Case report

Intracystic Papillary Carcinoma in a Male Breast: Thirty Years after Orchiectomy

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SUMMARY

Papillary carcinoma of the breast is an extremely rare form of breast carcinoma in males. We report a case of an 83-year-old man who presented with a retroareolar palpable mass, associated with bloody nipple discharge. Cytological characteristics were rare clusters and papillary formations with enlarged hyperchromatic nuclei, surrounded by erythrocytes. Having in mind that a reliable cytological diagnosis of papillary carcinoma cannot be made and that all papillary lesions observed in cytological material should be excised for histological examination, breast conserving surgery (BCS) was performed.

Histologically, cystic ducts were lined with atypical papillary formations bridging the duct lumen, but with the absence of both myoepithelial cells and stromal invasion. Prolactin and testosterone serum levels were decreased. Immunohistochemical examination on actin, estrogen and progesterone receptors was negative.

The authors have pointed out that the patient had left orchiectomy, induced by tuberculous orchiepididymitis with scrotal fistula, performed thirty years before. Twelve months after breast conserving surgery, the patient is still alive.

Key words: male breast papillary carcinoma, cytology, pathohistology, immunohistochemistry, tuberculous orchiepididymitis

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INTRODUCTION

Papillary carcinoma of the breast is an extremely rare form of breast carcinoma in males, with a handful of case reports in the literature entity (1-4). In contrast to the breast carcinoma in women (4), it tends to affect older age group having an excellent prognosis due to low malignant potential (1, 5, 6).

The goal of the current study was to expand the knowledge regarding the cytological, histological and immunohistochemical characteristics, but also to suggest the possible pathogenesis of the male intracystic papillary breast carcinoma.

CASE REPORT

A 83-year-old male presented with bloody nipple discharge in the left breast occurring in the previous three months. Clinically, there was a palpable mobile retroareolar plaque like induration. The nipple and the overlying skin were normal. The right breast and the nodes in the axillary and supraclavicular fossa were not palpable. On ultrasonography, the mass was heterogeneous with solid and cystic components, measuring 15 x 12 mm. His past medical history included a diagnosis of pulmonary tuberculosis at the age of 18. Due to tuberculosis infection of testis and formed scrotal fistula, orhiectomy was performed 30 years before. His familial

history was not significant. During the last 30 years of his life, he was in good health.

Cytological smears of the bloody nipple discharge were stained by HE (Haematoxylin and Eosin), PAP (Papanicolaou) and MGG (May Grünvald Gimsa) methods and analyzed three times, to the time of the breast conserving surgery (BCS). Paraffin sections were stained with HE, histochemical Van Gieson and immunohistochemical ABC methods, using the antibodies to actin, estrogen and progesterone receptors (Er and Pr). Cytologically, the atypical cells formed rare large cohesive clusters surrounded by erythrocytes and papillary formations with increased cellularity, irregular nuclear contour and hyperchromasia (Figure 1), suggesting the diagnosis of "an atypical papillary adenoma".

Histological examination of specimens showed cystic dilatation of the ducts lined with papillary lesions, having the characteristics of non-invasive papillary carcinoma: round to polyhedral cells with mild nuclear atypia and hyperchromasia, but with rare mitoses. They were without the vascular connective tissue core, arranged in solid and micropapillary formations and forming cribriform pattern (Figure 2). Excision margins were free of tumor.

Immunohistochemical examination with antibodies to actin, estrogen and progesterone was negative. Serum prolactin (5.2 ng/ml) and testosterone (1.22 ng/ml) were at the normal level (Eliza).

Figure 1. Cytological features of the male breast tumor: cluster of dysplastic cells, surrounded by erythrocytes. MGG x 300

Figure 2. Cribriform pattern of intracystic papillary carcinoma. HE x 300

DISCUSSION AND CONCLUSION

Intracystic papillary carcinoma is a term used to describe a solitary, centrally located, malignant papillary proliferation involving a cystically dilated ducts (7-9). It is an extremely rare condition in men and accounts for 0.6 % of all breast cancers (1, 3-5), with a favorable prognosis due to low malignant potential. Mean age of patients with male breast papillary carcinoma is 68.2 years (2, 4). Our patient was 83 years of age, being one of the oldest patients in this category (4, 8). Clinical presentation, mammographic appearance and ultrasonography are not specific (1, 6, 7). Bloody nipple discharge, the clue clinical symptom for duct papillary carcinoma, directed us to repeat the cytological examination three times during one week and enabled us to discover an "atypical intracystic papilloma". Differential histological diagnosis between an atypical intracystic papilloma and intracystic papillary carcinoma was based on both histological and immunohistochemical examinations of the surgical biopsies (1, 9-11).

From our results, we have excluded the possible role of prolactin, estrogen and progesterone in the pathogenesis of breast carcinoma (11, 12).

Our patient had unilateral orchiectomy thirty years before, caused by tuberculous infection. The decreased serum testosterone level in our patient not only confirms that it has an important role in the genesis of the male breast carcinoma but also explains the fact why the main victims of male breast cancer are elderly males.

The association of cancer and tuberculosis is well known (13). Mycobacterium tuberculosis-infected macrophages produce various cytokines and the most important is epiregulin, a potent epithelial growth factor for premalignant epithelial cells, indicating that chronic inflammation caused by tuberculosis may stimulate carcinogenesis (14, 15). Medical history of our patient was positive for tuberculosis which may contribute, with addition of other factors, to the development of breast cancer. The linkages between breast cancer and epiregulin is currently weak, but further investigation needs to be done.

Wide local surgical excision, without axillary dissection, is currently the treatment of choice. No other treatment was performed. He was doing well at the 12-month follow-up.

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Intracistični papilarni karcinom u dojci muškarca: trideset godina nakon orhiektomije

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SAŽETAK

Papilarni karcinom je izuzetno redak karcinom dojke kod muškaraca. Prikazuje se bolesnik star 83 godine sa retroareolarnom palpabilnom masom udruženom sa krvavom sekrecijom iz dojke.

Na citološkom preparatu nađeno je prisustvo retkih klastera tumorskih ćelija i papilarnih formacija sa uvećanim hiperhromatskim jedrom okruženim eritrocitima. Imajući u vidu da pouzdana dijagnoza papilarnog karcinoma nije moguća na osnovu citološkog nalaza i da papilarne formacije nađene u citološkom materijalu zahtevaju biopsiju promene, izvršena je poštena hirurška operacija dojke.

Na patohistološkom materijalu nađen je cistično prošireni duktus obložen atipičnim papilarnim formacijama, koje premošćuju njegov lumen, odsustvo mioepitelnih ćelija i stromalne invazije.

Serumske vrednosti prolaktina i testosterona bile su snižene. Imunohistohemijsko bojenje na aktin, estrogenske i progesteronske receptore bilo je negativno.

Autori naglašavaju da je bolesnik imao levu orhiektomiju izvršenu zbog tuberkuloznog epididimoorhitisisa sa skrotalnom fistulom pre 30 godina. Dvanaest meseci nakon poštene operacije dojke, bolesnik je zdrav.

Ključnereči: papilarni karcinom kod muškarca, citologija, patohistologija, imunohistohemija, tuberkulozni orhiepididimitis