

## INTRACYSTIC PAPILOMA IN MALE BREAST

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### ERKEK MEMESİNDE İNTRAKİSTİK PAPILOM

#### ÖZET

Erkek memesinde intrakistik papillom (İKP) son derece nadir görülür. Sağ memesinde İKP saptanan literatürdeki 13. erkek olguyu sunduk. Olgunun yaklaşık 6 aylık genişleyen ağrılı kitle hikayesi vardı. Ultrasonografide intrakistik bileşenli bir kist saptandı. Benign İKP nin radyolojik ve klinik özellikleri intrakistik karsinom ile benzer olduğundan, solid komponenti olan kompleks kistik kitleler biyopsi gerektirir. Benign İKP ve intrakistik karsinom ayırıcı tanısında ultrasonografi rehberliğinde biyopsi uygulanabilir. Aksi taktirde cerrahi rezeksiyon İKP için tanı ve tedavi yöntemi olarak önerilmektedir. Olgumuza lokal anestezi altında eksizyonel biyopsi uygulandı. Patolojik inceleme intrakistik bileşeni papillom olarak saptadı.

Anahtar sözcükler: papillom, erkek, meme kisti

### ABSTRACT

Intracystic papilloma (ICP) of the breast in males is extremely rare. We reported the 13th male patients presented in literature, complaining about ICP in his right breast. The patient had a history of a painful enlarging mass for about 6 months. Ultrasonography revealed a cyst with an intracystic component. Because the radiologic and clinical features of benign ICP are similar as intracystic carcinoma, complex cystic masses with solid components require biopsy. Sonographically guided core needle biopsy can be performed for differential diagnosis of benign ICP and intracystic carcinoma. Otherwise surgical resection is recommended as a diagnostic and therapeutic method for ICP. Excisional biopsy was performed under local anesthesia for our patient. Pathological examination revealed the intracystic component to be benign IP.

Key words: papilloma, male, breast cyst

### Introduction

Intracystic papillary tumors of the breast are extremely rare lesions (1). Even more rare, is the presence of an intracystic papillary mass in the male breast because of the rudimentary state of the mammary gland (2-5). Intracystic papillary tumors of the breast are generally benign and more frequently include intracystic papilloma (ICP). The clinical features of ICP in males are a palpable mass and nipple discharge (2, 4, 5). However, there have been only a few cases published on ICP of the male breast. Through a search of pubmed about ICP in male breast cases reported in the medical literature published during the years 1950–2009, this is the 13 th reported case of ICP of male breast in the world. We present the clinicopathological features of intracystic papilloma in a 23 year old male.

### Case report

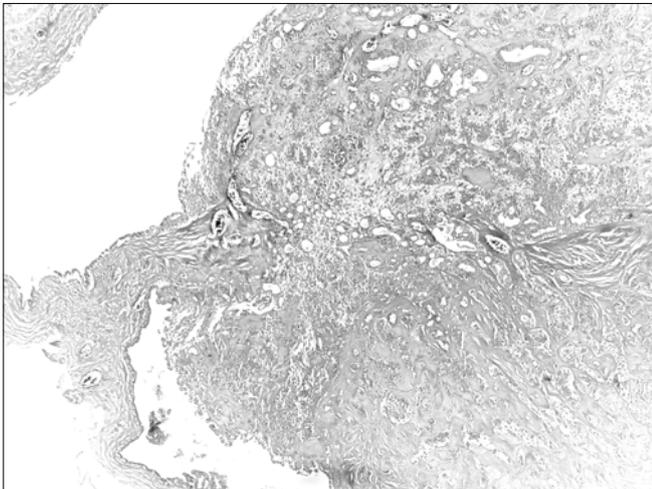
A 23 year old man presented with a history of painful right breast mass, which he had noticed approximately six months prior to presentation. The mass had gradually enlarged in size within 6 months. There were no relevant features in his medical history including gynecomastia or chest trauma. In addition, there was no history of breast cancer in his family. The patient had no nipple

retraction, discharge, or bleeding. Physical examination revealed an elastic and movable mass of 3.0 x 2.0 cm in size that localised at 4 cm far away from the areola in the superior medial quadrant of the right breast. The mass had a smooth surface and clear margins. There was no ulceration of the overlying skin, nor were there any palpable axillary lymph nodes observed. The left breast was unremarkable.

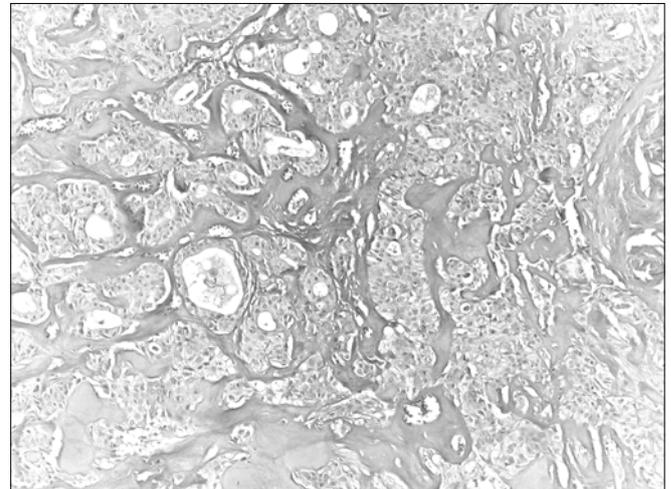
Serum hormonal levels, including prolactin, follicle-stimulating hormone, luteinizing hormone, estradiol, and testosterone were all normal.

