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## Case Report

# Asymptomatic syringomatous adenoma of the nipple: A rare nipple neoplasm<sup>☆</sup>

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## ABSTRACT

Syringomatous adenoma of the nipple is a very rare benign tumor, with radiographic and clinical characteristics that are difficult to differentiate from malignant tumors. Less than 60 cases have been identified thus far and most studies have not included radiographic findings. As such, the neoplasm requires more characterization within the literature. The usual clinical presentation of syringomatous adenoma of the nipple is symptomatic with a solitary, unilateral nipple mass within the subareolar region which may be tender and/or painful and may cause nipple inversion or discharge. We present a case of a 43-year-old woman that is unique, as the patient was asymptomatic and only presented following standard screening mammography. Moreover, we offer further characterization of the neoplasm through documented imaging and histologic findings.

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## Introduction

Syringomatous adenoma of the nipple (SAN) is a rare and benign tumor, the exact etiology of which is unknown, but is hypothesized to originate from sweat glands within the nipple-areolar complex [1]. It was first described by Rosen in 1983 [1]. The mean age of presentation is 46 years of age, and the tumor typically ranges in size from 1 to 3 cm [2]. SAN is typically unilateral and tends to show local infiltration without metastasis [3,4]. It can be misdiagnosed as carcinoma due to the infil-

trating pattern and requires careful pathologic examination to differentiate between them [2]. Standard treatment includes wide excision of the tumor [4]. Without complete resection, the tumor has a recurrence rate of 25%-55% [5]. Patients typically present with an enlarging palpable mass, change in the appearance of the nipple, nipple discharge, or breast pain, and some have been discovered after rapid increase in size after pregnancy [3–11].

We present a case of an asymptomatic 43-year-old woman that presented for screening mammography, which revealed microcalcifications in the left nipple that were eventually confirmed to be SAN.

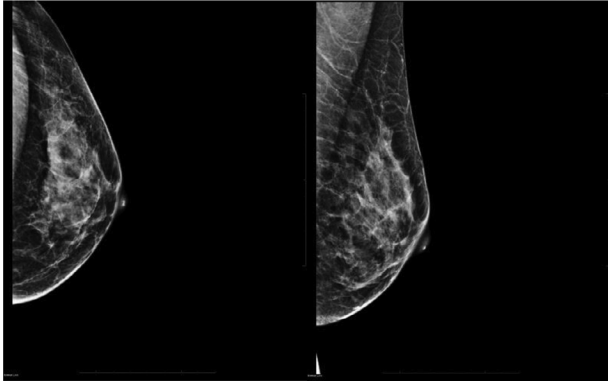
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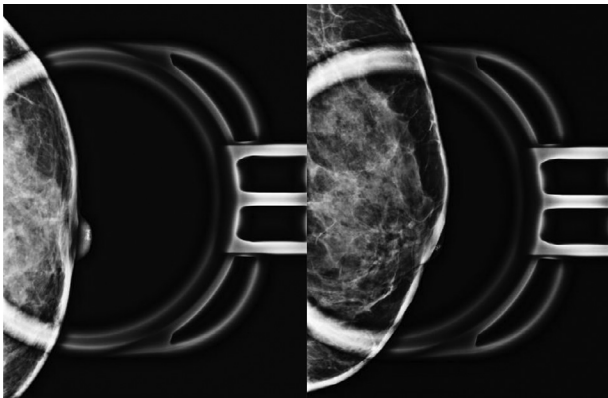
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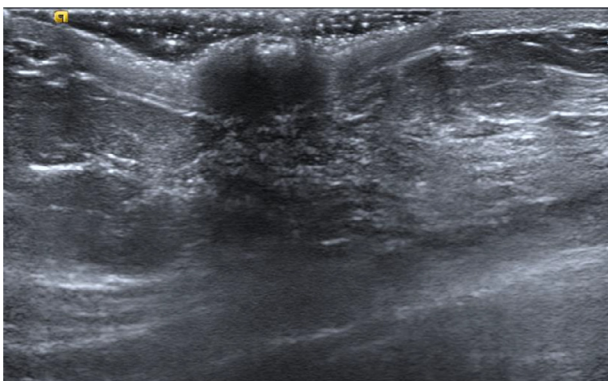
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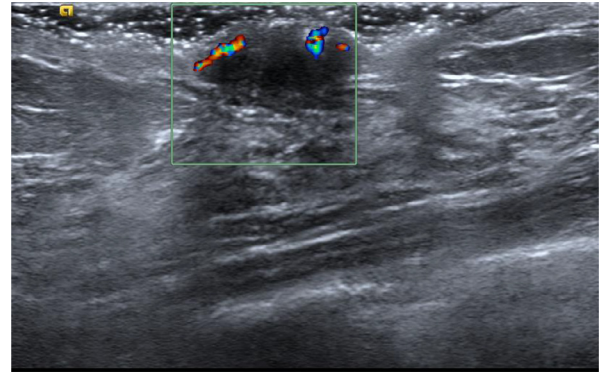
**Fig. 1 – Screening mammogram. Craniocaudal (left) and mediolateral oblique (right) views of the left breast demonstrating heterogeneously dense breast with microcalcifications in the nipple.**



