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Case Report

Syringomatous tumor of the nipple: master of deceit

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ABSTRACT

Syringomatous tumor (SyT) of the nipple is a locally infiltrative, benign neoplasm known to rarely involve breast. It usually presents as a unilateral retroareolar mass and may be associated with pain, nipple discharge or inversion. Microscopically tubules and ducts with comma-like configuration lined by double layered epithelium along with keratinizing cysts are seen similar to cutaneous adnexal neoplasm of eccrine origin. Since it shows infiltration into smooth muscle and nerves on microscopy, it is prone to be misdiagnosed as invasive carcinoma of breast. Initial management of SyT of the nipple requires complete resection of the lump with negative histological margins as this tumor is known to reoccur if incompletely excised. So, awareness of this entity is helpful when dealing with retroareolar breast lump clinically, radiologically and histologically. We present a case of SyT reported on a Tru-cut biopsy specimen in a 40 year old female presenting with retroareolar lump, for being a rare entity in breast and a diagnostic dilemma.

Keywords: Syringomatous tumor of the nipple, Syringomatous adenoma, Subareolar

INTRODUCTION

Syringomatous tumor (SyT) of the nipple/syringomatous adenoma was first described by Rosen in 1983 in his study of 5 patients as a benign, locally infiltrating tumor which showed histological similarities to syringoma of skin adnexal origin.[1] SyT of nipple tends to be infiltrative in nature thus notoriously reoccurs if excision is incomplete without negative histological margins.

Also, due to rarity in breast and similarity to other tumors on various fronts i.e. clinically, radiologically and histologically, it is often either under diagnosed or over diagnosed as invasive carcinoma resulting in overtreatment.[2]

We present a case of SyT of nipple reported on a trucut biopsy specimen from a 40-year-old female highlighting its clinical presentation, mammographic findings, histology and differential diagnosis to avoid misdiagnosis and provide adequate treatment to prevent relapses.

CASE REPORT

A 40-year-old female patient presented to the surgical OPD with complaint of awareness of lump in the nipple areola region of her left breast since 3 years. On palpation a non-tender, retro-areolar mass firm in consistency was identified. Overlying skin and nipple were normal. No complaints of nipple discharge, pruritis were present. Mammography revealed a well-defined irregular, dense mass measuring 18×41×45 mm^3 in nipple areola region of the left breast without any architectural distortion/calcification or raised vascularity and was graded as BIRADS 3 (Figure 1). Left nipple was not visualized separately from the mass radiologically. Tru-cut biopsy from the mass was done. Histopathology of the biopsy cores revealed densely fibrotic stroma and proliferation of elongated ductules and tubules with double layered epithelium infiltrating the muscle. Few comma shaped ducts were also seen (Figure 2-4). Diagnosis of SyT was given on histopathology and IHC for myoepithelial layer to rule out malignancy was suggested

as this facility was not available in our institute. Immunohistochemistry was positive for p63 and SMA, highlighting the myoepithelial layer. Patient refused excision of lump and is on follow up.

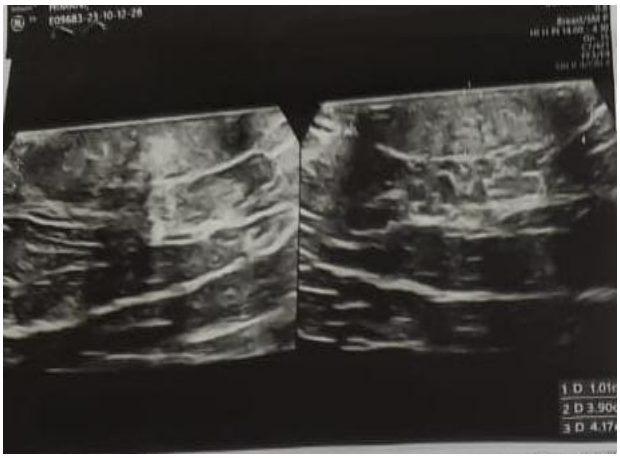

Figure 1: Mammography of the right breast. Oval iso-echoic mass in left nipple areolar region.

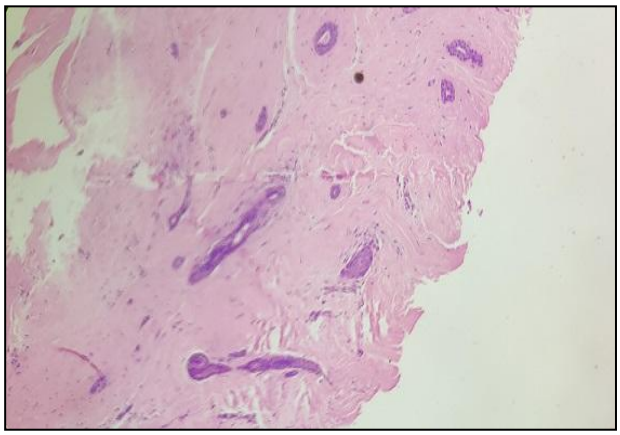

Figure 2: Proliferating ducts assuming a comma-shaped configuration lined by double-layer epithelium. (hematoxylin and eosin; 40×).

