Case Report

Secretory Carcinoma of the Breast in a 20-year-old Male: Case Report and Review of Literature

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Abstract

Secretory breast carcinoma (SBC) is a very rare low-grade subtype of breast carcinoma. It was initially termed "juvenile breast cancer," but it is now known to occur in adults of both sexes, and only few male cases have been reported in the literature. SBC is of importance because of its unique morphology and excellent prognosis. We report here a case in a 20-year-old male presenting with right pedunculated and ulcerated breast mass with associated contact bleeding. Histopathology was performed and it revealed a secretory carcinoma. This tumor is morphologically characterized by the presence of abundant eosinophilic secretions in intracellular vacuoles and intercellular spaces. The objective of this article is to review the epidemiological and clinical aspect of secretory carcinoma from a case report and literature review.

Keywords: *Breast, male, secretory carcinoma*

Introduction

Carcinoma of the breast is among the most common human cancers throughout the world, and male breast carcinoma is a relatively rare malignant tumor accounting for <1% of all breast cancers.[1] Secretory breast carcinoma (SBC) is a very rare subtype of breast carcinoma and comprises <0.15% of invasive breast cancers. Most of the patients are females, and it is very rare in males with a relatively good prognosis.[2] The typical clinical presentation is a slow-growing, painless, well-circumscribed, mobile, palpable mass. SBC is morphologically characterized by the presence of abundant eosinophilic secretions in intracellular vacuoles and intercellular spaces. The secretory material in cells, lumen, and stroma is mucicarmine, Alcian blue, and periodic acid–Schiff (PAS) positive. Mucinous carcinoma, acinic cell carcinoma, and apocrine carcinoma are primary malignant breast lesions that may resemble secretory carcinoma morphologically. Recently, the tumor was found to be associated with a distinct ETV6-NTRK3 mutation which confers the tumor proliferative and survival advantage. As a rare type of breast carcinoma, there are at present no consensus guidelines for treatment. Although recommendations vary among authors, surgical intervention is the primary mode of treatment for secretory carcinoma.[3] A review of literature revealed few cases of secretory carcinoma of the breast in males, and this is the first case seen and reported from our center.

Case Report

A 20-year-old male student presented with a lump on the right breast. The lump was first noticed 2 years ago as a small pea-sized nodule which grew initially slowly but became rapidly increasing in size over the last 3 months before presentation. It was painless and ulcerated 1 month before presentation with associated history of contact bleeding. He has no family history of breast cancer. The physical examination revealed a 4 cm × 3 cm ulcerated tumor mass located in the subareolar region of the right breast. The mass was mobile and not painful. No skin dimple, no nipple retraction, or nipple discharge was observed. No clinical axillary lymph node involvement was detected. Routine blood tests including urea, electrolytes, and full blood counts were within normal limits. The liver ultrasound and chest X-ray were

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negative for metastases. The mass was excised and sent for histological analysis.

Grossly, the mass was fairly circumscribed partly skin covered with some ulcerated areas. The mass measured 4 cm × 3 cm and had grayish white-to-tan solid surfaces on cut section. On microscopic examination, hematoxylin and eosin stain revealed abundant eosinophilic secretions in intracellular vacuoles and intercellular spaces [Figure 1a and b]. The secretion was mucicarmine and Alcian-blue positive. No tumor infiltration was present at the nipple or at surgical margins.

On immunohistochemistry, the tumor cells were positive for epithelial membrane antigen (EMA) and S100 [Figure 2a and b] but negative for human epidermal growth factor 2 (HER2) and estrogen receptor.

Postoperatively, the patient was fairly stable and was not regular at his clinic attendance. He was seen for only 2 months and later lost to follow-up.

Discussion

Male breast cancer constitutes 6.1% of all breast cancer cases in our center, which is more than the 1% found in developed countries.[4] In general, high incidence rate was also documented in other parts of Nigeria as well as other African countries.[5-6]

Secretory carcinoma is a very rare subtype of breast carcinoma, most commonly seen in females and its occurrence in male is even rarer with male-to-female ratio of 1:6.[7-9] Lamovec and Bracko reported four cases of SBC in their retrospective series of 7038 breast carcinoma cases,[10] and Botta *et al*.[11] found one case of SBC among 3000 breast carcinoma cases. Li *et al*. reported 15 cases in their pathologic review of 10,000 breast carcinoma cases.[12] To the best of our knowledge, no case of secretory carcinoma of the male breast has been reported in Nigeria.

