



Non-Operative Treatment of Intracystic Papilloma in the Male Breast

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Abstract

Intracystic Papilloma (ICP) in male breast is rare and only 13 cases of ICP in men have been reported worldwide so far. In all cases, local excision or mastectomy is performed immediately upon diagnosis for treatment of ICP. This case report describes a patient who was diagnosed with a benign cyst by core-needle biopsy and had received only non-operative management for 10 years. Then, this patient underwent local excision and diagnosed with ICP. The lump did not become malignant after 10 years from initial diagnosis. We suggest that immediate surgical excision may not always be necessary for ICP of the male breast after diagnosis by core-needle biopsy.

Introduction

Intracystic Papilloma (ICP) of the breast is one of the benign breast tumour in which a papillary lesion grows inside a cyst. ICP is rare in male breasts, and only 13 cases of ICP in men have been reported worldwide so far [1,2].

In a review of the literature on men with ICP, there was no predilection for age or the right or left breast [1]. All patients had palpable cystic masses. The size of these lesions ranged from 2 cm to 10 cm. The duration of the disease before diagnosis was more than 1 year in 40% of the patients [2]. The clinical features of ICP in men are similar, including a palpable cystic mass, bloody or serous nipple discharge, and pain. Clinically, there are no medical conditions associated with ICP in the male breast. However, 2 cases raised the possibility of a relationship between ICP in men and long-term therapy with phenothiazine, which has been known to increase the level of prolactin [2,3]. However, an explanation for this possible association is not clear yet.

Mammography or sonography is required to diagnose ICP and a definitive diagnosis can be made by a sonographically-guided core needle biopsy [1]. Pathologically, ICP is a hyperplastic polypoid lesion within the duct. The cyst of ICP is actually a cystically dilated duct containing the papilloma. Therefore, there is no histopathologic difference between intraductal papilloma and ICP, and the origin of its cyst is likely different from that of cysts that arise in the lobules [1].

In most cases, local excision or mastectomy is performed immediately upon diagnosis for treatment of ICP [1,2,4,5]. Here, however, we report a case of ICP in an 83-year-old man who was treated with aspiration, not surgery, for 10 years after diagnosis.

Case Presentation

An 83-year-old man visited our hospital with a lump in his left breast and nipple. He first discovered the lump 10 years ago. Ultrasonography and mammography were performed when the lump was originally found, and a definitive diagnosis of a benign cyst was made by core-needle biopsy. The lump size increased gradually after the diagnosis and was reduced by aspirating the cystic fluid. The aspiration treatment was repeated every time the cyst reappeared.

During his visit at our hospital, physical examination showed a 6 cm hard, movable lump below the nipple in the left breast. There were no enlarged axillary lymph nodes or nipple retraction. The patient had no record of medications that cause male breast enlargement, no known medical conditions, and no pleural injury.

Lumpectomy was performed under local anaesthesia without additional diagnostic examinations

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in our hospital because the patient had already been diagnosed with a benign cyst. The excised specimen was a cystoma with a volume of $6 \times 6 \times 3 \text{ cm}^3$ and the intracystic papillary lesion was 1.5 cm.

The cyst was identified as ICP by histopathology after surgery. In our case, the epithelium lining the papillary fibrovascular fronds in the lesion was mostly apocrine, containing large nuclei, nucleoli, and abundant cytoplasm. The patient has shown no recurrence for 1 year after the surgery.

Discussion

ICP in the male breast is very rare because of the rudimentary state of the mammary gland [1]. Since Simpson and Barson first reported ICP of the male breast in 1969, it has been reported in only 13 other patients [2,6].

Intracystic tumours can now be detected easily using new imaging modalities, including ultrasonography, computed tomography and magnetic resonance imaging. However, even with these imaging modalities, diagnosis of these lesions is difficult except in cases of intracystic carcinoma with invasive features on imaging [2].

Complex cystic breast masses have a possibility of being malignant and therefore confirmative diagnosis is usually indicated [7]. In order to make appropriate treatment decisions, ICP should be distinguished from Intracystic Papillary Carcinoma (ICPC). However, it is difficult to distinguish between benign and malignant lesion preoperatively by imaging or cytologic examinations. In particular, cytologic examination of specimens obtained by puncture aspiration are frequently misleading because cellular atypia is very low in the majority of ICPCs, and the floating cells in the cyst fluid are easily denatured [8]. Therefore, histological diagnosis is required, which means an excisional procedure. Alternatively, sonographically guided core needle biopsy can be performed as a less invasive method for differential diagnosis of ICP and ICPC when a solid component is present in the cyst [1].

The natural history or treatment of ICP in male breasts is not clearly defined. Therefore, the patients are being treated according to the protocol for intraductal papilloma in women. In cases of intraductal papilloma in women, surgery is recommended immediately upon diagnosis based on a report saying that the risk for breast cancer is elevated two-fold [4]. All previously reported cases of men diagnosed with ICP have also received local excision or mastectomy immediately once intraductal papilloma was diagnosed [1,2,4,5]. However, it cannot be concluded that the incidence of breast cancer in men with ICP is equal to that in women, because there have been no studies about the relationship between breast cancer incidence and ICP in men.

Navas, et al. reported the case of a patient who was diagnosed with ICP based on the results from laboratory tests after local excision of a lump that had existed for 6 years [5]. The current case had received only conservative management without surgery for 10 years. Then, our patient underwent local excisional surgery and was diagnosed with ICP based on laboratory test results. In both cases, the lump did not become malignant after 6 and 10 years, respectively, from the initial appearance or diagnosis.

In conclusion, appropriate treatment of ICP is controversial. We suggest that immediate surgical excision may not always be necessary for ICP of the male breast after diagnosis by core-needle biopsy. Case studies so far have only discussed the clinical features or pathogenesis of ICP in men. However, we have focused on the natural progression or treatment of ICP in men. We currently cannot come to any definitive conclusions about its natural progression or treatment due to the small number of cases of ICP in men that have been reported to date. However, it is expected that accumulation of studies like this will help to reveal the clinical features and treatment principles of ICP in men in the future, and make it possible to analyse the differences in ICP between men and women.

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