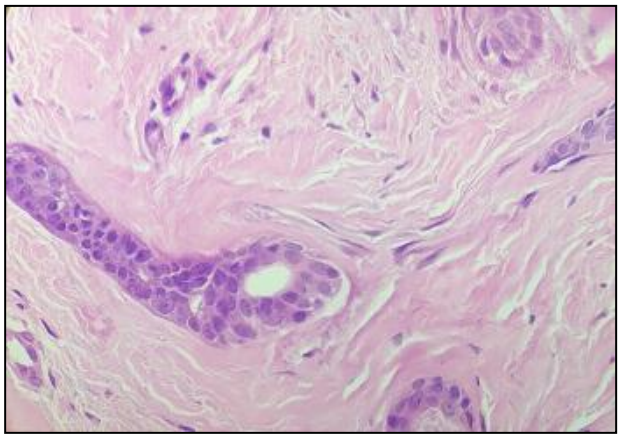

Figure 3: Proliferating ducts assuming a comma-shaped configuration lined by double-layer epithelium. (hematoxylin and eosin; 400×).

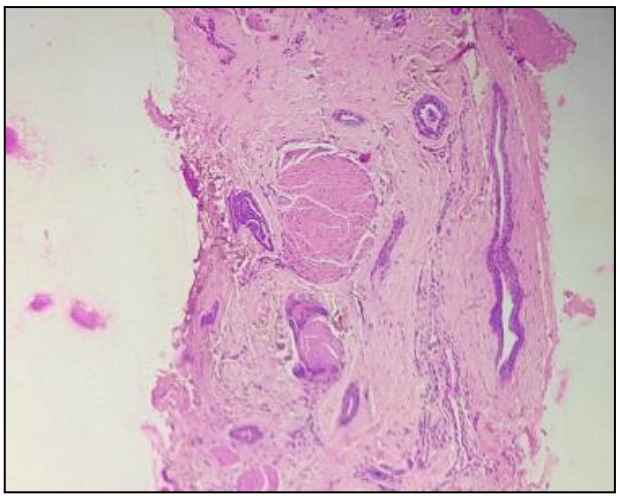

Figure 4: Tumor cells infiltrating the dense stroma between the smooth muscle bundles (hematoxylin and eosin; 100×).

DISCUSSION

SyT of the nipple is a rare, under diagnosed tumor in breast. It usually manifests as a firm, solitary mass in the subareolar region though it may occur in breast parenchyma also.[3] Occasionally it is known to present as fungating mass mimicing malignancy and is also known to involve bilateral breasts in extremely rare cases.[4] Nipple discharge, tenderness, pruritus and nipple retraction are other presenting symptoms.[3] It usually measures about 1 to 3 cm in size. Mean age at the time of presentation is 40 years as was the current scenario. Commonly affecting females, occasionally SyT involves male breast and supernumerary nipple.[5-7] SyT is visualized as high-density mass with irregular margins and microcalcification on mammography while on ultrasonography it appears as an ill-defined mass with heterogeneous echotexture.[8] These radiographic findings are not very helpful so as to differentiate SyT from malignant mass. Histologically SyT reveals tubules and ductules with comma shaped structures that infiltrate smooth muscle bundles. Keratinous cysts like structures are also seen.[1] Immunohistochemistry demonstrates intact myoepithelium.[9]

Since SyT shares many nonspecific features with other types of neoplasm morphologically leading to incorrect diagnosis, affecting" with incorrect diagnosis will affect the patient management and outcome. Nipple adenoma, seen in females in their fourth decade is a benign epithelial hyperplasia of the lactiferous duct of nipple and has clinical presentation similar to SyT of nipple with nipple erosion and deformity. Thus it is difficult to differentiate nipple adenoma from SyT unless clinical, radiological and morphological features are studied in unison.[10] Tubular carcinoma underlying nipple areola region and SyT of nipple both infiltrate smooth muscle and have similar morphology but tubular carcinoma tends to occur deeper in the breast parenchyma, often has accompanying foci of DCIS, shows pagetoid disease and has absent myoepithelial layer.[11] Certain variants of low-grade

adenosquamous carcinoma involving nipple are also difficult to differentiate from SyT however they are known to occur deep in breast parenchyma unlike SyT.[12] Imaging modalities like Mammography, ultrasonography, and MRI render little help to differentiate from other close morphological entities. Management of SyT includes complete resection of tumor with histologically negative margins to avoid local tumor recurrence.[11]

CONCLUSION

The diagnosis of SyT is a challenge not only clinically but radiologically and microscopically, especially on needle biopsy done preoperatively both due to its rarity and morphological mimics. Therefore, in the setting of a retroareolar lump prior awareness of various morphological entities including SyT of nipple is necessary for an accurate diagnosis and better patient outcome.

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