INTRODUCTION

Pseudoangiomatous stromal hyperplasia (PASH), first described by Vuitch et al., is a rare benign lesion, shows the proliferation of the breast stromal tissue mimicking the low grade angiosarcoma (1-7). The most common mammographic and ultrasound finding of PASH is a circumscribed mass without calcification and it is difficult to distinguish from the phyllodes tumor and fibroadenoma (1-4, 8). To our knowledge, PASH presenting as rapid bilateral breast enlargement, as seen in our case, is very rare. In addition, several English medical literature were reported in this kind of manifestation of PASH (3, 4, 8).

We described imaging findings of diffuse, infiltrating, and bilateral manifestation of PASH.

CASE REPORT

A 19-year-old female had massive, progressive enlargement of both breasts for eight months. Although there was a history of dull ache in both breasts, breast enlargement was not associated with nipple discharge, fever, weight loss or any other clinical symptoms. She was nulliparous and had no prior history of breast disease or trauma.

Physical examination revealed markedly enlarged breasts, measuring over 30 cm in the transverse and craniocaudal dimension of each breast. Both breasts were firm in consistency and movable without any discrete palpable mass. The overlying skin appeared red, and tense with engorged superficial vein. Skin ulceration was not present. There was no evidence of axillary or supraclavicular lymphadenopathy. Routine hematologic and biochemical examinations were within normal limits.

Due to huge size of both breasts, mammography could not be evaluated. On ultrasound, a diffuse, infiltrating and heterogeneous hypoechoic lesion was present with difficulty in delineating the normal architecture. Ill-defined tubular shaped cystic areas were scattered within this lesion without vascular flow (Fig. 1).

The core needle biopsy was done to eliminate the possibility of malignancy.
of malignancy. The result of biopsy suggested a possibility of phyllodes tumor. Therefore, further work up was performed for surgical excision and detecting malignant features, such as invasion, or metastatic lesion.

CT and MRI scans revealed markedly enlarged breast parenchyma without discrete mass. Normal breast parenchyma couldn't be delineated from this lesion. Both enlarged breasts appeared not to extend or invade into the chest wall muscles on CT and MRI, which suggested benign lesion rather than malignancy. No area of necrosis or hemorrhage was present on both CT scans and MR images. There was no evidence of enlarged lymph node in the axillary or supraclavicular area. On CT scans, both breasts showed homogeneous soft tissue density and a few fat densities were scattered throughout the whole breast. No calcification was detected on CT scans. On MR, both breasts showed homogeneously low intensity on T1 as compared with the muscle and heterogeneously high signal intensity on T2 weighted images, respectively. Both breasts showed a mild, heterogeneous enhancement on fat suppressed and enhanced images (Fig. 1).

The patient underwent bilateral simple mastectomy and mammoplasty. The final right-sided excised lump measured $18 \times 16 \times 15$ cm, and weighed 1200 grams. The left-sided lump measured $22 \times 17 \times 11$ cm and weighed 1200 grams. The microscopic examination showed proliferation of the stromal components. There was a proliferation of stroma with diffuse slit-like network of spaces, looking like vascular channels in the collagen stroma. No red blood cell was seen in this structure. Immunohistochemistry was positive for CD34. Because no leaf-like growth pattern was noted, the phyllodes tumor has been ruled out. Also, cytological atypia or mitosis was absent. These features are regarded as being suitable for PASH (Fig. 1).

The patient is on regular follow up without evidence of recurrence.

**DISCUSSION**

PASH is a benign stromal proliferation first described by Vuitch et al. Histologically, its most common feature is a complex pattern of the vascular channel, like a space lined by myofibroblasts, without containing red blood cells, and within the stro-
enhancement pattern (4, 8, 9). Although it is a very rare manifestation of PASH, it should be considered to be one of the differential diagnoses of massive bilateral breast enlargement in young women.

REFERENCES


Immunohistochemically, PASH are known to be positive for CD34 and negative for vascular markers CD31 and factor VIII-related antigen, so it can be distinguished from angiosarcoma (4-6, 8). PASH mainly occurred in young premenopausal women; in some cases, it also occurred in postmenopausal women in hormonal replacement therapy. This suggests that the development of PASH may be related with hormonal stimulation (1, 3, 4, 7-9). It is found as an incidental finding of breast biopsy specimens, up to 25% and can occur as mass forming tumor. PASH is known not to progress to malignancy, but it can be present within malignant lesions. However, some reports also had produced the possibility of recurrence. As such, final diagnosis by core needle biopsy alone was not recommended (1-6, 8).

Imaging findings of PASH are non specific. The most common mammographic finding of PASH is a circumscribed mass without calcification, followed by increased stromal density. Some reports were suggesting that PASH could show as slow-glowing asymmetry on mammography, although, some PASH lesions cannot be detected on mammography. The case is shown as a well-defined mass, can mimick the phyllodes tumor and the fibroadenoma. The size of PASH lesions varies, ranged from a few millimeters to a huge mass (1, 3, 4, 8).

On ultrasound, it is mainly presented as a hypoechoic mass, also mimicking fibroadenoma or phyllodes tumor. It usually are associated with tubular or round shaped cystic space (4, 8). Celliers et al. (2) reported the normal ultrasound as a second common finding. During pregnancy, PASH lesions can occur as a breast mass, causing breast enlargement with skin necrosis (1). Manifestation of PASH woman as rapid bilateral breast enlargement is very rare. Further, a few English medical literature were reported in this kind of manifestation of PASH (3, 4, 8).

On MRI, which had been reported less frequently, PASH showed intermediate or low signal intensity on T1 weighted images in reference of the muscle, as our case. Low signal intensity mixed with high signal intensity is a common finding on T2 weighted images. High signal intensity area is thought to be a cystic space on ultrasound images. After contrast injection, PASH was known to show avid enhancement with benign, persistent enhancement pattern (4, 8, 9).

Although it is a very rare manifestation of PASH, it should be considered to be one of the differential diagnoses of massive bilateral breast enlargement in young women.
양측성 가성혈관종성 기질 증식증: 증례 보고

고희선 · 제수경

양측성 유방통과 유방 확대를 주소로 내원한 19세 여자 환자에서, 유방 초음파, 전산화단층촬영, 자기공명영상 시행하였다. 시행한 영상소견상 경계가 불분명한 종괴양의 병변이 정상 유방 조직과 서로 섞여 있는 양상을 보였으며 수술 후 병리조직 검사상, 양측성 가성혈관종성 기질 증식증으로 확인되어 증례 보고를 하고자 한다.

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