Intracystic Papillary Carcinoma of the Breast in Male: Case Report and Review of the Japanese Literature

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We report a case of intracystic papillary carcinoma of the breast in a 62-year-old Japanese man, who came to our hospital complaining of a right subareolar mass. Imaging diagnosis was a cyst with an intracystic component. Since repeated aspiration biopsy cytology was interpreted as a borderline lesion, no additional treatment had been given. Because he wanted the removal of the mass 14 months after his first visit, an excisional biopsy was performed under local anesthesia. Pathological examination revealed the intracystic component to be non-invasive papillary carcinoma. Although the incidence of male breast cancer is ~1% of all breast cancer, intracystic papillary carcinoma in the male is very rare. From a review of the literature on this disease in Japanese men, an intracystic component can often be demonstrated by ultrasound. However, it is difficult to confirm malignant cells by aspiration biopsy cytology of cyst fluid. Since some cases have a local relapse, excisional biopsy and long-term follow-up are indicated.

Key words: intracystic papillary carcinoma – male breast cancer

INTRODUCTION

Male breast cancer is a rare disease and the incidence is ~1% of all breast cancer (1). We experienced a case of intracystic papillary carcinoma of the breast in a male, whose diagnosis had been finally given after excisional biopsy. We discuss the clinicopathological features of cystic breast cancer in the male from a review of the Japanese literature and the present case.

CASE REPORT

A 62-year-old man, complaining of a round mass in his right subareolar region, visited the Division of Breast Surgery at the National Cancer Center Hospital East in August 1995. He had a past history of asthma, but did not use medication. He had no gynecomastia. The tumor was 3 × 2.7 cm in size with a smooth surface and clear margin. Bilateral axillary lymph nodes were not palpable. Mammogram showed a cyst, and ultrasound revealed a cystic lesion with an intracystic component [Fig. 1(a) and (b)]. The initial aspirate contained clusters of columnar cells with enlarged nuclei and a moderate nucleus/cytoplasm (N/C) ratio (borderline malignancy) (Fig. 2). Aspiration biopsy cytology had been performed three times during the following one-year period. Since cytology had diagnosed a borderline lesion, the patient had been followed every six months. Because he wanted the removal of the mass, he underwent an excisional biopsy with local anesthesia in November 1996. The resected specimen was a cyst with an intracystic papillary lesion (Fig. 3). Grossly, the size of the lesion was 3.2 × 2.5 cm. The small protruded lesion was 8 × 5 × 7 mm in size. 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 [Fig. 4(a) and (b)]. The carcinoma cells were round to polyhedral with mild atypia and rare mitoses. There was no evidence of stromal invasion. The tumor was diagnosed as intracystic papillary carcinoma, low grade. Upon review, the initial aspirates revealed clusters of mildly atypical cells with slight nuclear hyperchromasia and small nucleoli, but the cytological diagnosis was not suggestive of malignancy. Systemic survey (chest X-ray, liver ultrasound and bone scintigram) showed no evidence of distant metastases. Serum tumor markers [carcinoembryonic antigen (CEA) and carbohydrate antigen 15-3 (CA15-3)] were within normal limits. Since non-invasive carcinoma in this case was completely resected, no additional surgical treatment was performed. He was free of disease about one year after the excision.

DISCUSSION

Intracystic papillary carcinoma of the breast in the male is a very rare disease. The clinical features extracted from the Japanese literature of cystic breast cancer in males and the present case are

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Abbreviations: CEA, carcinoembryonic antigen; CA15-3, carbohydrate antigen 15-3; N/C ratio, nucleus/cytoplasm ratio; LOH, loss of heterozygosity
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Figure 1. Diagnostic imaging. A mammogram shows a cyst (a). An intracystic component is demonstrated by ultrasound (b).

Figure 2. Initial aspiration cytology. The specimen contains clusters of columnar cells with enlarged nuclei and a moderate N/C ratio (borderline malignancy).

Figure 3. Macroscopic features of the cyst. The inner surface of the cyst was smooth, associated with a protruding papillary lesion.

Figure 4. Microscopic features of the cystic lesion. The cyst wall (right lower corner) was lined by a single layer of flat epithelial cells, and a papillary epithelial lesion with a fibrous stalk is present in the wall (a). The tumor consists of round to polyhedral epithelial cells (b). 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.

