



Spontaneous Infarction of Phyllodes Tumor of the Breast in a Postpartum Woman: A Case Report

산후 여성에서 유방 엽상 종양의 자발적 경색: 증례 보고

Yoogi Cha, MD¹, Hye-Won Kim, MD^{1*}, Hun Soo Kim, MD², Tae Wan Won, MD³

Departments of ¹Radiology, ²Pathology, ³Surgery, Wonkwang University School of Medicine, Wonkwang University Hospital, Iksan, Korea

A rare tumor of the breast, phyllodes tumor is uncommon in pregnant women, and spontaneous infarction of this tumor has not been reported to date. Infarction develops in malignant tumors of the breast, but the mechanism and pathogenesis of this complication is not fully understood. Breast tumor infarction in pregnant women is uncommon, except in cases of fibroadenomas. The authors report a case of spontaneous infarction of a benign phyllodes tumor in a 30-year-old postpartum woman that exhibited rapid growth during late pregnancy; this is followed by a discussion of imaging findings.

Index terms

Breast
Infarction
Phyllodes Tumor
Magnetic Resonance Imaging
Ultrasonography

Received April 24, 2017

Revised June 14, 2017

Accepted July 4, 2017

*Corresponding author: Hye-Won Kim, MD

Department of Radiology, Wonkwang University School of Medicine, Wonkwang University Hospital, 895 Muwang-ro, Iksan 54538, Korea.
Tel. 82-63-859-1920 Fax. 82-63-851-4749
E-mail: khw@wonkwang.ac.kr

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

INTRODUCTION

Rapidly growing breast masses in lactating or pregnant women can be due to many underlying etiologies including physiologic glandular hyperplasia, cysts, fibroadenomas, abscesses, galactoceles, lactating adenomas, phyllodes tumors, and breast cancer (1-3). In particular, phyllodes tumor is rare during pregnancy or breast feeding (1). Furthermore, spontaneous infarction of these pregnancy-related breast diseases is rare and is usually not included in the list of differential diagnoses. Here, we report the first case of spontaneous infarction of a benign phyllodes tumor in a postpartum woman.

CASE REPORT

A 30-year-old woman visited our hospital one month after delivery, with a huge palpable non-tender mass in her left breast.

The mass had progressively grown during pregnancy and hardened during the postpartum period. After delivery, she breast-fed for 2 weeks and then stopped. In addition, she had a history of vacuum-assisted breast biopsy in the left breast for fibroadenoma 5 years back.

Physical examination revealed a hard, palpable mass with erythematous skin change in the bulging left breast (Fig. 1A). Mammography showed a large oval hyperdense mass in the left breast (Fig. 1B). Breast ultrasound (US) depicted a 14 cm sized oval circumscribed mass with a multiloculated cystic portion, and color Doppler US showed penetrating vascularity within the solid portion (Fig. 1C). This complex cystic and solid mass was classified as Breast Imaging Reporting and Data system (BI-RADS) category 4. US-guided core needle biopsy was performed using a 14-gauge needle, and subsequent histological analysis revealed a fibroadenoma with infarction. Magnetic resonance imaging (MRI) was recommended due to the imaging-