**Fig. 2 – Diagnostic mammogram. Magnification views of the left breast. Craniocaudal (left) and mediolateral (right) views demonstrating a group of fine pleomorphic calcifications within the nipple.**



**Fig. 3 – Static gray scale ultrasound of the left retroareolar breast demonstrating echogenic foci within the left nipple, which are consistent with microcalcifications.**



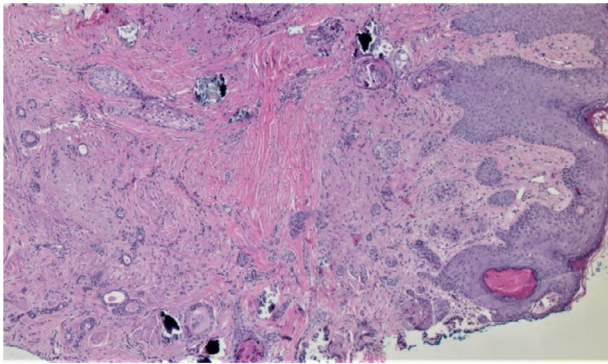
**Fig. 4 – Static doppler ultrasound image of the left retroareolar breast demonstrating increased vascularity of the left nipple.**

### Case

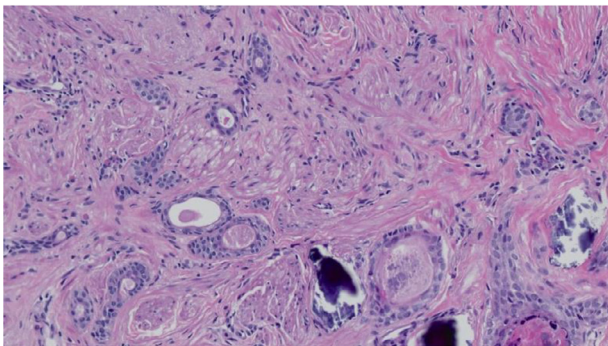
The patient was a 43-year-old woman with a history of right breast phyllodes tumor status post excisional biopsy who presented for screening mammography, which demonstrated calcifications in the left nipple (Fig. 1). The patient was recalled from screening mammography for a diagnostic mammogram and ultrasound.

Diagnostic mammogram demonstrated fine pleomorphic microcalcifications in the left nipple spanning 5 mm (Fig. 2). Further examination with ultrasound also revealed evidence of microcalcifications as well as increased vascularity of the left nipple (Figs. 3 and 4). No suspicious left breast retroareolar masses were identified. The diagnostic mammogram and ultrasound were given a BI-RADS 4 classification with recommendation for surgical consultation, as stereotactic needle biopsy could not be performed because of the location of the microcalcifications within the nipple.

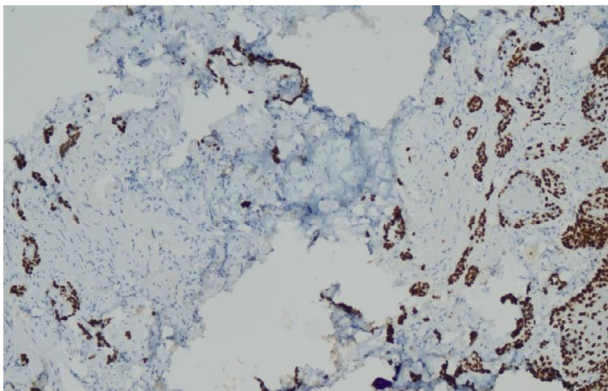
The patient was referred for surgical consultation as well as nipple punch biopsy. Physical examination of the breasts was conducted in the sitting and supine positions and found no asymmetries other than a well healed right breast periareolar scar from previous excisional biopsy. The exam revealed no dominant masses, nipple inversion or discharge, nor any associated skin dimpling. Furthermore, the patient was not found to have any axillary, supraclavicular, or cervical lymphadenopathy. She denied any palpable masses bilaterally, nipple discharge or skin changes prior to the exam. Needle punch biopsy of the nipple shows a 5 mm tumor. Histologically, the tumor consists of an infiltrating bland small ductular proliferation lined by double layer of epithelial cells. The outer layer comprised of small, cuboidal cells with scanty cytoplasm while the inner layer consisted of cuboidal to flat cells with eosinophilic cytoplasm. In some of the lumens there are amorphous eosinophilic material in addition to calcifications. In addition, squamous metaplasia of the tubules and keratinizing cysts, also containing eosinophilic debris, are found scattered throughout. The tumor cells infiltrated the stroma in between the smooth muscle bundles (Fig. 5).



A.



