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Case Report

Syringomatous Tumor of the Breast: A Case Report

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Abstract: Syringomatous tumor is a rare and benign clinical condition, first described in 1983, characterized by infiltrative glandular proliferation that can affect the nipple-areola complex. Due to its rarity and clinical presentation, the differential diagnosis with malignant neoplasms represents a challenge. In this report, through the collection and analysis of medical records, we present the case of a 29-year-old female patient with a history of a persistent eczematous lesion on the left nipple, accompanied by nipple discharge. After several clinical and radiological evaluations, including ultrasounds and biopsies, surgical excision of the lesion was performed and histopathological analysis revealed a syringomatous tumor, with compromised surgical margins, but no evidence of malignancy. The patient was initially treated with local resection, but developed postoperative necrosis, requiring additional interventions. Despite being a benign lesion, local recurrence is a risk when excision is not complete. The present case emphasizes the importance of an accurate diagnosis and an appropriate therapeutic approach to prevent recurrences and unnecessary surgical complications.

Keywords: Breast Neoplasms; Breast Tumor; Diagnosis Differential; Syringomatous tumor; Case Report.

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1. Introduction

In 1983, Rosen first described syringomatous adenoma of the nipple (SAN), a benign breast tumor characterized by locally infiltrative proliferation and histologically resembling syringomas [1]. In 2012, the WHO recommended a nomenclature change to syringomatous tumor (SyT) due to the locally invasive nature of the lesion [2]. SyT is a rare tumor, and its diagnosis can be challenging. Based on the histological classification of mammary gland tumors, the differential diagnoses include mammary adenomas and low-grade adenosquamous carcinomas (LGASC).

Complete excision is curative, with local recurrences occurring in 30% of incompletely excised lesions. A single case of possible micrometastasis to a sentinel lymph node of such a tumor has been reported [3]. The infiltrative syringomatous tumor of the nipple clinically mimics an underlying breast malignancy and therefore usually draws the attention of the surgeon and surgical pathologist, but less so the dermatologist and dermatopathologist. Some patients have undergone mastectomy due to the erroneous interpretation of the lesion as tubular carcinoma [4] or adenosquamous carcinoma [5].

This case report aims to describe the history of a patient with a syringomatous tumor presenting with an eczematous lesion on the left nipple, as well as to compare the case

with findings in the literature, highlighting its rarity and the importance of differentiating it from breast neoplasms.

2. Case Report

A 29-year-old female patient was initially followed up at the Centro Municipal de Atenção Especializada (CEMAE) in Vitória da Conquista - BA, and from 2020, at the Instituto de Mastologia (IMASTO). She had a family history of breast cancer, nulliparous, G0P0A0, was on Puran T4 (levothyroxine) and Combined Oral Contraceptive (COC) Primera 20 (desogestrel and ethinylestradiol) for two and a half years, with no known drug allergies. In 2014, she initially presented with a painless mass in the left breast, accompanied by purulent and foul-smelling discharge, which was slow to heal, and evolved with deformities and hardening of the ipsilateral areola. Upon seeking medical attention, a biopsy of the left nipple revealed no malignancy, showing infundibular cysts and subepithelial vascular proliferation without atypia. Additionally, she reported having previously undergone three biopsies.

Ultrasonography (US) performed in 2017 and 2019 were normal (BI-RADS 1). On physical examination in 2020, an eczematous lesion was observed on the left nipple and areola, with no suspicion of malignancy. Initially, a punch biopsy or referral to a dermatologist was suggested, with the patient opting for the latter. One month later, a new US was performed, revealing thickening of the periareolar and papillary skin on the left side, with subtle Doppler flow and intermediate elastography, categorized as BI-RADS 2, with no solid lesions or cysts. Given this scenario, the management plan was annual US follow-up.

In January 2022, the patient returned with complaints of sharp pain and discharge from the left nipple, expressing a desire to undergo surgery to remove the lesion. Physical examination revealed enlargement of the eczematous lesion on the left nipple, measuring approximately 4 cm in diameter. At that time, the plan was to perform a new US and prepare for surgical intervention. The US indicated thickening of the areolomammillary complex with an edematous appearance, possibly of inflammatory nature (BI-RADS 2). Five months later, she underwent surgical resection of the lesion. Histopathological examination of the surgical specimen revealed a syringomatous tumor, with coincident resection margins, and immunohistochemistry (IHC) was recommended. Postoperatively, the patient developed local necrosis with granulation tissue.

Following ambulatory dressing care and improved healing, the patient returned with a scar on the left breast due to excision of the areolopapillary complex after undergoing a second surgery for margin removal. The second surgery occurred after the IHC result confirmed a syringomatous tumor with no signs of malignancy; however, complete removal of the lesion with clear surgical margins was crucial to avoid recurrence. One month later, she returned with the result of a Magnetic Resonance Imaging (MRI) that revealed a 7mm lymph node in the superolateral quadrant (SLQ) of the left breast (BI-RADS 2). Thus, the patient remains without complications to date and is scheduled for a follow-up with US and Mammography (MMG) in one year.

