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Case study

Basal cell adenocarcinoma arising in salivary gland metaplasia of the breast: a novel salivary gland–type tumor developing in the breast

Christopher Flynn MD, PhD[a], Maryam Kadivar MD[b], Meeae Y. Kwon MD[c], Mariella Bonzanini MD[d], G. Kenneth Haines III MD[a], Fattaneh A. Tavassoli MD[a,*]

[a]*Department of Pathology, Yale University School of Medicine, New Haven, CT 06520, USA*
[b]*Department of Pathology, Rasool Akram Medical Complex, Tehran University of Medical Sciences, Tehran, Iran*
[c]*Community Memorial Hospital PTH, Ventura, CA 93003, USA*
[d]*Department of Surgical Pathology, S Chiara Hospital, Pisa 38100, Italy*

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Summary A variety of salivary gland–type lesions occur in the breast. Three cases of a novel mammary carcinoma arising in a background of salivary gland metaplasia and morphologically similar to basal cell adenocarcinoma of the salivary gland are presented. The clinical presentation, morphologic features, treatment, and follow-up of these cases are discussed.

1. Introduction

Salivary gland–type tumors of the breast are rare [1]. In a recent review of uncommon cases of breast neoplasms, less than 100 cases of salivary gland–type tumors primary to the breast were identified [1]. Most (n = 54) of these cases were pleomorphic adenomas [1]. Also described in the review were adenoid cystic carcinoma (ACCA), benign and malignant myoepitheliomas, syringomatous carcinoma, adenomyoepithelioma, acinic cell carcinoma, oncocytic carcinoma, and mucoepidermoid carcinoma [1]. However, not a single case of basal cell adenocarcinoma, the low-grade malignant counterpart of basal cell adenoma [2,3], has been reported in the breast in any of the prior reviews on the topic [1,4,5]. Basal cell adenocarcinoma of the salivary gland was first identified as a distinct entity in 1990 and constitutes only 1% to 2% of salivary gland cancers [3,6]. In this report, we describe 3 examples of basal cell adenocarcinoma of the breast arising in a background of salivary gland metaplasia in the breast.

2. Presentation of cases

The 3 cases presented during a 1 1/2-year period among the consultations of one of the authors (F.A.T.) and are summarized in Table 1. The patients, all women, were 43, 55, and 55 years of age. Two lesions presented in the left breast and 1 in the right. Two were subareolar in location, one associated with nipple retraction, the other with cystic dilation of the adjacent ducts. The third was central in location as well, but deeply and inferiorly located. They were all mammographically visible as irregular mass densities

* Corresponding author. Fattaneh Tavassoli, Department of Pathology, Yale University School of Medicine, 310 Cedar St, LH222, New Haven, CT 06520-8023, USA.
E-mail address: Fattaneh.Tavassoli@yale.edu (F. A. Tavassoli).

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(Fig. 1). There was no clinical evidence of axillary node involvement or metastases to distant sites. After a biopsy, 2 women had mastectomy, whereas 1 had lumpectomy and radiation therapy. In one case, the sentinel node was negative and the mastectomy contained a low axillary node that was also negative. In a second case, 15 axillary nodes were removed, all of which were negative. The third case had no nodes excised because none were clinically palpable. None of the patients has had either local recurrence or distant metastases at the latest follow-up 15, 23, and 25 months postdiagnosis.

Grossly, the lesions were irregular to stellate masses, 1.6, 2.5, and 3.0 cm in maximum diameter. Histologically, the 3 lesions showed substantial similarity. At low magnification, the striking feature was an admixture of solid nests of basaloid neoplastic cells with salivary gland–type acini as well as intercalated-, striated-, and excretory-type ducts (Figs. 2A and 3). The acini were lined by cells with eosinophilic to basophilic cytoplasmic granules concentrated along the luminal aspect (Fig. 2B and C). In every case, a transition was evident from ducts with distinct and often prominent basal cells (the term *basal cell* is used as implicated in the salivary glands and distinct from myoepithelial cells) to ducts with proliferation of the basal cell component either in a pure form or variably admixed with residual luminal cells (Fig. 3). The luminal cells with more abundant eosinophilic cytoplasm also showed variable degrees of proliferation in some of the metaplastic salivary gland–type tissues surrounding the adenocarcinoma. A complete or partial layer of basal cells persisted around some nests. The nests of neoplastic basal cells, which originated in the ducts and were variably scattered in the area of salivary gland metaplasia, gradually coalesced to form larger irregular nests and expansile masses with variable necrosis. Normal breast tissue with terminal duct-lobular units was evident around the lesion. A few salivary gland-type acini were entrapped by the carcinoma (Fig. 4A). Numerous mitotic figures and necrosis were observed within the proliferating cells in both smaller and larger nests but were more common in the latter (Fig. 4B-D).

