 highlighted the epidermoid cells (Figure 3). The Ki -67 index was approximately 20%. Hormonal markers ER, PR and HER2/Neu were negative. Based on histology and IHC, the final diagnosis was high grade mucopidermoid carcinoma.

**DISCUSSION**

The first case of primary MEC of the breast was reported in 1979 by Patchefsky et al. This condition is very rare and accounts for only 0.2 to 0.3 % of all the primary breast tumors. The true incidence may be higher as many cases may be misdiagnosed as metaplastic carcinoma with squamous differentiation due to shared histological features. At present, only 44 cases of MEC of the breast have been reported in the literature. It occurs in adult women with a wide age range varying from 29 to 80 years, usually presenting as a solid cystic mass involving any quadrant but retroareolar cases may cause nipple discharge. Unlike the index case, none of the reported cases presented in such locally advanced stage with ulceration and abscess-like pus discharging mass leading to the initial management with incision and drainage. The histological and immunohistochemical
features of MEC arising in the breast are similar to those of the salivary gland origin. This can be explained by the fact that both mammary and salivary glands are exocrine glands comprising tubules and acini with an ectodermal origin. Both have luminal epithelial cells surrounded by myoepithelial cells. These histological similarities account for the similar malignant lesion.1

Figure 2. Immunohistochemistry showing cytoplasmic (a) Mammaglobin, and (b) GCDFP15, (c) GATA3 (nuclear), (d) mCEA, and (e) EMA (membranous) mainly in mucinous lining epithelial cells, and (f) p63 (nuclear) in the basal cells (IHC x100).

Figure 3. Immunohistochemistry showing (a) High molecular weight cytokeratin (b) CK5/6, (c) CK14 and (d) CK8 positivity in epidermoid cells (IHC x100).

Microscopically, the tumor contains basaloid, mucinous, intermediate and epidermoid cells in varying proportions. The histopathological grade serves as an important predictor of outcome. The parameters usually considered for high grade MEC include cystic component (<20%), perineural invasion, necrosis, mitotic activity (≥4/10 hpf), and anaplasia.6 On the basis of semi-quantitative evaluation of these criteria, MEC can be further categorised into low, intermediate and high grade. Moreover, clinical features like rapid progression, locally advanced stage, lymph node positivity and large tumor size also favours a poor prognostic outcome. The index case fulfilled both histological and clinical criteria of the high grade MEC.

Intraductal papilloma, and adenomyoepithelioma form important differentials for a low-grade MEC. The immunohistochemistry of breast adenomyoepithelioma is unusual, where HMWCK shows a unique paradoxical staining pattern with diffusely positive inner epithelial cells and completely negative outer myoepithelial cells.7 In
MEC, HMWCK stain basaloid and epidermoid cells uniformly. Clear cell hidradenoma is also reported to have some similar histological features and may pose a challenge to differentiating it from low grade MEC. However, identification of SOX10-positive cuboidal cell lined ductal or tubular structures and hyalinized stroma with regular circumscribed borders in clear cell hidradenoma help in distinguishing from MEC. Absence of overt keratinization and squamous pearls is very important to differentiate high grade MEC from metaplastic breast carcinoma with squamous differentiation. Diffuse GATA3 expression can be seen in mammary analogue secretory carcinoma and salivary duct carcinoma; however, they usually do not show the expressions of p63, CK5/6 and HMWCK which is helpful to differentiate them from MEC.

Immunohistochemically, these tumors are ER, PR and HER2/neu negative (triple negative breast cancer, TNBC). Grading is the most important pathological prognostic factor. Both low and intermediate grade MEC behaves indolently and shows much better survival in comparison to other TNBCs. However, high grade MEC has a poor prognosis usually due to metastatic disease. Therefore, correct diagnosis of MEC in breast is crucial, as most of these tumors are amenable by breast conservative surgery and may not require neoadjuvant or adjuvant chemotherapy compared to other TNBCs. A few studies have shown that similar to MEC of salivary glands which shows frequent translocation t(11;19)(q21;p13) translocation resulting in the fusion of CRTC1 with MAML2, breast MEC harbours the same molecular alteration. These molecular analyses further support shared pathogenesis between salivary gland and breast MECs. We have not performed molecular analysis due to the limitations of the resources.

CONCLUSION
Mucoepidermoid carcinoma of breast is a very rare tumor under TNBC group with a very good overall prognosis. Histopathological features with appropriate IHC are the mainstay for correct diagnosis. Low and intermediate grade MEC behaves indolently, can be managed by breast conservative surgery and may not require neoadjuvant or adjuvant chemotherapy. Hence, identifying MEC is very important to warrant optimal treatment and avoid unnecessary extensive surgery or chemotherapy.

ETHICAL CONSIDERATIONS
Informed consent from the patient was obtained.

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CONFLICT OF INTERESTS
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REFERENCES


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