

Nipple Adenoma: Report of two Cases and Review of the Literature

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Case 1

A 34 years old Saudi single female presented with an ulcerated left nipple of two years duration. Patient gave no history of associated co-morbidities. It was sudden in onset with no predisposing factors. She was a regular visitor to many dermatology clinics and history of chronic use of topical creams and ointments with brief relieve and recurrence once the treatment was discontinued. On clinical examination general examination revealed an anxious young female otherwise unremarkable. Local examination revealed an ulcerated bleeding nipple with obvious fissures. The remaining examination of both breast was within normal limits. Ultrasound was reported as normal. Wedge excision of the medial aspect of the nipple and areola was performed and was reported as Nipple Adenoma with no evidence of malignancy. Local treatments were not effective, patient refused the suggested nipple excision. Patient was followed up for three months and then lost follow up.

Case 2

A 43-year-old Saudi female with no reported comorbid conditions presented with thickened, cracked right nipple and bloody discharge of 4 years duration. It was sudden in onset associated with discomfort but no pain. She gave no history of palpable breast masses or other local or systemic symptoms. The patient noticed progressive thickening of the nipple associated with persistent ulceration not responding to local or systemic antibiotics and anti-inflammatory treatments. Family history for similar lesion or breast cancer is negative.

On examination: general examination was unremarkable. Local examination revealed an ulcerated flattened right nipple with bloody discharge from its surface. (Figure I) the surrounding was indurated with a small palpable retro areolar nodule measuring about 1x1 centimeter in diameter. There were palpable axillary lymph nodes could. Left breast was unremarkable. Paget's disease was the clinical diagnosis at presentation.

Mammography and Ultrasound revealed elements of duct ectasia bilaterally with no suspicious masses. Consistent with BI-RADS II findings. (Figure II)



Figure I: demonstrates the replacement of the nipple with a sharp margin ulcer with deformity of the nipple areolar complex denoting chronicity.

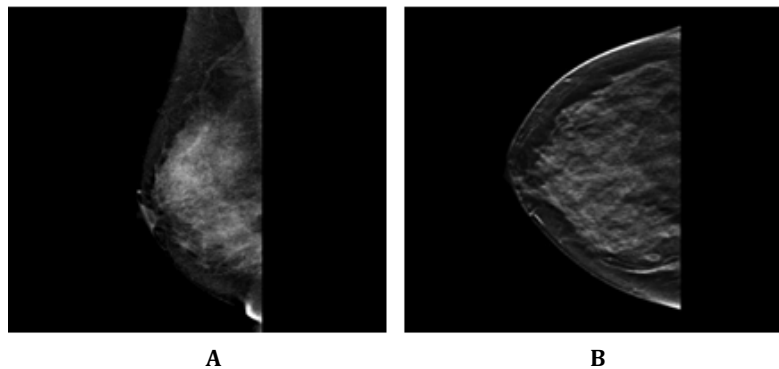


Figure II (a & b): The mediolateral and craniocaudal views demonstrate the normal glandular breast tissue with no evidence of suspicious lesions or retro-areolar architectural distortion.

In view of the chronic and persistent lesion and the suspicious of Paget's disease a wedge biopsy was performed under local anesthesia and was reported as proliferative epithelial lesion with features suggestive of nipple adenoma with no malignancy. The discrepancy between the clinical manifestation and the and histopathology report mandated another biopsy under general anesthesia and was reported as the same histopathological findings. Dermatology consultation and local treatment was again initiated with mild improvement of the symptoms. Patient underwent total nipple excision and pathology report revealed microscopic examination of the excised nipple shows a relatively well circumscribed lesion composed of compacted proliferative ducts and tubules in the dermis and nipple stroma. The overlying epidermis showed ulceration and granulation tissue formation. On higher magnification, the ducts showed a spectrum of hyperplastic changes ranging from mild papillary hyperplasia to florid ductal hyperplasia. The lining ductal cells lack cytological atypia. Calponin and p63 highlight the myoepithelial layer surrounding each duct. These features were diagnostic of nipple adenoma\florid papillomatosis of the nipple. (Figure III) At three months follow up the patient was satisfied with complete healing of postoperative site.