The immunohistochemical profile of all 3 cases was very similar as outlined in Table 2 and represented in Figs. 5 and 6. The antibodies used and their sources and dilutions are shown in Table 3. Briefly, the neoplastic cells were negative for estrogen receptor (ER), progesterone receptor, androgen receptor (AR), CD10 (Fig. 6C), calponin, lysozyme, α_1-antitrypsin, and α_1-antichymotrypsin. The neoplastic basal cells were positive for S100 (predominantly in the peripheral cells, a reverse of the cytokeratin (CK) 903 staining; Fig. 5), CK903 (with sparing of the peripheral cells; Fig. 6A), CK5/6, and CD117 (c-kit). More than 40% of the nuclei in the nests of tumor cells were positive for Ki-67. Immunostain for p63 decorated only the peripheral basal cells in some of the smaller nests, whereas in other areas, up to two-thirds of the proliferating cells were p63 positive in irregular patches (Fig. 6B). In one case, some of the smaller nests of proliferating cells in the surrounding salivary gland–type areas were positive for

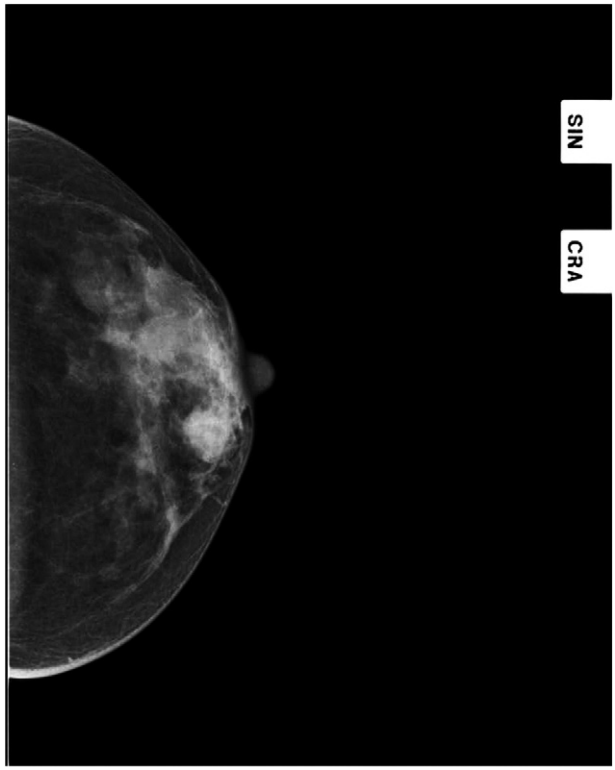

Fig. 1 A dense stellate mass is identified on mammogram suspicious for an invasive carcinoma.

Table 1 Patient information

Case	Age (y)	Tumor size (cm)/shape	Salivary gland metaplasia	Ductal intraepithelial neoplasia	Invasive carcinoma	Lymph node status	Location	Treatment	Follow-up
1	55	2.5, stellate	Present	Present	Present	None excised	Right, subareolar with nipple retraction	Mastectomy	Free of disease, 23 mo
2	55	1.6, lobulated	Present	Present	Present	Negative (0/15)	Central, inferior, deep	Lumpectomy, axillary dissection, and radiation	Free of disease, 25 mo
3	43	3.0, irregular	Present	Present	Present	Negative (0/2)	Left, retroareolar	Mastectomy with sentinel node	Free of disease, 15 mo