Breast Cancer
Vol. 11 No. 4 November 2004

Case Report

A Case of Intracystic Papillary Carcinoma with a Multilocular Cyst of the Breast in Male

Minoru Kihara, Natsumi Mori, Akira Yamauchi, and Hiroyasu Yokomise

Second Department of Surgery, School of Medicine, Kagawa University, 1750-1 Miki-cho, Kita-gun, Kagawa 761-0793 Japan.

Intracystic papillary carcinoma of the breast in males is a very rare disease but has an excellent prognosis. We report the case of a 68-year-old man who had a right subareolar soft mass. Imaging examinations showed a multilocular cyst with an intracystic component, and benign disease was diagnosed. Fine needle aspiration of the cyst fluid revealed many malignant cells, and modified radical mastectomy was performed under general anesthesia. Histological examination showed intracystic papillary carcinoma with a multilocular cyst. No positive lymph nodes were involved. The diagnosis of intracystic papillary carcinoma of male breast should be made carefully to avoid misdiagnosing benign disease.

Breast Cancer 11:409-412, 2004.

Key words: Intracystic papillary carcinoma, Male breast cancer, Multilocular cyst

Male breast cancer is a rare disease and the incidence is less than 1% of all breast cancers[1]. In addition, primary intracystic papillary carcinoma of the male breast is an extremely rare disease accounting for less than 1% of all male breast cancers[2-5]. Most cases of intracystic papillary carcinoma of the breast in men had a single cystic lesion with a malignant intracystic component in the literature[2-10]. We report the case of intracystic papillary carcinoma occurring in a multilocular cyst of the male breast.

Case Report

A 68-year-old man had first noticed a painless tumor in the subareolar of his right breast in March 2003. It had grown in size rapidly within a short time, so he was referred to our hospital after a month. Physical examination demonstrated a soft, well defined, movable tumor measuring 5.0 × 4.0 cm and retraction of the nipple, but without nipple discharge (Fig 1). Bilateral axillary lymph nodes were not palpable. His past medical history and family history revealed no evidence of malignancy. He had no additional gynecomastia. Mammography of the right breast showed a well-defined dense nodule (Fig 2). Ultrasonography revealed multiple cystic lesions, and a large cyst with a mural hyperechoic nodule (Fig 3). Computed tomography and magnetic resonance imaging (Fig 4) scans revealed a multilocular cyst.

Aspiration showed dark brown fluid, and cytology of this fluid revealed adenocarcinoma (Fig 5). Serum tumor marker (carcinoembryonic antigen, carbohydrate antigen 15-3, BCA225, and NCC-ST439) levels were all within the normal limits. The blood estradiol, estriol, follicle stimulating hormone, luteinizing hormone, testosterone, progesterone, and prolactin levels were normal. No evidence of other lesions was observed by bone scintigraphy or computed tomography of the chest or abdomen. Modified radical mastectomy was performed in May 2003. The resected specimen showed multiple cystic lesions measuring 2.3 × 1.5 cm and a solid papillary lesion measuring 2.6 × 1.5 cm, with necrotic change and some hemorrhagic areas. Histopathologically, the papillary lesion was an intracystic papillary carcinoma, and the inside of a part of the cyst wall was lined by multiple layers of malignant cells (Fig 6, 7). There was no evidence of malignancy in the other cysts. Therefore, intracystic papillary carcinoma of the breast was diagnosed. On immunostaining, the cancer cells were strongly positive for estrogen receptor (> 95% of cells positive) and proges-

Reprint requests to Minoru Kihara, Second Department of Surgery, School of Medicine, Kagawa University, 1750-1 Miki-cho, Kita-gun, Kagawa 761-0793 Japan.

Received December 10, 2003; accepted April 10, 2004

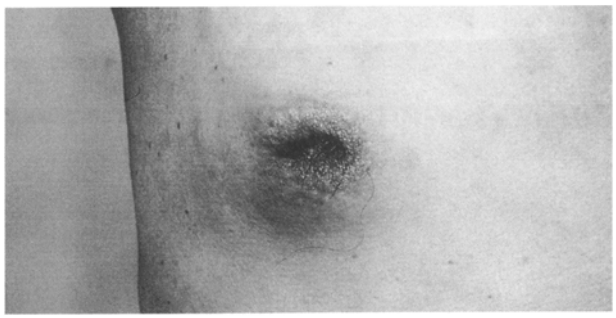

Fig 1. A mass, 5 cm in diameter, in the right subareolar area with retraction of the nipple.

