

CASE REPORT

Syngomatous adenoma of nipple; A case report and review

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Abstract:

Syngomatous adenoma is a benign, rare but locally infiltrative tumor¹. It is very difficult clinically to differentiate between syngomatous adenoma and malignant conditions². Clinically, it presents as sub-areolar lesion but ultrasound and mammography show features mimicking malignancy³. Therefore it is important to have proper pre-operative diagnosis in the form of FNAC and tru-cut biopsy to diagnose malignancy. In our case we did tru-cut biopsy of the peri-areolar region which revealed no malignancy. Excision biopsy of the lesion done and undue mastectomy was avoided.

We represent a case report of 35 years old multiparous lady who presented with history of painless swelling in retro-areolar region of right breast for 7 years. clinically proven as syngomatous adenoma of nipple and literature review

Keywords: sub-areolar lesion of breast, tru-cut biopsy, FNAC (fine needle aspiration cytology), locally infiltrative tumor, Syngomatous adenoma, infiltrative ductus carcinoma breast, infiltrative lobular carcinoma of breast.

Introduction:

A 35 year old multiparous lady presented with history of painless swelling in retro-areolar region of right breast for 7 years. She did not notice any progression in size and shape of lump since it was first noticed. No history of blood or any other discharge from nipple. There was no past history of breast disease and no risk factors of breast malignancy in this patient.

On examination of right breast, there was nipple retraction with thick nipple areolar complex. No nipple ulceration or discharge.(Fig.1) On palpation, 2×2 cm hard, non-tender mass was palpable in retro-areolar region. No regional lymph-adenopathy. Left breast was normal. Mammography revealed irregular dense mass in retro-areolar region with dendritic margins; no enlarged axillary lymph nodes were seen in axilla. It was labeled as BI-RADs category IV (fig 2, 3).

Tru-cut biopsy revealed no malignant cells. Ex-

cision biopsy of retro-areolar lump was done which confirmed the diagnosis of syngomatous adenoma of nipple. Subsequently Wide excision of lump was done (Fig. 4) which showed that the lesion has been completely excised, closest margin was 0.3 cm away from tumor and the histopathological features reconfirmed the diagnosis of syngomatous adenoma. (Fig.5)

Lesion composed of well-formed tubules with lumen formation. These tubules are scattered in streaming fashion and are set in a sclerotic stroma. These tubules are lined by double layer of epithelium.

Basal myoepithelial layer is highlighted on P63 immunohistochemical stain. Luminal epithelium is composed of round cells with scant cytoplasm. Some ducts appear compressed.

Discussion:

Syngomatous adenoma of nipple is a benign, rare locally invasive tumor of nipple areolar re-

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Figure 1:



Figure 4:

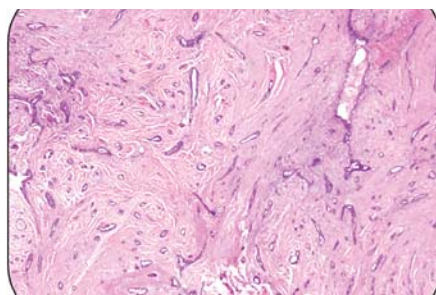


Figure 5:

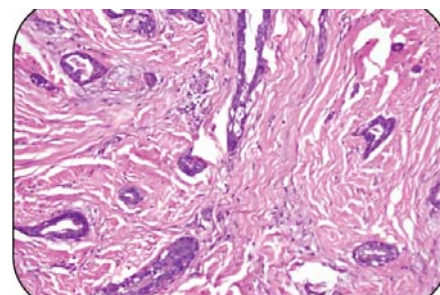


Figure 6:

gion which does not metastasize, show sweat gland duct differentiation. It is locally recurrent.⁵ In 1983; it was first described by Rosen.⁶

The term “Infiltrating” was given in 1989, in a clinical and pathological study of 11 cases to emphasize locally infiltrative nature of syringomatous adenoma.⁷

The reported age of patients with syringomatous adenoma is 11-76 years with an average age of 40 years.^{6,7} Our patient also falls in this range. Common clinical presentation is single, painful nipple swelling which may lead to nipple discharge, itching or inversion of nipple.⁶⁻⁸

Medical literature has reported that 65-70% patients commonly presented with serous or bloody discharge from nipple which is followed by nipple enlargement and induration associated with nipple ulceration.⁹⁻¹¹ Our patient presented with retro areola mass radiologically presented as a subareolar, spiculated mass that cannot be differentiated from carcinoma. Mammogram in our patient showed BI-RAD category IV. No radiological features are characteristic to this lesion.^{12,13} Grossly, syringomatous adenoma is typically a dermal nodule in a sub-areolar region.¹⁴ It was described as a tumor like proliferation of small squamous islands in which there is lack of

connection between surrounding parenchyma of breast and squamous structures.¹⁵ Probably, it develops from pluripotential cells i.e. adnexal keratinocyte which has a capability to differentiate into follicular and as well as sweat gland.¹⁶ For diagnosis of this rare tumor, histopathology proof is mandatory.¹⁷ It is necessary to differentiate syringomatous adenoma from low grade adenosquamous carcinoma and tubular carcinoma especially when nipple areolar complex is involved.¹⁸ Toyoshima et.al reported a case in which probable low grade adenosquamous carcinoma was misinterpreted in FNAC that was followed by mastectomy and dissection of axilla but, syringomatous adenoma was revealed in microscopic sections.¹⁹ In 1987, another case reported, in which frozen section biopsy was misinterpreted. It revealed features of Paget’s disease that resulted into mastectomy.²⁰

Surgical management ranges from excisional biopsy to radical mastectomy.²¹ 25-55% of patients had recurrence which was initially treated with some form of local excision or subcutaneous mastectomy.⁷ Local recurrence can occur in incomplete excision.²² It had not been found when aggressive local therapy was given to patients; which includes complete excision of nipple or mastectomy.¹⁶ Therefore to avoid it, treatment of

choice is complete excision. After confirmation of complete excision histologically, nipple reconstruction is usually planned.¹⁷ However, no proof of metastasis has been found.¹⁶ It is proved from literature that patients with this adenoma who had undergone axillary dissection, axillary lymphadenopathy has not been found. During follow up no patients reported to have distant metastasis as long as 24 years follow up.⁷

Role and contribution of authors:

Dr Lubna Habib, Professor of Surgery at Hamdard University Hospital, wrote the primary case report and clinical research of the case report.

Dr Anum Ikram Khan is FCPS fellowship resident at Hamdard University Hospital, collected the data, gathered the references for syringomatous adenoma and help the author in writing down the case report.

Conflict of Interest: none

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