Co-Existence of Two Rare Entities in the Male Breast: Intraductal Papilloma and Angiolipoma

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ABSTRACT
Intraductal papilloma and angiolipoma lesions are very rare in male breasts and gynecomastia is the most common male breast pathology. A 52-year-old healthy Caucasian male patient with right nipple pain for one month and two subareolar and periareolar masses had no other abnormal clinical or laboratory findings. After ultrasound examination, pull-through excision was made with a circumareolar incision in both lesions and the samples were sent for pathological examination. Histopathological examination revealed intraductal papilloma and angiolipoma on the basis of gynecomastia. This case is unique because both lesions are extremely rare and this is the first report of concurrent occurrence in a male breast.

Keywords: Papilloma, intraductal; angiolipoma; gynecomastia; male; breast

Introduction
Morphologically male breasts are composed of glandular and fatty tissues, as in females. These glandular units only consist of ducts that are typically delimited below the nipple-areolar complex (1).

Gynecomastia is the most common male breast pathology, and its prevalence in males with breast-related disorders varies between 32% and 100% according to age groups (1, 2).

Intraductal papilloma is a proliferative lesion of the mammary ducts and is usually completely benign, but can sometimes contain atypical or even malignant cells. The benign intraductal papilloma consists of abundant stroma containing both luminal epithelium and myoepithelial cells, forming several broad fronds (2-4).

Angiolipoma is an unusual vascular variant of the lipoma, the etiology of which is controversial and represents 5%-17% of all benign fatty tumors. This lesion is mostly localized in the subcutaneous tissues of the trunk and extremities, and breast angiolipoma is extremely rare. In addition, differential diagnosis of breast angiolipomas can be difficult as they can be confused with malignant lesions clinically, radiologically and pathologically (5, 6).

Both intraductal papilloma and angiolipoma lesions in male breasts are very rare and a few cases have been reported in the literature (1-3, 7).

Here, a case in which the co-existence of intraductal papilloma and angiolipoma in the male breast with gynecomastia is presented with the help of ultrasonographic and pathological images. In addition, the relevant literature is reviewed.
Case Presentation

A 52-year-old healthy Caucasian male patient was admitted to the surgical outpatient clinic with complaints of right nipple pain and two subareolar and periareolar masses for one month. Physical examination revealed firm, tender, well-circumscribed, nodular masses of approximately 2 cm and 1 cm in diameter, which could be palpated in the right retroareolar region. The remaining breast areas were symmetrical and had normal nipple-areolar complex. No erythema or pitting of the skin was observed. No palpable bilateral axillary or supraclavicular lymph nodes were found. Except for antiarterial hypertension drugs, he had no history of local trauma, recent weight loss, or use of anabolic steroids or other drugs that could cause gynecomastia. There was no relevant family history. The patient had no other abnormal clinical or laboratory findings.

Mammography could not be performed because the breast of the case was not large enough and was extremely painful. Ultrasound examination revealed a hypoechoic solid mass with a maximum diameter of 2 cm in the right retroareolar region with coarsely lobulated contours (Figure 1) and a well-circumscribed hyperechoic solid mass with a maximum diameter of 1 cm immediately medially (Figure 2).

Total pull-through excision with circumareolar incision was performed for both lesions and the samples were sent for pathological examination. Histological examination revealed an intraductal papilloma (Figure 3) in the large lesion, with no evidence of atypia or malignancy, on a background of gynecomastia (Figure 4), and an angiolipoma (Figure 5) in the small lesion.

**Figure 1.** Ultrasound image shows a retroareolar, hypoechoic solid mass with coarse lobulated contours

**Figure 2.** Ultrasound image shows a medial retroareolar, hyperechoic solid mass with well circumscribed contours

**Figure 3.** Intraductal papilloma. a) It is observed that papillary structures with fibrovascular cores in the enlarged duct are lined with epithelial and myoepithelial cells (H&E, x10) and b) (H&E, x20). c) Immunohistochemical staining of myoepithelial cells with p63 was observed in intraductal papilloma areas (H&E, x10)

H&E: hematoxylin and eosin stain
Male breast disease is often not recognized due to rarity, lack of awareness, and the scarcity of epidemiological data in the literature when compared to the female breast. Male and female breasts are similar at birth. Subareolar ducts in males are histologically similar to those in prepubertal females. An adult normal male breast usually consists of large ducts that do not extend beyond the central subareolar segment without the formation of lobules and acini. These ducts are embedded in the fibrous stroma and adipose tissue (8).

Gynecomastia can occur in any age group, and the risk factors for all the same breast lesions are similar. These include age, family history, medications, obesity, endocrine and hormonal imbalance, systemic disease, liver disease, neoplasm, history of orchitis or thoracic radiotherapy, and genetic predisposition in patients with Klinefelter syndrome, or BRCA2 and the P53 gene positivity (8).

In males, a retroareolar mass can be benign, such as an intraductal papilloma or any soft tissue tumor, or sometimes malignant. Intraductal papillomas of the male breast are rare, in contrast to females (3, 8).

The clinical presentation of intraductal papilloma and malignant lesions is similar, with a unilateral bloody or serous discharge associated with a palpable, unilateral, firm, fixed lesion in the subareolar region in males. It may be associated with skin changes or axillary lymphadenopathy (8).
The authors certify that they have received a consent from the patient. The form gave consent for patient pictures and other clinical information to be reported in the journal.

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