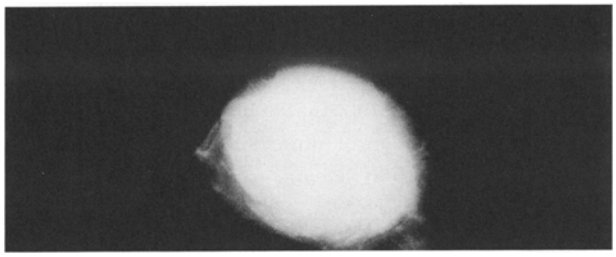

Fig 2. Mammography of the right breast showed a well-defined dense nodule without calcification or spiculation.

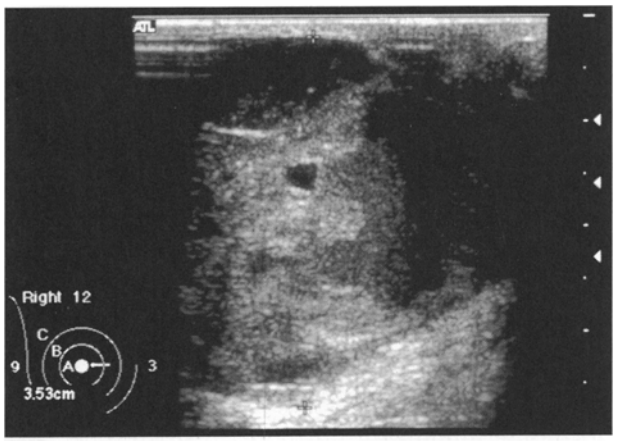

Fig 3. Ultrasonography revealed multiple cystic lesions, and a large cyst with a mural hyperechoic nodule.

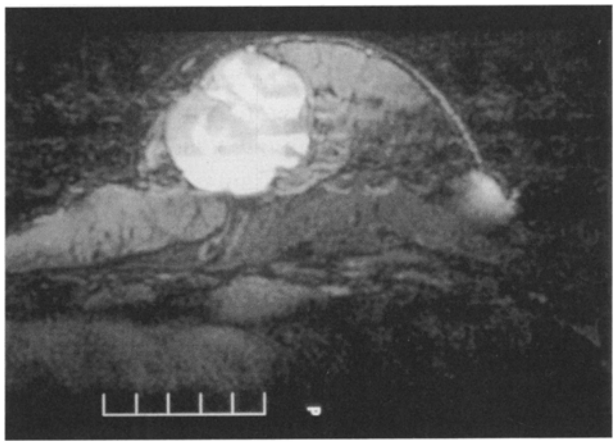

Fig 4. Magnetic resonance imaging scan revealed a multilocular cyst.

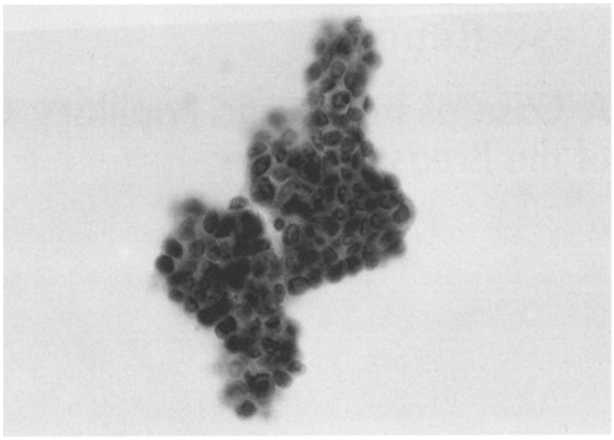

Fig 5. Cytology of cystic fluid revealed adenocarcinoma. Papanicolaou stain × 100.

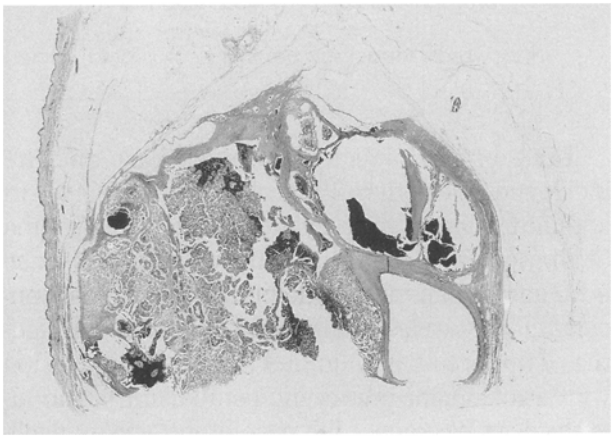

Fig 6. Histopathological findings showed multiple cysts, and solid papillary tumor appeared in the inside of one of these cysts (HE × 1).