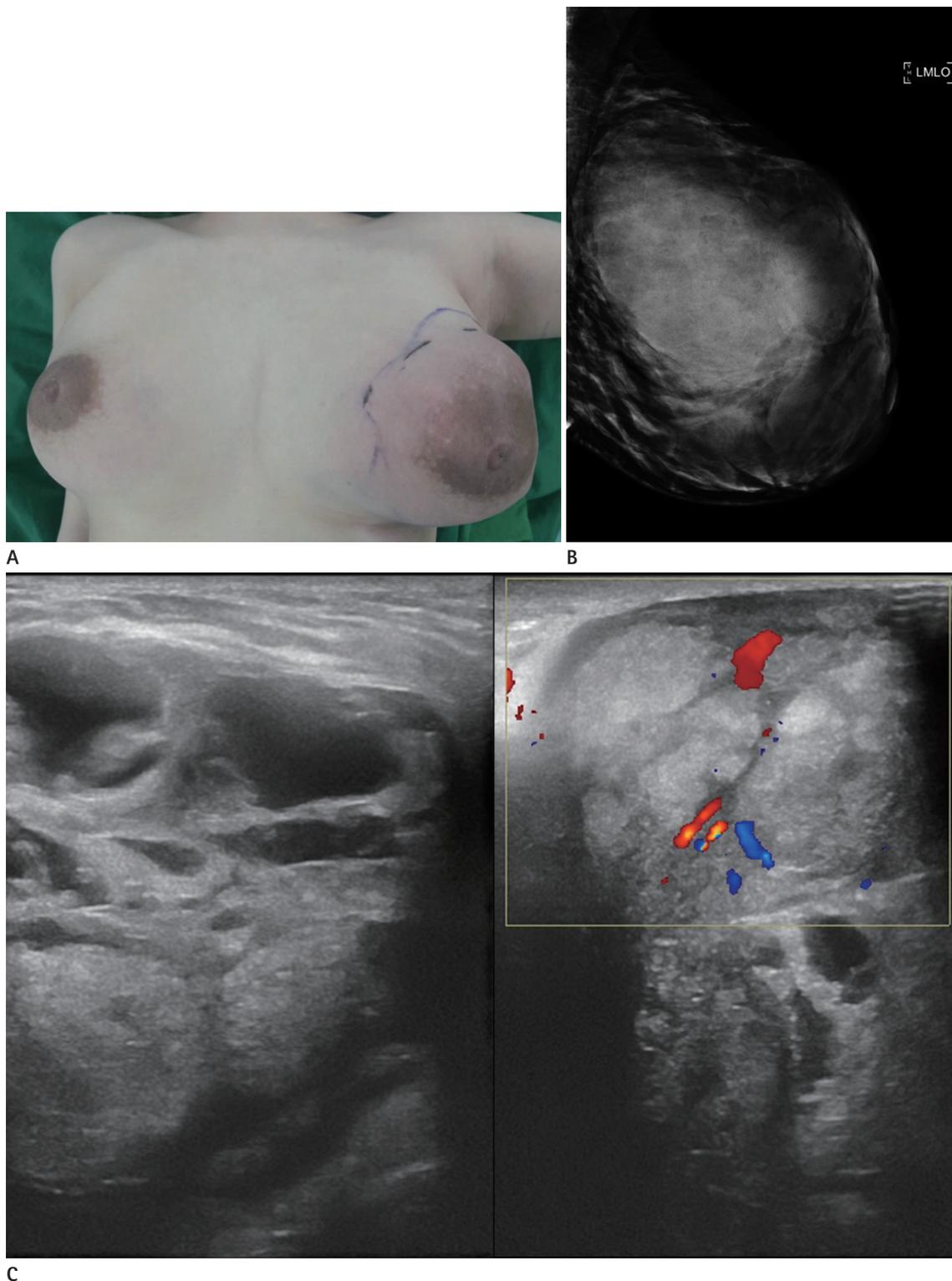


Fig. 1. A 30-year-old postpartum woman with spontaneous infarction of phyllodes tumor, presenting as a huge palpable non-tender mass in her left breast.

A. Clinical photograph shows a hard, palpable mass with erythematous skin change in a bulging left breast.

B. Mammography shows a huge oval hyperdense mass in the left breast.

C. US reveals a large circumscribed mass with a multiloculated cystic portion, and color doppler US shows penetrating vascularity within the solid portion of the mass.

