









Syringomatous Adenoma of the Nipple on Screening Mammography: A Case Report

선별 유방촬영술에서 발견된 유두의 한선 종양: 증례 보고

Min Hyeok Yoon, MD¹ , Hye Jung Kim, MD^{2*} , Won Hwa Kim, MD² ,
Jeeyeon Lee, MD³ , Ji-Young Park, MD⁴ , Jin Young Kim, MD⁵ 

¹Department of Radiology, School of Medicine, Kyungpook National University, Kyungpook National University Hospital, Daegu, Korea
Departments of ²Radiology, ³Surgery, and ⁴Pathology, School of Medicine, Kyungpook National University, Kyungpook National University Chilgok Hospital, Daegu, Korea
⁵Kim Jin Young Breast & Thyroid Clinic, Daegu, Korea

Syringomatous adenoma of the nipples, first reported in 1983, is an extremely rare benign tumor extending to the subareolar area and, pathologically, has a shape similar to that of sweat gland tumors. Radiologically, infiltrating patterns and calcifications can cause misdiagnosis of malignant tumors. The authors report a case of syringomatous adenoma that shows only calcifications of the nipple in a screening mammography.

Index terms Syringoma; Adenoma; Nipples; Mammography

INTRODUCTION

Syringomatous adenoma of the nipple (SAN) is an extremely rare benign tumor extending to the subareolar region (1). Most of the patients have a unilateral palpable mass in the nipple, pain and discharge (1-3). However, a review of literature showed microcalcifications are uncommon (4). We report the case of a 42-year-old female with SAN showing only microcalcifications.

CASE REPORT

This study was approved by the Institutional Review Board of our hospital and the requirement for informed consent was waived (IRB No. 2021-05-040).

A 42-year-old female visited the local clinic for a breast cancer screening examina-

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***Corresponding author**

Hye Jung Kim, MD
Department of Radiology,
School of Medicine,
Kyungpook National University,
Kyungpook National University
Chilgok Hospital,
807 Hoguk-ro, Buk-gu,
Daegu 41404, Korea.

Tel 82-53-200-3376
Fax 82-53-200-3349
E-mail mamrad@knu.ac.kr

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ORCID iDs

Min Hyeok Yoon 
<https://orcid.org/0000-0002-9997-3528>
Hye Jung Kim 
<https://orcid.org/0000-0002-0263-0941>
Won Hwa Kim 
<https://orcid.org/0000-0001-7137-9968>
Jeeyeon Lee 
<https://orcid.org/0000-0003-1826-1690>
Ji-Young Park 
<https://orcid.org/0000-0002-7571-1064>
Jin Young Kim 
<https://orcid.org/0000-0002-9225-8206>

tion. She does not have any breast symptoms, such as palpable lump, nipple discharge, or pain. She had no other history of breast disease or family history of breast cancer. Mammography (MMG) was performed, which showed grouped coarse heterogeneous microcalcifications in the left nipple without associated suspicious masses, abnormal lymph nodes, or architectural distortions (Fig. 1A). Ultrasonography (USG) was conducted subsequently. The examination revealed several microcalcifications in the left nipple (Fig. 1B).

The patient visited our hospital for further evaluation of the left nipple. Incisional biopsy was performed resulting in mild acanthosis with perivascular lymphocytic infiltration. Craniocaudal and mediolateral magnification view showed no change of the microcalcifications of the left nipple on 3 months follow-up. An excisional biopsy was performed to rule out the possibility of malignancy and to reduce the difficulty in localizing all calcification of the nipple. The specimen contained the whole of the left nipple and was diagnosed as syringomatous adenoma with complete excision. Syringomatous adenoma was located in the dermis of the nipple and infiltrated around lactiferous sinuses and into the areolar smooth muscle. Cysts and dilated lactiferous sinuses containing keratin materials were noted in the superficial portion with multiple dystrophic calcifications. The histologic section of the tumor showed a configuration composed of comma-shaped ductal structures and tubules in the background of fibrotic stroma (Fig. 1C).

DISCUSSION

SAN is an extremely rare tumor that was initially described in 1983 by Rosen (1). Although the term “infiltrating syringomatous adenoma” was used when the tumor locally extends to the subareolar region with the potential of recurrence, SAN is benign because it does not metastasize and is curable with complete excision of the lesion (5). The age of patients with SAN ranged from 11 to 76-years-old, and the most affected age group is the 40s (6). A majority of the patients have variable clinical symptoms such as a unilateral palpable mass in the nipple and

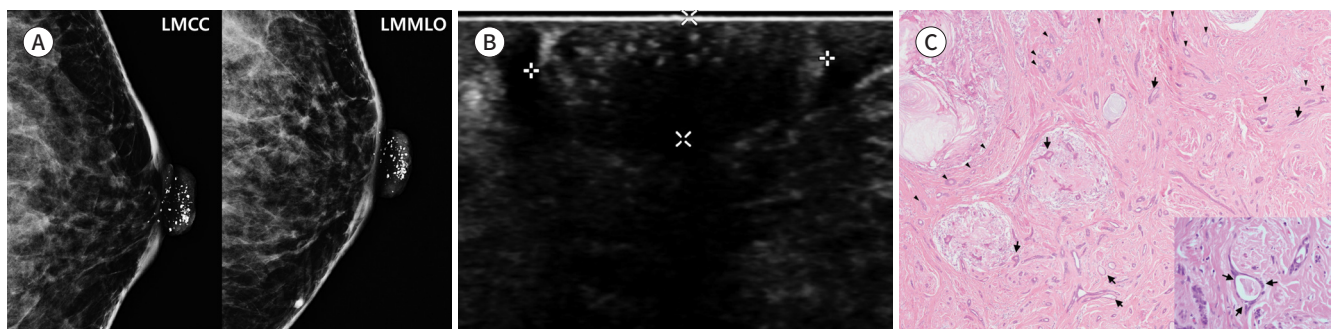
Fig. 1. A 42-year-old female patient with syringomatous adenoma of the left nipple.