3. Discussion

We present the case of a 29-year-old female patient with a family history of breast cancer, who has never been pregnant and has been using Puran T4 and the Combined Oral Contraceptive (COC) Primera 20. The first biopsy in 2015 revealed infundibular cysts and subepithelial vascular proliferation without atypia; between 2017 and 2019, she had normal ultrasonographies (BI-RADS 1); and in 2020, an eczematous lesion was found on the left nipple and areola (BI-RADS 2). In 2022, a quadrantectomy was performed to remove the lesion, and histopathology confirmed a syringomatous tumor with coincident resection margins, leading to a second surgery, ultimately achieving clear margins. Postoperatively, there was local necrosis and healing, and an MRI revealed a 7mm lymph node

(BI-RADS 2), interpreted as reactive. The patient remained without complications and was scheduled for annual US and MMG monitoring.

The diagnosis of a syringomatous tumor is rare; it is a benign tumor of eccrine glands, locally infiltrative, non-metastatic, with a generally benign prognosis. However, a high level of suspicion is necessary to avoid overtreatment, as it is difficult to distinguish from malignant tumors on imaging studies [6,7,8,9]. It is commonly misinterpreted even in pathology, as carcinoma in frozen section biopsy or by fine needle aspiration.

The literature confirms that syringomatous tumor represents a diagnostic challenge for conventional imaging methods [10,11]. The infiltrative growth pattern of SyT is characteristically superficial, restricted to the nipple dermis and capable of infiltrating the smooth muscle of the areola, while only rarely extending into the deep dermis [12]. This superficial infiltrative architecture does not produce the typical radiological features of malignancy (spiculated margins, deep architectural distortion, or abnormal Doppler vascularization), resulting in repeatedly benign BI-RADS classifications despite the presence of a clinically evident lesion [10,11].

The BI-RADS 1 and 2 classifications assigned over several years in this patient, despite a persistent and progressive lesion, reflect the intrinsic limitations of imaging modalities in detecting superficial adnexal tumors of the nipple-areolar complex. Although ultrasound is the preferred modality for evaluating nipple lesions, it frequently underestimates the true extent of superficial infiltrative lesions such as SyT [10]. Breast MRI has been described as a superior modality for assessing involvement of the nipple-areolar complex; however, it was not performed during the preoperative period in this case [10]. This diagnostic difficulty, combined with the methodological limitations of fragmented biopsies for this specific histological subtype, contributed to the delayed excisional biopsy because of a combination of factors: limited access to specialized care, slow and initially minimally symptomatic clinical progression, and imaging studies repeatedly categorized as benign.

SyT exhibits stromal infiltration and tear- or comma-shaped tubules. Immunohistochemistry typically shows positivity for p63 and SMA [13]. Its main differential diagnosis is low-grade adenosquamous carcinoma, a distinction that represents one of the most complex challenges in breast pathology because both entities share significantly overlapping morphological and immunohistochemical features and may be virtually indistinguishable on microscopic examination [2,14]. In the present case, the findings that allowed exclusion of malignancy and confirmation of SyT included: superficial anatomical location restricted to the nipple-areolar complex; characteristic histological architecture; and absence of LGASC features, such as deep infiltration of the breast parenchyma, band-like desmoplastic stromal reaction surrounding neoplastic glands, or peripheral lymphocytic aggregates [2,12,14].

Its clinical presentation is variable. Other cases described in the literature predominantly involve female patients aged between 11 and 83 years (mean age 40 years), with mostly unilateral breast symptoms such as a nipple mass, which may present as dense, swollen, painful or painless, and not reduced by compression or manually. Symptoms may also include itching, discharge, crusts, and nodules [6,7,8,9]. Thyroid comorbidities, such as multinodular goiter, as well as irritable bowel syndrome and anxiety, have also been described [6].

Tumor sizes range from 0.75 to 7 cm, and size is not related to the quality of outcomes after treatment. These tumors are treated surgically, primarily via local or complete excision, as clear margins are important to prevent recurrence [6,8]. Despite this, when negative margins are achieved and the tumor is completely removed, there is generally no recurrence within six years [13]. Awareness of this tumor is essential to prevent delayed diagnoses, overtreatment, such as mastectomy, aggressive local excisions, or lymph node biopsies. The main differential diagnosis is low-grade adenosquamous carcinoma, and follow-up can generally be conducted with clinical monitoring [6,7,9,13].

4. Conclusion

The syringomatous tumor of the breast, although rare, presents a diagnostic challenge, mainly due to its clinical and histological similarity to malignant breast lesions, such as low-grade adenosquamous carcinoma. The reported case highlights the importance of precise diagnosis and complete excision with clear surgical margins to avoid recurrence; as well as the early consideration of excisional biopsy, regardless of BI-RADS classification, in the presence of persistent nipple-areolar complex lesions showing discordance between clinical and radiological findings. The patient, with a family history of breast cancer and no prior pregnancies, presented with an eczematous lesion on the left nipple, initially interpreted as benign. However, the clinical evolution and subsequent histopathological findings confirmed the presence of a syringomatous tumor.

This case emphasizes the need for awareness among surgeons, pathologists, and dermatologists about the existence of SyT to avoid misdiagnosis and overtreatment, such as unnecessary mastectomies. The literature points to a generally benign prognosis when treatment is performed appropriately. However, due to the rarity of the tumor, it is essential to continue reporting and discussing cases to improve diagnostic and therapeutic strategies, thus ensuring the best possible management for these patients.

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