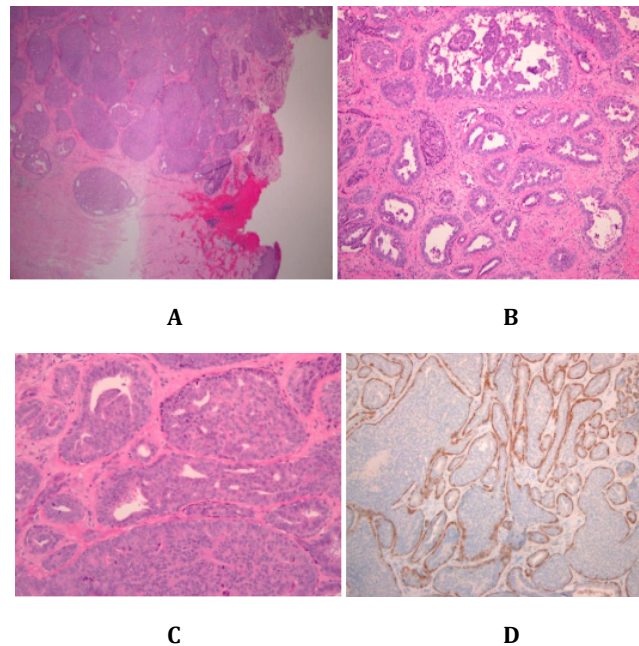


Figure III (a-d): Microscopic examination of the excised nipple. A. Low power view shows hyperplastic ducts and tubules in the dermis and nipple stroma with ulceration of the overlying dermis. B. Medium power view shows usual ductal papillary hyperplasia. C. Medium power view shows ducts with florid ductal hyperplasia along with slit-like secondary luminae. D. Calponin immunohistochemical stain demonstrates the presence of myoepithelial cells.

Discussion

Breast Cancer is commonest malignancy affecting women worldwide and is the second most common female in Saudi Arabia [1]. The concerns of breast cancer in any breast complaint dictate the thorough clinical evaluation and focused investigations of all symptomatic breast patients. Nipple adenoma is a rare condition which is usually missed and unrecognized by many breast care specialties. [2]

It is proven histologically that it is a benign ductal proliferative disease of middle aged women. Patients may present with a palpable nodule, or commonly presents as nonhealing ulcer characterized by erosion and crusting. [3] The age, chronicity, the presence of ulceration and nipple destruction mimic the clinical manifestations of paget's disease [4,5].

The standard imaging dependent on age groups, ultrasonography or mammography are the initial steps in diagnosis. The limitations of both modalities are noted due to the fact that nipple adenomas have the same density as the surrounding breast tissue with minimal architectural distortion. Few reports favored ultrasound despite its limited yield in detecting the echogenicity and hypervascularity which aid the diagnosis of adenoma [4,7,8].

Nipple adenomas are categorized under the benign ductal proliferative disease. Occurrence is in the middle age group with prolonged symptoms. [3]

The common presentation is non-healing ulcers, however a less commonly patients may present with breast masses. [9] Pathology may show inflammatory reaction and benign ductal epithelial cells on the excised wedge with repeated negative cytology. [10]

The benign pattern of nipple adenoma should not be taken for granted is not always the case. Reports had shown the close association of nipple adenoma with malignant breast conditions [11,12]. It has also been reported at rare sites such as in an accessory breast [6]. Nipple adenoma is not an exclusive female disease as it had been seldom reported in males. [10,13].

Unilaterality is the usual presentation of this entity however it may occur as bilateral disease. [11]. Nipple adenoma might mimic wide range of benign and potentially malignant conditions. Thus, Biopsy of the nipple lesion is an essential tool for primary diagnosis [6,13].

Histologic appearance of benign adenoma shows 2 layers of epithelial cells and myoepithelial cells which is the main component to differentiate it from malignancy [14].

Complete surgical excision of the nipple skin, underlying subcutaneous tissues and surrounding skin with histologic examination remains the gold standard for diagnosis and treatment [7,13,14].

However, others reported successful complete excision of the adenoma with preservation of the nipple [8,14]. The literature supported the good prognosis of this benign condition. However, the risk and relation to malignancy is still unknown and thus regular follow up is warranted [5,11,13].

These two rare presentations had posed challenges on both diagnosis and management. Biopsy followed by complete surgical excision of the nipple skin, underlying subcutaneous tissues and surrounding skin with histologic examination remains the gold standard for the management of nipple adenoma.

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