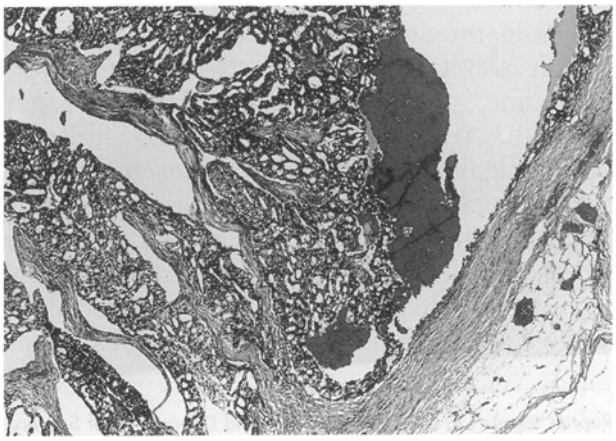

Fig 7. Histopathological findings revealed papillary proliferation of tumor cells. The inside of a part of the cyst wall was lined by manylayers of malignant cells (HE × 20).

terone receptor (> 80% of cells positive) antibodies. A total of 13 lymph nodes were removed, all showing no signs of metastatic disease.

The patient recovered well from surgery and was discharged from hospital after 1 week. Adjuvant therapy with tamoxifen 20 mg/daily was started.

Discussion

Intracystic papillary carcinoma of the breast is a type of breast cancer in which papillary carcinoma grows inside a cyst[11]. This disease is rare in females and exceedingly rare in males[2-10, 12]. Intracystic papillary carcinoma accounts for 0.5-2% of breast cancers in females, and an even lower percentage in males[2-10, 12]. Twenty-seven cases of intracystic carcinoma have been reported in men in the world[1-10, 13-15]. Generally, intracystic carcinoma is a ductal carcinoma *in situ*, but an invasive case of intracystic carcinoma in a male patient has been reported[7].

The mechanism of origin of intracystic carcinoma is still unclear. In a review of the literature, almost all cases of intracystic papillary carcinoma of breast in male had a single cyst[2-10]. Fallentin *et al.*[13] reported an intracystic breast carcinoma with a multilocular cyst in a male. The case described herein also had a multilocular cyst containing papillary carcinoma. According to Rosemond *et al.*[16], the malignant degeneration rate of breast cysts is 0.1%. Therefore, the malignant potential of a multilocular cyst may be higher than that of a single cyst.

The diagnosis of intracystic papillary carcinoma by physical examination and radiological imaging tends to be difficult[6-8, 17]. The cystic nature of this lesion commonly presents as a benign-appearing, well-circumscribed mass on these examinations[4, 6, 7, 9, 10, 13]. Ultrasonography can detect intracystic solid lesions on the wall of the cyst[6, 14], and many studies have reported ultrasonography to be useful in the diagnosis[4, 6, 7, 9, 17]. Computed tomography and magnetic resonance imaging can give additional information about the morphological features and vascularisation of the tumor and about the local spread[7, 14]. Generally ultrasound-guided fine needle aspiration can be performed safely to achieve a pathological diagnosis. However, fine needle aspiration may fail to establish the diagnosis of malignancy[4]. Pacelli *et al.*[9] reported that core needle biopsy was useful for diagnosing intracystic papillary carcinoma of the male breast. In addition, Blaumeiser *et al.*[7] described that some papillomas were at risk of malignant degeneration; therefore, biopsy should be applied liberally on diagnostic and therapeutic grounds whenever a papillary lesion is suspected. However, simple and easy fine needle aspiration is the first choice because there are some cases in which the malignancy is diagnosed by cytology of the intracystic lesion or cyst fluid, including the present case[15, 17, 18].

A review of the literature showed that almost all patients with intracystic papillary carcinoma of the male breast underwent modified radical or simple mastectomy[4, 6, 7, 9, 13, 14]. In addition, this disease in men is usually estrogen and progesterone receptor rich[6-8, 19]. The appropriate adjuvant endocrine or chemotherapy has not established because there are no prospective randomized trials; however, generally this disease receives the same adjuvant treatment as other types of female breast carcinoma[4, 7, 8]. Males and females with breast cancer have similar outcomes based on the stage of the disease and typically exhibit excellent prognosis with intracystic carcinoma[4, 7, 8].

The etiology remains unknown. Loss of heterozygosity (LOH) on chromosome 16q was a useful marker for intracystic papillary carcinoma, since intraductal papilloma showed no LOH. Lanzafame *et al.*[19] described that expression of BCL-2 in intracystic papillary carcinoma of the breast was consistent with an indolent clinical course and very favourable prognosis.