Ultrasonography revealed a 26x19 mm cystic lesion with an 8x7 mm solid papillary lesion extending into the cyst lumen at the superior medial quadrant of the right breast. There were no suspicious lymph nodes on sonography.

Excisional biopsy was performed under local anesthesia. The resected specimen was a cyst, measuring 28x22x20 and intracystic papillary lesion, measuring 7x7x6 mm, on the inside of the chest wall. Histopathologically, this intracystic papillary lesion was



**Figure 1.** Intracystic papiller lesion surrounded with myoepithelial cells.



**Figure 2.** Cyst wall covered by myoepithelial cells and cubic-columnar epithel.

papilloma. Sections revealed chest wall covered by myoepithelial cells and cubic-columnar epithel and papiller lesion surrounded with myoepithelial cells containing proliferating epithel islands with fenestration. Features of gynecomastia were not seen. The histopathological diagnosis was intracystic papilloma (Figure 1, 2). He had shown no recurrence one year after surgery.

### Discussion

The development of ICP in males is extremely rare and it has been reported only in twelve previous patients (2, 5-8). ICP of the breast is a type of breast tumor in which a benign papillary lesion grows inside a cyst. The clinical features of ICP in males showed similarity including palpable cystic mass, bloody or serous nipple discharge and pain. Our patient's clinical presentation was a painful mass in the breast. The age of the patients published, ranged from seven months to 82 years (2, 9). Our patient was 23 years old. Generally, the cyst with intracystic papilloma of the breast is small. The sizes of ICPC cysts are larger than those of ICP, and intracystic papillary lesions larger than 3 cm, are generally malignant. Our patient had a cyst smaller than 3 cm. There were 4 cases of intracystic papilloma of the breast in male forming greater than 5 cm in size.

No dominance was observed in left or right breast. Our patient had ICP in his right breast. In addition, ICP usually occurs in retroareolar localization, however in our patient the palpable mass was 4 cm away from the areola.

Some authors tried to show a relationship between male intracystic papilloma and hormonal status. Yamamoto et al. reported a case given long-term phenothiazine therapy, which is known to increase serum prolactin levels (2). And since two of the males with intracystic papillomas were given long-term phenothiazine therapy, they suggested that phenothiazines or prolactin may participate in the tumorigenesis of this rare tumor (2, 4). But this theory remained unclear. Similarly with the other 11 ICP male patients our patient hormonal status was normal.

Imaging features of the previously reported cases showed typical findings of a complex cystic mass. Complex cysts contain cystic and solid components and are associated with a variety of benign, atypical, and malignant pathologic diagnoses. Mammography mostly shows a discrete dense mass in the subareolar portion (5, 10). In our patient preoperatively, only US was performed and it showed a 26x19 mm cystic lesion including an 8x7 mm solid papillary lesion.

Complex cystic breast masses have a substantial chance of being malignant therefore percutaneous or surgical biopsy is usually indicated (11, 12). ICP should be distinguished from intracystic papillary carcinoma (ICPC). However, it is difficult to distinguish between ICP and ICPC by imaging or cytologic examinations preoperatively. Especially, the cytological picture revealed by aspiration is also frequently misleading because cellular atypia is very small in the majority of ICPCs, and the floating cells in the cyst fluid are easily denatured. Techniques that may be used for sampling of complex cystic breast masses include fine-needle aspiration, core-needle biopsy (with an automated spring-loaded or vacuum-assisted device), and surgical excision. Intracystic masses detected on US are commonly aspirated to confirm the diagnosis. However the clinical validity of atypical findings identified in cyst aspirate fluid is often very low. The final diagnosis depends on excisional biopsy. There have been some reports that describe the methods to distinguish between ICP and ICPC preoperatively. Inaji et al. reported that the combination assays of epidermal growth factor receptor 2 and carcinoembryonic antigen in cyst fluid produced very encouraging results in diagnosing intracystic breast cancer. Their sensitivity of the combination test for intracystic cancer was higher than that of cytology (13). Mammary ductoscopy is suggested as a useful diagnostic tool in patients with pathological nipple discharge. Berna et al. reported a percutaneous endoscopic approach to intracystic breast lesions as a potential alternative to open surgery (14). However all of these reports included female patients. In cases of ICP in males, the histological

diagnosis was made by excisional procedure in 10 cases and mastectomy in three cases. In our case, excisional biopsy was used as both a diagnostic and therapeutic tool.

ICP without atypia is histopathologically composed of multiple branching fibrovascular cores lined by two layers of epithelial and myoepithelial cells. On the other hand, microscopically, ICPC are characterized by one, or occasionally several, nodules of papillary carcinoma surrounded by a thick fibrous capsule. Myoepithelial cells are not present in the papillae of encapsulated papillary carcinomas (3,12).

In follow up of these cases most of these cases showed no recurrence. Only Sara's case showed recurrence locally six months later that was successfully treated by reexcision (4). Our patient showed no recurrence in their one year follow up.

ICP in males very rarely develops. The differential diagnosis of ICP and ICPC is extremely difficult preoperatively. Frequently they show complex cystic breast masses that are suspicious for malignancy and usually warrant biopsy. US guided biopsy might be useful but excisional biopsy is required for both diagnostic and therapeutic purposes.

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