



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

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Key Points

- Both intraductal papilloma and angiolipoma are extremely rare entities in males. To the best of our knowledge, this is the first report of concurrent occurrence of intraductal papilloma and angiolipoma in a male breast on the background of gynecomastia.

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.

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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 anti-arterial 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).



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

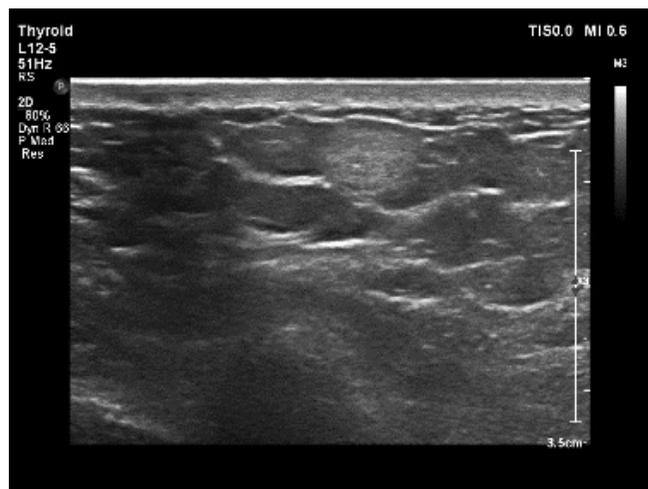


Figure 2. Ultrasound image shows a medial retroareolar, hyperechoic solid mass with well circumscribed

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 angioliipoma (Figure 5) in the small lesion.

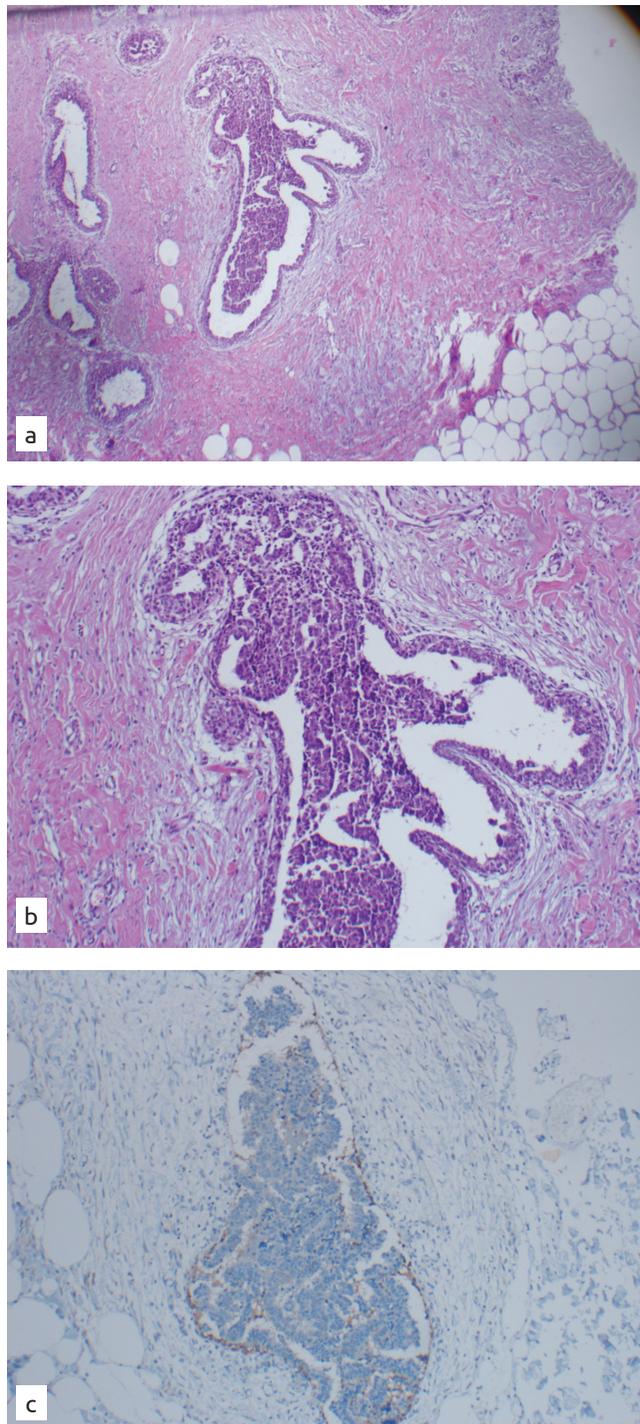


Figure 3. Intraductal papilloma. a) It is observed that papillary structures with fibrovascular cores in the enlarged duct are lined with epithelial and myoepithelial cells (H&E, x10) and b) (H&E, x20). c) Immunohistochemical staining of myoepithelial cells with p63 was observed in intraductal papilloma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