In conclusion, intracystic papillary carcinoma of the male breast is very rare. Mammography and ultrasonography for this lesion demonstrate benign disease. It should be kept in mind that the diagnosis of intracystic papillary carcinoma is suspected when imaging examinations reveal an intracystic mass. Pathological examination by fine needle aspiration or core needle biopsy is necessary. Excisional biopsy is recommended when possible.

References

1) Vetto J, Jun SY, Paduch D, Eppich H, Shih R, Padduch D: Stages at presentation, prognostic factors, and outcome of breast cancer in males. *Am J Surg* 177:379-383, 1999.
2) de Araujo PM, Dias MC: Papillary carcinoma of the male breast. *Acta Med Port* 2:221-223, 1989.
3) Madden CM, Reynolds HE: Intracystic papillary carci-

noma of the male breast. *Am J Roentgenol* 165:1011-1012, 1995.
4) Imoto S, Hasebe T: Intracystic papillary carcinoma of the breast in male: case report and review of the Japanese literature. *Jpn J Clin Oncol* 28:517-520, 1998.
5) Jonasson JG, Agnarsson BA, Thorlacius S, Eyfjord JE, Tulinius H: Male breast cancer in Iceland. *Int J Cancer* 65:446-449, 1996.
6) Tochika N, Takano A, Yoshimoto T, Tanaka J, Sugimoto T, Kobayashi M, Matsuura K, Araki K, Ogawa Y, Moriki T: Intracystic carcinoma of the male breast: report of a case. *Surg Today* 31:806-809, 2001.
7) Blaumeiser B, Tjalma WA, Verslegers I, De Schepper AM, Buytaert P: Invasive papillary carcinoma of the male breast. *Eur Radiol* 12:2207-2210, 2002.
8) Gupta D, Torosian MH: Intracystic breast carcinoma in a male: Unusual case presentation and literature review. *Oncol Rep* 9:405-407, 2002.
9) Pacelli A, Bock BJ, Jensen EA, Heerden JA, Reynolds C: Intracystic papillary carcinoma of the breast in a male patient diagnosed by ultrasound-guided core biopsy: a case report. *Breast J* 8:387-390, 2002.
10) Andres B, Aguilar J, Torroba A, Martinez-Galvez M, Aguayo JL: Intracystic papillary carcinoma in the male breast. *Breast J* 9:249-250, 2003.
11) Carter D, Orr SL, Merino MJ: Intracystic papillary carcinoma of the breast. After mastectomy, radiotherapy or excisional biopsy alone. *Cancer* 52:14-19, 1983.
12) Czernobilsky B: Intracystic carcinoma of the female breast. *Surg Gynecol Obstet* 124:93-98, 1967.
13) Fallentin E, Rothman L: Intracystic carcinoma of the male breast. *J Clin Ultrasound* 22:118-120, 1994.
14) Rosemond GP, Maier WP, Brobyn TJ: Needle aspiration of breast cysts. *Surg Gynecol Obstet* 128:351-354, 1969.
15) Ravichandran D, Carty NJ, al-Talib RK, Rubin C, Royle GT, Taylor I: Cystic carcinoma of the breast: a trap for the unwary. *Ann R Coll Surg Engl* 77:123-126, 1995.
16) Anan H, Okazaki M, Fujimitsu R, Hamada Y, Sakata N, Nanbu M: Intracystic papillary carcinoma in the male breast. A case report. *Acta Radiol* 41:227-229, 2000.
17) Gorczyca W, Olszewski W, Tuziak T, Kram A, Woyke S, Ucinski M: Fine needle aspiration cytology of rare malignant tumors of the breast. *Acta Cytol* 36:918-926, 1992.
18) Sonksen CJ, Michell M, Sundaresan M: Case report: intracystic papillary carcinoma of the breast in a male patient. *Clin Radiol* 51:438-439, 1996.
19) Lanzafame S, Emmanuele C, Torrisi A, Magro G: Correlated expression of BCL-2 protein, estrogen receptor, cathepsin D and low growth fraction (PCNA) in intracystic papillary breast carcinoma. *Pathol Res Pract* 194:541-547, 1998.
20) Tsuda H, Uei Y, Fukutomi T, Hirohashi S: Different incidence of loss of heterozygosity on chromosome 16q between intraductal papilloma and intracystic papillary carcinoma of the breast. *Jpn J Cancer Res* 85:992-996, 1994.