A. Grouped, coarse, heterogeneous microcalcifications are seen in the left nipple on the craniocaudal (left) and mediolateral oblique (right) magnification views.

B. Ultrasonography shows microcalcifications in the left nipple (markers indicate the nipple).

C. The lesion showed an infiltrative proliferation of the small glandular and tubular structures, often angulated features with a tear-drop shape or a comma shape (inset, arrows, $\times 400$; H&E stain). The glands were composed of two cell layers—the inner epithelial cells (arrows) and the outer flattened myoepithelial cells (arrowheads) ($\times 40$, H&E stain).

H&E = hematoxylin and eosin, LMCC = left magnified craniocaudal, LMMLO = left magnified mediolateral oblique



subareolar region, tenderness with swelling, nipple discharge, and inversion (1-3). The patient in this case study is 42-years-old as well, but these symptoms and abnormalities were absent during the physical examination.

Histologically, the compositions of SAN are tubules and ducts, which is lined by double or multiple metaplastic squamous cells, in a fibrous stroma. Comma- or tadpole-shaped nests exist in these proliferating ducts which have a syringomatous appearance. Keratotic cysts found in the superficial dermis are another feature of the tumor. The tumor cells are thought to be proliferated from pluripotential keratinocytes and infiltrate smooth muscle bundle and even perineural regions (7).

The typical radiologic findings upon MMG is a subareolar mass with high density and irregular shape or spiculated margin, but some cases without any radiological features also exist (3). Microcalcifications with or without mass are uncommon (4). USG findings include irregular or circumscribed mass in the subareolar region with or without skin thickening (3, 6, 8, 9). Both MMG and USG in this study revealed only grouped microcalcifications in the nipple. This appearance of the lesion requires to distinguish SAN from other conditions or diseases having similar calcifications.

Some situations in the nipple that may be suspected of being benign that are accompanied by calcifications may include skin calcifications, fat necrosis, suture site calcification, and intraductal papilloma. Skin calcification, originating from the gland and hair follicle, can be extensive and have a spherical pattern with a central lucent area. Fat necrosis can occur if the patient has undergone trauma or previous surgery or radiation treatment. Thus, an investigation of the patient's history is required. Radiologic findings include a rim-like or eggshell calcification accompanied by an oil cyst. Sutural calcification may occur after undergoing surgical biopsies. MMG can show nipple calcifications in the case of papilloma in which it shows rounded, crescentic, coarse, or eggshell form. Sixty percent of the patients have normal MMG because papilloma is relatively small (4).

Paget's disease is a malignant disease that can be first considered in situations where nipple calcifications are shown. Approximately 90% of the patients with Paget's disease have concurrent malignant diseases. Thus, differentiating SAN from Paget's disease is essential. Typical clinical manifestations are eczema, scale skin, retraction or erosion of the nipple, and bloody discharge from the nipple. MMG and USG may also show nipple-areolar complex-related abnormalities, suspicious mass, and malignant calcifications with segmental distribution (4).

Although there are several different characteristics between SAN and other diseases, a biopsy is mandatory for patients to establish appropriate management especially when radiologic findings are suspicious and not specific to eliminate other diseases or when it is too early to identify clinical courses. The subject in this case study has only suspicious calcifications in the nipple without a suspicious mass, the biopsy was needed to exclude Paget's disease. However, SAN does not involve the epidermis, an inadequate sampling of dermis can produce a limited diagnosis (10).

The treatment of SAN varies from local excision to total mastectomy, and the recurrence rate was 25%–55% when incomplete excision was performed (1, 2). The first consideration is to reduce the recurrence rate while avoiding aggressive surgical treatment by doing a com-

plete excision like in the patient of this case report.

In conclusion, SAN can show suspicious microcalcifications without mass in the nipple on MMG. Therefore, it is important to be aware of this mammographic SAN finding to prevent more aggressive surgical treatment.

Author Contributions

Conceptualization, Y.M.H., K.H.J.; resources, L.J., P.J., K.J.Y.; supervision, K.H.J.; writing—original draft, Y.M.H.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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선별 유방촬영술에서 발견된 유두의 한선 종양: 증례 보고

윤민혁¹ · 김혜정^{2*} · 김원화² · 이지연³ · 박지영⁴ · 김진영⁵

유두에 발생하는 한선 종양은 1983년 처음 보고된 매우 드문 양성 종양으로 유륜 하부에서 발생하며 병리학적으로 땀샘종과 닮은 형태를 보이는 특징이 있다. 영상 소견에서 침윤하는 양상과 석회화로 인해 악성 종양으로 오진하는 경우가 있다. 저자들은 선별 유방촬영술에서 유두의 석회화만을 나타내는 한선 종양의 증례를 보고하고자 한다.

¹경북대학교 의과대학 경북대학교병원 영상의학과,
²경북대학교 의과대학 칠곡경북대학교병원 영상의학과, ³외과, ⁴병리과,
⁵김진영유외과의원