H&E: hematoxylin and eosin stain

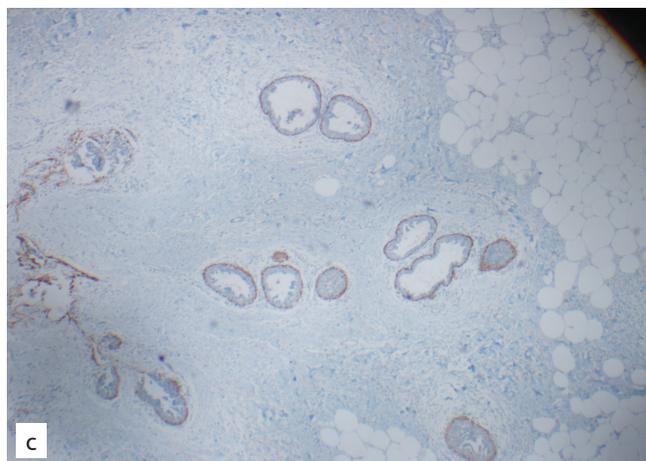
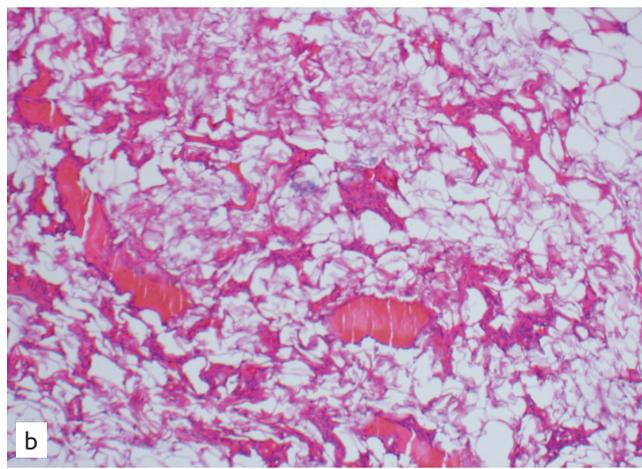
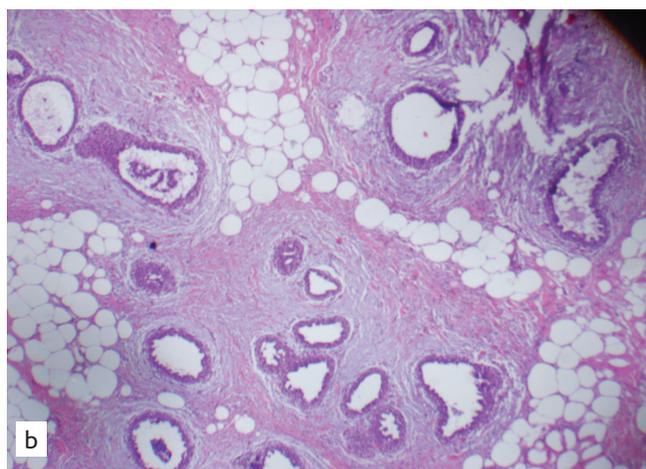
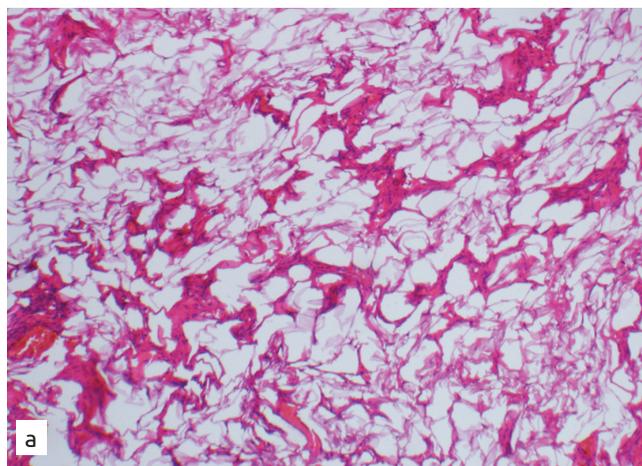
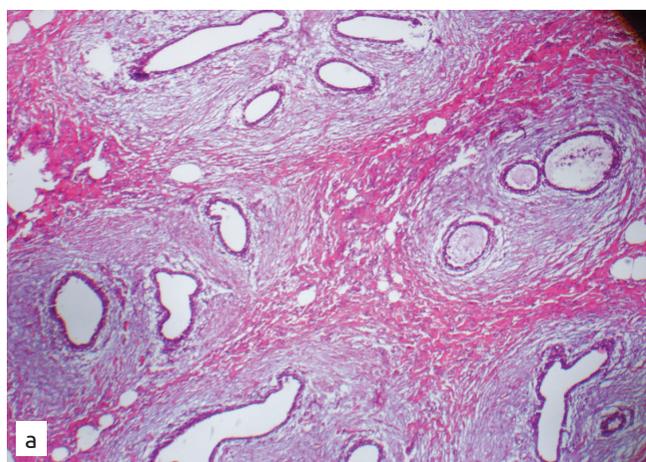


