Online Case Report

Papillary carcinoma in a male breast cyst: a diagnostic challenge

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Intracystic papillary breast carcinoma is a rare form of non-invasive carcinoma with an excellent prognosis. It accounts for less than 0.5% of breast cancers. We report the case of a 75-year-old man presenting with a painless cystic lump in the right breast. Ultrasonography showed a cystic lesion and aspiration revealed blood-stained fluid with suspicion of malignancy. Excisional biopsy was necessary to confirm the diagnosis and also indicated that local treatment was adequate.

Keywords: Intracystic – Papillary carcinoma – Male breast cancer

Male breast cancer is a rare disease and the incidence is 1% of all breast cancer. Intracystic papillary breast carcinoma is an extremely rare condition in men and represents only 5–7.5% of all male breast carcinomas.¹ Diagnosis is often delayed because of the benign appearance on physical examination and radiological imaging. Such carcinomas have a better prognosis than other types of breast malignancy. Fine needle aspiration is often inaccurate and excision biopsy may be necessary for the diagnosis.² We report a case of 75-year-old man presenting with a cystic lesion of the right breast which recurred after aspiration. An excisional biopsy confirmed the diagnosis and also indicated that local treatment was adequate.

Case report

A 75-year-old male presented with a painless mass in the right breast of 1-month duration. On examination, he had a well-defined 2 cm x 2 cm, firm, non-tender, non-adhering lump lateral to the right nipple. The left breast and both axillae were normal. Ultrasound showed a cyst. Aspiration of the lump revealed blood-stained fluid.

Figure 1 Macroscopic examination shows a well-defined cystic lesion with a fleshy papillary lesion in its wall.

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Figure 2 Histology shows a cyst containing a papillary and glandular lesion on its wall with extensive microcalcification. The tumour consists of round to polyhedral epithelial cells. Nuclear atypia is mild and mitoses are rare. Since no evidence of stromal invasion was found, the lesion was diagnosed as intracystic papillary carcinoma, low grade.

Microscopic examination revealed foamy macrophages in addition to numerous epithelial cells arranged as three-dimensional papillary clumps with nuclear crowding. There was also variation of nuclear size and prominent nucleoli. The appearance was suspicious of malignancy, with the possibility of a papillary lesion being raised.

The cyst recurred 2 weeks later and the patient subsequently underwent a lumpectomy. Macroscopic examination of the specimen showed a cyst of 30 mm diameter with an intracystic fleshy papillary lesion in its wall (Fig. 1). Multiple sections were taken for microscopic examination. The cyst wall was lined by a single layer of epithelial cells without atypia. The papillary lesion was a non-invasive papillary carcinoma. The carcinoma cells were round to polyhedral with mild atypia and rare mitoses. There was no evidence of stromal invasion. The tumour was diagnosed as intracystic papillary carcinoma, low grade (Fig. 2). Excision margins were free of tumour. No other treatment was performed. He was doing well at 1-year follow-up.

An electronic literature search was undertaken using the key words: intracystic papillary carcinoma, male breast cancer on the Medline, Pubmed, Google and Embase databases from 1966 to date and the bibliographies of extracted articles examined.

Discussion

Intracystic breast carcinoma is rare in females and exceedingly rare in males with a handful of case reports in

the literature.¹⁻³ It accounts for 5–7.5% of all breast cancers in males.¹ It is a localised, non-invasive breast cancer with papillary proliferation arising within or on the wall of a large cyst.²

It commonly presents as a benign-appearing, well-localised lump due to its underlying cystic nature. In a case report and subsequent review of Japanese cases in men, Tochika *et al.*³ reported the mean age of intracystic carcinoma in males as 68.2 years. Most of the patients presented with a palpable lump. In addition to abnormalities felt on palpation, a few patients presented with mild pain, bloody nipple discharge and pruritus.³

Radiological studies are helpful. Ultrasonography of these lesions typically reveals a hypo-echoic area (representing the cyst) with soft tissue echoes projecting from wall of the cyst (intracystic tumour).⁴ Biopsy should be aggressively performed under a tentative diagnosis of intracystic breast carcinoma when ultrasound reveals cyst with mural nodules and aspiration fluid is bloody.³ Fine needle aspiration is rarely diagnostic and an excisional biopsy is usually required to establish the diagnosis.² Intracystic papillary carcinoma tends to be well-defined on mammography; an irregular margin suggests the presence of invasion.⁵

Tsuda *et al.*⁶ reported that loss of heterozygosity (LOH) on chromosome 16q was a useful marker for the intracystic papillary carcinoma, since intraductal papilloma showed no LOH. Using the polymerase chain reaction, the malignant potential of intracystic papillary lesions may be more clearly determined.⁶ There is

currently no consensus regarding the management of intracystic papillary carcinoma, with the potential for over-treatment. A study by Harris *et al.*⁷ reviewed patients who underwent varying combinations of local excision, radiotherapy and tamoxifen. The absence of axillary involvement and low recurrence rate after local excision suggests that wide local excision without axillary dissection is currently the treatment of choice for pure IPC.⁷ The role of sentinel lymph node biopsy has not been evaluated in this disease but it may be an excellent alternative to full axillary dissection in patients with IPC associated with invasive carcinoma. The role of radiation therapy in these patients remains undefined. IPC associated with DCIS or invasive cancer or both and should be treated on the basis of this associated pathology. Prognosis of pure IPC is usually excellent because the malignant potential and the proliferative activity of the cancer is low.¹

Conclusions

Intracystic papillary breast carcinoma is extremely rare in males. Clinically it presents as a benign-looking cystic lesion. Mammographic appearance is typically of benign, well-circumscribed, mass lesion. The diagnosis of an intracystic malignancy should be suspected when

ultrasonography shows intracystic mural nodule, the mass persists or recurs after fine needle aspiration or cellular atypia is identified in the aspirate specimen. Wide, local excision without axillary dissection is currently the treatment of choice and the prognosis is favourable.

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