US = ultrasound

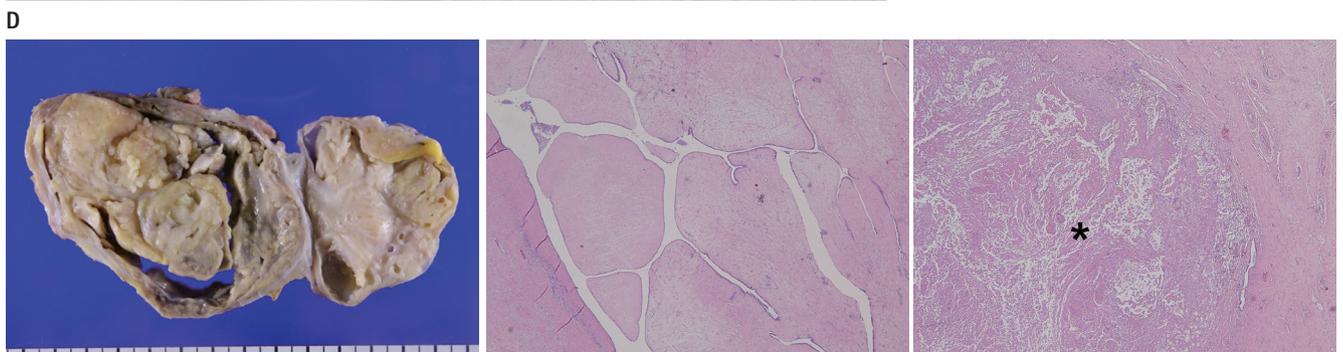
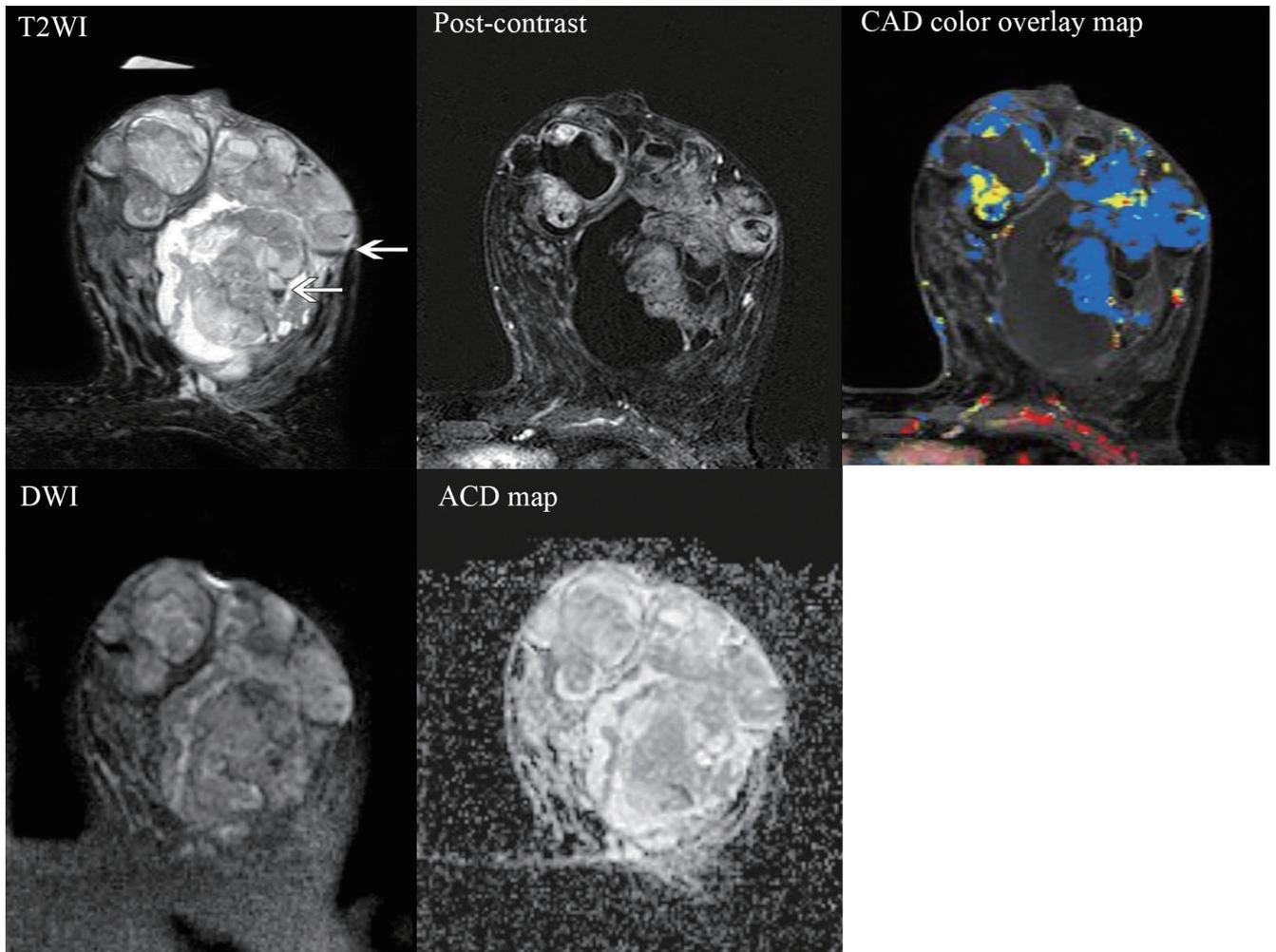


Fig. 1. A 30-year-old postpartum woman with spontaneous infarction of phylloides tumor, presenting as a huge palpable non-tender mass in her left breast.

