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# A rare case of giant papillary adenoma of the breast

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SUMMARY: A rare case of giant papillary adenoma of the breast.

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The authors present a case of giant papillary adenoma of the breast and discuss their therapeutic strategy. The patient subsequently returned due to a local recurrence, which was treated with oncoplastic surgery, with satisfactory aesthetic results.

The authors conclude by stressing the considerable rarity of this disease and the need for effective cooperation between surgeons and pathologists. RIASSUNTO: Su un raro caso di adenoma papillare gigante della mammella.

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Gli Autori espongono un caso di adenoma papillare gigante della mammella giunto di recente alla loro osservazione e discutono della strategia terapeutica adottata. La paziente è ritornata alla loro osservazione per recidiva locale trattata con chirurgia oncoplastica, con soddisfacenti risultati estetici.

Gli Autori concludono sottolineando la notevole rarità della patologia e la necessità di una fattiva collaborazione tra chirurghi e anatomopatologi.

KEY WORDS: Giant papillary adenoma - Oncoplastic surgery - Subcutaneous mastectomy. Adenoma papillare gigante - Chirurgia oncoplastica - Mastectomia sottocutanea.

## Introduction

Giant papillary adenoma of the breast is a rare tumor which although itself benign, has features suggesting malignity, making it easy to under- or overestimate its biological behavior. Haagensen (6) considers it to originate from a papilloma of the nipple, i.e. of the large collecting ducts, which can deform and erode the nipple with clinical signs similar to those of Paget's disease. It is characterized by a sometimes papillary proliferation seeming to infiltrate the glandular tubules branching in the stroma, which becomes fibrous.

There are two anatomopathological forms (2, 3), which although having similar clinical signs, are completely distinct. The less common, solitary papilloma, grows in

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a dilated lactiferous duct and can become large enough to be visible on the surface of the nipple through the opening of the duct (8, 11). The most common type is a diffuse adenomatous and papillary epithelial proliferation which normally develops (15) on the surface of the nipple and at a relatively early stage erodes the epithelium, presenting on the surface as a granulated area. Haagensen coined the term papillary adenoma for this form, but it has also been defined as florid papillomatosis of the nipple or erosive adenomatosis. Histological examination reveals ductule structures, sometimes dilated, lined with normal epithelial and myoepithelial cells (7, 13).

### **Case report**

A 38-year-old woman recently came to our attention with a large growth, completely occupying the outer quadrants of the right breast, which had developed six months earlier while she was breastfeeding. On her doctor's advice, she had stopped nursing and taken antibiotics and anti-inflammatories, with little benefit. She also reported a milky secretion from the right nipple.

On inspection, the right breast appeared larger than the other. Palpation revealed a hard, irregular growth which could not easily be dissociated from the remaining parenchyme, occupying the ou-



Fig. 1 - Surgical specimen.

ter quadrants of the breast as well as part of the upper inner quadrant and the retroareolar area. Ultrasound revealed an area of diffusely inhomogeneous tissue with a predominantly solid component, probably neoplastic. Tru-cut biopsy lead to the suspicion of a carcinomatous lesion.

As surgery was indicated, the patient was informed of the need to remove the tumor completely and warned that radical mastectomy might be performed if the intraoperative histological examination proved positive for carcinoma.

The surgery involved a radial incision at the confluence of the outer quadrants of the right breast, separation of the cutaneous layer above the growth, mobilization of the tumor and resection from the deep glandular and retroareolar layers. After removal, the growth was found to be oval, hard, and 10 x 6 cm in size. Cutting revealed it as a homogenous parenchymatous mass with fibrous aspects, with no signs of normal glandular tissue visible to the naked eye (Fig. 1). The histological examination revealed it to be a giant papillary adenoma. A mastoplasty was thus carried out, with suturing in layers and positioning of a suction drain.

#### The post-operative course was normal.

The final pathological examination revealed a "bi-lobed multinodular structure lined with several rows of cylindrical cells consisting of large, moderately polymorphous papillary and cell formations, with signs of apical secretion; no signs of infiltration" (Fig. 2).

During a routine follow-up examination two years after the operation, an ultrasound scan revealed the presence of tumoral tissue with a solid nodular appearance in the retroareolar area. Recurrence was diagnosed and the patient was re-operated to remove the growth. To restore the aesthetics of the breast affected by the previous surgery, it was decided to perform a subcutaneous mastectomy with the insertion of a tissue expander and complementary construction of a cutaneous rotator flap (Fig. 3).

Recurrence of the papillary adenoma was confirmed by histological examination.

# **Discussion and conclusions**

A review of the literature revealed that no cases of giant papillary adenoma of the breast similar to that described here have ever been reported. They are in fact reported as uncommon (12) tumors with a hard, fibrous, nodular appearance, which can cause deformation of the skin and nipple but never reach a size of more than 3 cm. They have been described using various terms (10), such as flo-

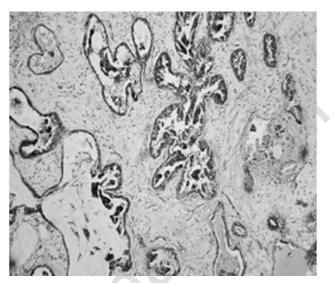


Fig. 2 - Histologicy (see text).



Fig. 3 - Subcutaneous mastectomy.



Fig. 4 - Final result.

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rid papillomatosis of the nipple and erosive adenomatosis (16); they affect both males and females, normally in adulthood. There is often a bloody or serous secretion and sometimes erosion of the epidermis, thus mimicking Paget's disease of the nipple. Histological examination reveals ductule structures, sometimes dilated, lined with normal epithelial and myoepithelial cells.

This case differed from classic papillary adenoma, confirming that uncritical use of diagnostic imaging may lead to the overestimation - or still worse, the underestimation - of diseases, like this, whose rarity means that they

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are often not recognized during diagnostic imaging (9) and are only diagnosed by intraoperative histological examination. In any case, the ever more widespread use of screening for breast cancer means that cases such as this are normally found at an early stage.

The multidisciplinary cooperation of radiologists, pathologists and plastic surgeons enables not only diagnosis and timely treatment (1, 4), but also the restoration of an aesthetic condition enabling women, like our patient, to face the diagnostic and therapeutic process with greater confidence (5, 14).

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