The age of patients ranged between 46 and 91 years. Most patients presented with a palpable mass or lump. Bloody nipple discharge occurred in one case (5). Ultrasound often demonstrated intracystic papillary lesions, and was thus useful for clinical diagnosis. However, it was difficult to obtain a definite diagnosis of malignancy by aspiration cytology specimens. Seven Japanese male patients in Table 1 showed negative or borderline cytology. There was only one case exhibiting positive cytology in the world literature (12). The cytological diagnosis in the present case could not be suspicious for carcinoma. Excisional biopsy has been recommended in this condition.
Table 1. Review of the clinical features of cystic breast cancer in Japanese males

<table>
<thead>
<tr>
<th>First author and reported year</th>
<th>Age (year)</th>
<th>Ultrasound</th>
<th>Cytology class</th>
<th>Method of diagnosis</th>
<th>Treatment</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Non-invasive carcinoma</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1. Noguchi, 1983</td>
<td>80</td>
<td>Intracystic tumor</td>
<td>BPL</td>
<td>Biopsy</td>
<td>Simple M</td>
<td>3 y alive</td>
</tr>
<tr>
<td>2. Watanabe, 1986</td>
<td>46</td>
<td>Intracystic tumor</td>
<td>–</td>
<td>Biopsy</td>
<td>Modified RM</td>
<td>–</td>
</tr>
<tr>
<td>3. Sasahashi, 1992</td>
<td>64</td>
<td>Intracystic tumor</td>
<td>BPL</td>
<td>Biopsy</td>
<td>Standard RM</td>
<td>11 m alive</td>
</tr>
<tr>
<td>4. Kato, 1997</td>
<td>54</td>
<td>Intracystic tumor</td>
<td>BM</td>
<td>Biopsy</td>
<td>Modified RM</td>
<td>7 y alive</td>
</tr>
<tr>
<td>5. Present case</td>
<td>62</td>
<td>Intracystic tumor</td>
<td>BM</td>
<td>Biopsy</td>
<td>Biopsy only</td>
<td>1 y alive</td>
</tr>
</tbody>
</table>

| Invasive carcinoma            |            |            |                |                    |           |         |
| 6. Yabuki, 1982               | 54         | Bloody cyst | –              | Biopsy | Standard RM | 12 y dead |
| 7. Abe, 1985                  | 69         | Intracystic tumor | – | Biopsy | Modified RM | 2 y alive |
| 8. Kasumi, 1987               | 73         | –          | –              | –          | Modified RM | 9 y 10 m dead |
| 9. Kasumi, 1987               | 91         | –          | –              | –          | Simple M | 8 m alive |
| 11. Nishizawa, 1993           | 70         | Intracystic tumor | BPL | Biopsy | Modified RM | 1 y alive |
| 12. Ikeda, 1994               | 84         | Intracystic tumor | BPL | Biopsy | Standard RM | 2 y alive |

BM, borderline malignancy; BPL, benign papillary lesion; M, mastectomy; RM, radical mastectomy; y, year; m, month; –, not described.

Cutuli et al. (13) reported that there were seven papillary carcinomas and five papillary and cribriform carcinomas among 31 male cases of ductal carcinoma in situ of the breast. Six of the 31 cases underwent lumpectomy and 25 mastectomy. All cases had negative nodes. Relapse-free survival was 83% at 10 years, since four cases had a local relapse. In addition, Leffkowitz et al. (14) reported 77 female cases of invasive or non-invasive intracystic papillary carcinoma of the breast. These cases had been treated with excisional biopsy or mastectomy, and had a 10-year disease-free survival rate of 91%. In the Japanese literature reviewed, most patients had undergone modified or standard radical mastectomy, but long-term follow-up data are not available. However, case no. 4 in Table 1 developed a local relapse of non-invasive ductal carcinoma (5). These results suggest that the optimal treatment for intracystic papillary carcinoma is excisional biopsy in cases of non-invasive carcinoma, and simple or modified radical mastectomy in cases of invasive carcinoma. Anyway, long-term follow-up is necessary after surgical treatment.

The etiology remains unknown, but it is thought that intracystic papillary carcinoma arises in larger, more centrally placed ducts, and tumor development and its secretion cause the cystic dilatation (14). The differential diagnosis of intracystic papillary lesions should be made on histopathological specimens. Tsuda et al. (15) reported that loss of heterozygosity (LOH) on chromosome 16q was a useful marker for intracystic papillary carcinoma, since intraductal papilloma showed no LOH. Using this method by the polymerase chain reaction, the malignant potential of intracystic papillary lesions may be more clearly determined.

In conclusion, we report a rare case of intracystic papillary carcinoma of the breast in a male. An excisional biopsy and long-term follow-up are indicated in this disease.

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References

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