Figure 4. Florid type gynecomastia. **a)** Budding is seen in the proliferating ducts in the fibroblastic stroma (H&E, x20). **b)** In addition to the surrounding fibroadipose tissue, budding is also seen in the proliferating ducts in the fibroblastic stroma (H&E, x20). **c)** Immunohistochemical staining of myoepithelial cells with p63 is observed in areas with gynecomastoid changes (H&E, x10)

H&E: hematoxylin and eosin stain

Discussion and Conclusion

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

Figure 5. An encapsulated nodular lesion (angiolipoma) containing mature adipose tissue and vascular tissue proliferation. **a)** Mature adipose tissue contains thick-walled vessels with branching capillaries and pericytes (H&E, x20). **b)** Fibrin thrombi were seen in the lumen of some vascular structures (H&E, x20)

H&E: hematoxylin and eosin stain

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 diagnostic approach to intraductal papillary lesions includes physical examination, mammography, ultrasonography, magnetic resonance imaging, and more rarely, breast ductoscopy or ductography. When there is discharge, cytological examination of discharge material may also be required. Differential diagnosis between benign intraductal papilloma and a carcinoma with atypia or even malignancy is not possible with imaging alone. Therefore surgical excision and histological confirmation are required (3).

Microscopically, intraductal papilloma appears as an epithelium containing both luminal and myoepithelial cells and abundant stroma forming several broad leaves (8).

Lipomas are encapsulated proliferations of mature adipocytes but the cause of angioliipoma is unknown. Breast angioliipomas may present as solitary or multiple breast masses, and angioliipomas do not have a typical imaging appearance. The diagnostic key may be the homogeneous echogenic sonographic appearance, which is unusual for breast masses. However, in the differential diagnosis of masses with increased echotexture, focal acute hemorrhage or acute hematoma, focal fibrosis, hemangioma, spindle cell lipoma and malignancy are included, in addition to angioliipoma. The histological appearance of angioliipoma in the subcutaneous tissue of the breast is not different from comparable lesions at other subcutaneous locations. Microscopically, angioliipomas consist of mature adipose cells separated by a network of branching small vessels. Diagnosis of cellular angioliipoma can potentially lead to diagnostic pitfalls involving better-known formations such as angiosarcoma or Kaposi's sarcoma of the breast region (5).

In this case, intraductal papilloma with angioliipoma was observed, together with gynecomastia in the right breast. This case is unique because both lesions are extremely rare and are reported here for the first time in a male breast with gynecomastia. The diagnosis was confirmed by histological analysis as intraductal papilloma and angioliipoma without any atypia or evidence of malignancy.

The proportion of men with breast complaints is increasing day by day, and although mammography plays an important role in distinguishing between benign and malignant breast diseases (9), especially in the elderly, mammography examination could not be performed in our case.

In conclusion, both intraductal papilloma and angioliipoma lesions in male breasts are very rare in the literature. A systematic search in PubMed, Web of Knowledge, and EBSCO found only a few cases of intraductal papilloma and a few angioliipomas in the breast in males, but not together. Here, the co-existence of intraductal papilloma and angioliipoma in the breast of a male patient with gynecomastia

is presented as an extremely rare case that has not, to the best of our knowledge, been described before.

Informed Consent: 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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Authorship Contributions

Surgical and/or Medical Practices: E.C.Ö.; Concept: M.B.; Design: M.B.; Data Collection and/or Processing: M.B., H.B., E.C.Ö.; Analysis and/or Interpretation: E.C.Ö.; Literature Search: M.B., H.B.; Writing: M.B.

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