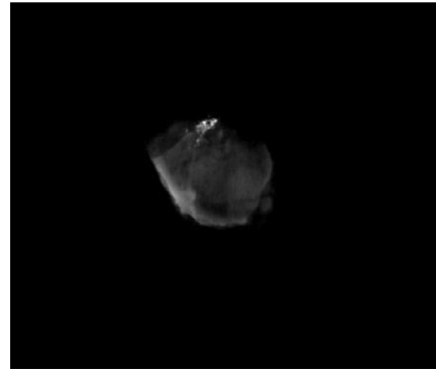
B.



C.

**Fig. 5 – (A) Low power 5X, H&E stain, infiltrating cytologically bland proliferation of small ducts, and tubules in between the smooth muscle bundles. (B) High power 20X, H&E stain, the ducts, and tubules are lined by double layer of epithelial cells. The lumens contain eosinophilic amorphous material. Squamous metaplasia and keratinizing cysts also with eosinophilic debris and calcifications are present. (C) Low power 5X, Immunohistochemistry stain p63, highlighting the myoepithelial cells, and squamous metaplasia.**

Following the punch biopsy, the breast surgeon performed wide local excision of the left nipple (Fig. 6). Final diagnosis was described as dermal scar with a 0.8 mm focus of residual syringomatous nipple adenoma, which was 3 mm from the nearest inked margin of the specimen.



**Fig. 6 – Plain radiograph of the left nipple excisional biopsy specimen demonstrating fine, pleomorphic calcifications.**

## Discussion

Syringomatous adenoma of the nipple is a very rare benign tumor, with radiographic and clinical characteristics that are difficult to differentiate from malignant tumors [5]. Age at diagnosis ranges from 11 to 87 years with a mean age of presentation of 46 years [3]. Tumor size typically falls between 1 and 3 cm with the largest tumor documented in the literature measuring 5 cm [6]. Less than 60 cases have been identified thus far, with only 2 of them affecting males [4,5]. Most studies have not included radiographic findings, thus requiring more characterization.

The majority of patients have a symptomatic presentation. The usual clinical presentation of syringomatous adenoma of the nipple is typically a solitary, unilateral nipple mass within the subareolar region which may be tender and/or painful, and may cause nipple inversion or discharge [12]. Our case is unique, as the patient was asymptomatic, and only presented following standard screening mammography. We also offer further characterization of the neoplasm through documented imaging findings.

Reported mammographic features include a high-density retroareolar mass that tends to be irregular with possible spiculations and/or microcalcifications [3]. The morphology of the microcalcifications in the nipple-areolar complex are often characterized as amorphous and fine pleomorphic in a regional or focal distribution [5]. Ultrasonography may demonstrate an ill-defined mass with heterogeneous internal echoes as well as skin thickening or retraction [3,5]. This case is unique because imaging did not demonstrate any masses and only showed a small group of microcalcifications. Moreover, a novel finding in our case was the presence of left nipple increased vascularity on doppler ultrasound, which has not been previously characterized. Our patient also did not have any associated skin changes, a common characteristic of this neoplasm.

SAN may be indistinguishable from carcinoma on imaging because findings tend to resemble those of malignant tumors: masses with suspicious morphology and microcalcifications [3]. As such, SAN needs to be diagnosed through histologic examination. Differential diagnosis on histology includes tubu-

lar and nipple adenoma, ductal carcinoma in situ, Paget's disease of the nipple, intraductal papilloma, and low-grade adenosquamous carcinoma [3]. Histologic features typical of syringomatous adenoma of the nipple often demonstrate focally infiltrative irregular compressed ducts and tubules involving the surrounding deep dermis smooth muscle bundles and stroma of the nipple. This tumor consists of infiltrating yet cytologically bland round to irregular tubules and ducts, often compressed and comma-shaped or tadpole-shaped in appearance, helping to differentiate them from other lesions [5]. The lumens of the ducts and tubules often contain amorphous, eosinophilic material and very rarely associate with calcifications. Squamous differentiation of the compressed tubules is frequently seen.

Management of SAN consists of complete local excision with histologically negative margins. Incomplete resection of the lesion may lead to recurrence [2]. In the literature, there was no documented evidence of recurrence during a follow-up period of 6 years in patients having negative margins after resection [2]. Regarding our case, negative margins were achieved with the tumor being 3 mm from the nearest inked margin of the specimen.

## Conclusion

We report the case of a 43-year-old woman who presented asymptotically for screening mammography, which revealed left nipple microcalcifications. Diagnostic mammogram confirmed the fine pleomorphic nipple microcalcifications and revealed no other lesions. The tumor was not amenable to stereotactic needle biopsy, requiring a nipple punch biopsy. Pathologic examination revealed the diagnosis of SAN with subsequent excisional biopsy confirming the diagnosis. The absence of symptoms or mass as well as the increased vascularity are an unusual presentation of SAN. Follow-up will be conducted to assure no recurrence.

## Patient consent

Written, informed consent for publication of case was obtained from the patient.

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