Secretory carcinoma has been known previously as juvenile breast carcinoma. The age at presentation varies from 3 to 66 years with a mean and median age of presentation of 33 and 40 years, respectively.[13] In a recent study of 15 male patients with breast cancer by de Bree *et al*., the median age was 17 years. Secretory carcinoma seems to occur at a younger age in males than females and reports suggest that the disease tends to be more aggressive in males.[14] Our case was a 20-year-old male which is in agreement with the average age reported for SBC in males, though a case was seen in a 66-year-old male as reported by Kuwabara.[15]

The patients commonly present with a slow-growing, painless, well-circumscribed, mobile, palpable, subareolar mass in the breast which was similar with the clinical presentation of our patient. The tumor size varies from 1 to 16 cm with an average diameter of 3 cm. The literature mentions nodal involvement in 15% of patients at presentation.[16] Although rare, some patients have been reported to present with metastatic disease.[17]

Even though the descriptive mammographic findings are sparse in the literature, it was not requested for when the patient presented. Many case reviews reported that the mammographic findings of SBC are variable and nonspecific, ranging from no abnormal findings or benign-looking nodular density to suspicious malignant lesion.[18]

Fine-needle aspiration cytology which was not done on this patient usually shows the presence of prominent intracytoplasmic vacuolization.[19] Vesoulis and Kashkari reported that cytological features of SBC resemble that of benign epithelial proliferative lesions, particularly lactating adenoma.[20]

Secretory carcinoma can demonstrate several histological patterns, including solid, microcystic, and ductal, with many tumors containing all the three patterns.[21] The tumor cells are polygonal with granular eosinophilic cytoplasm, with intracellular and extracellular PAS- and Alcian-blue-positive secretions.[21] Atypia is minimal or absent and mitotic activity is low.[22] In the current case on immunohistochemical studies, paraffin embedded tissue using heat method of antigen retrieval method was used. The neoplastic cells were positive for EMA and S100 but negative for estrogen and HER2 receptors. The immunohistochemical findings are similar with cases reported in other studies.[23] The final histologic diagnosis was a SBC.

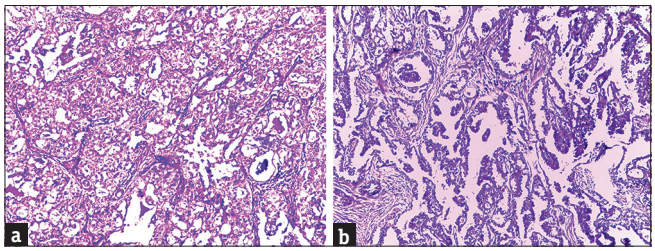

Figure 1: (a) Secretory carcinoma showing eosinophilic secretions in intracellular vacuoles and intercellular spaces (H and E, ×100). (b) Secretory carcinoma (H and E, ×100)

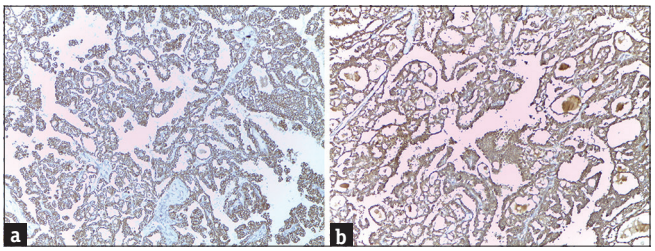

Figure 2: Immunohistochemical stains with the tumor cells demonstrating (a) positive cytoplasmic staining by epithelial membrane antigen. (b) Positive nuclear and cytoplasmic staining by S100 antibody

Surgery is considered the mainstay of treatment of secretory carcinoma; however, due to scarcity of reported cases, no published guidelines for surgical management exist. The demonstration of late local recurrence has led many to propose mastectomy for the patients with this disease.[24] Axillary lymph node dissection is considered by some authors for tumors >2 cm.

Adjuvant chemotherapy and radiation have been tried for the disease without much success. Therefore, there is at present insufficient evidence to recommend either approach in the management of secretory carcinoma.

Distance metastases are rare and have been reported in few cases. Local recurrence may occur after longtime postexcision, and therefore, longtime follow-up is really advisable. The good prognostic factors include tumor size <2 cm, age <20 years at the time of diagnosis, and tumors with circumscribed margins.[25]

Conclusion

Secretory carcinoma of the breast is a rare, slow-growing tumor and can metastasize to the axillary lymph node. Surgery in the form of mastectomy with axillary clearance is the treatment of choice. The role of adjuvant chemotherapy and radiotherapy also remains poorly defined and targeted therapies may be the treatment of choice in the future.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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