D. Magnetic resonance imaging of the left breast mass: T2WI shows a multiloculated heterogeneously iso- to high signal intensity mass with dark septations, containing a cystic portion and scant hemorrhagic fluid collection (arrows). Fat suppressed subtraction image at 1-minute post-gadolinium injection shows heterogeneous enhancement and a smooth wall of the mass. CAD color overlay map obtained by dynamic contrast enhancement study shows that the most of the solid portion displayed a persistent kinetic pattern. DWI and corresponding ADC map show no diffusion restriction or hyperintensity.

E. Gross photograph shows a 14 × 11.5 × 8 cm in size and yellow colored mass. A section of the mass revealed a multicystic lesion with inflammation and necrosis.

F. Microscopic examination shows a tumor composed of mesenchymal nodules and leaf-like projections with intervening clefts. Hematoxylin and eosin staining (× 40) shows coagulation necrosis (asterisks) in the mass induced by infarction.

ADC = apparent diffusion coefficient, CAD = computer aided detection, DWI = diffusion-weighted image, T2WI = T2-weighted image

histologic discordance, and to identify any suspicious features of this mass. MRI revealed a huge, heterogeneous, circumscribed mass containing multiloculated fluid. The solid portion of the mass exhibited iso- to high signal intensity and dark septa on T2-weighted images (T2WI), with no diffusion restriction on diffusion-weighted image (DWI) and isointensity on apparent diffusion coefficient (ADC) map. A gadolinium-enhancement study visualized a heterogeneous, well-enhanced mass, and a dynamic contrast enhancement study revealed a 77% persistent and 20% plateau kinetic pattern in the solid portion of the mass (Fig. 1D). The mass was classified as BI-RADS category 4.

The patient underwent mass excision of a 14.0 × 11.5 × 8.0 cm white-to-yellow colored mass containing a multiloculated cystic portion with inflammation and necrosis (Fig. 1E). Histopathologic analysis revealed benign phyllodes tumor with infarction (Fig. 1F).

DISCUSSION

Physiologic glandular hyperplasia, cysts, fibroadenomas (influenced by estrogen), abscesses, galactoceles, lactating adenomas, phyllodes tumors, and breast cancer are normally included in the differential diagnoses of rapidly growing breast masses in lactating or pregnant women (1-3). Since the treatment options of these conditions differ, it is very important to establish an accurate diagnosis (1).

Phyllodes tumor is a rare breast tumor, accounting for 1% of primary breast tumors, and rarely occurs during pregnancy and lactation. A previous study of 15 cases reported that phyllodes tumors arising during pregnancy or lactation could be benign, borderline, or malignant. Phyllodes tumor is rare in lactating breasts due to the presence of lobular and acinar hyperplasia, which are required to satisfy the physiological requirements during lactation. Thus, it is hypothesized that as stromal hyperplasia is not a feature of the lactating breast, the probability of developing phyllodes tumor is low (1). Furthermore, it is unknown if this rapidly growing mass is hormone dependent in pregnant women (4).

Spontaneous infarction of a breast mass is rare. However, it may be associated with pregnancy, lactation, recent fine needle aspiration, cardiovascular disease, or the use of oral contraceptives (3). Although the mechanism of breast mass infarction is un-

known, some hypotheses have been proposed, especially for fibroadenomas. One suggestion concerns relative vascular insufficiency due to increased metabolic need during pregnancy or lactation. Additional hypotheses suggest that infarction may be secondary to calcification with hyalinization and mechanical factors related to mass mobility, which may reduce the blood supply, or thrombo-occlusive vascular changes in feeder vessels (3, 5). In the case of giant juvenile fibroadenomas, the rapid growth and large size are proposed as the mechanisms of infarction, as the tumor may outgrow its blood supply (6).

There are few publications on the imaging findings of infarcted breast disorders. The majority of reported cases of infarcted benign tumors involve fibroadenomas, and some reports have described infarcted fibroadenomas or lactating adenomas during pregnancy or lactation (3). In one previous report, the sonographic findings of an infarcted fibroadenoma include a circumscribed complex cystic and solid mass with internal multiple small anechoic portions having posterior acoustic shadowing (3, 5).

US features of infarcted breast tumors include inhomogenous, low echoic, circumscribed lesions that may be accompanied by posterior acoustic enhancement. Oh et al. (7) concluded that an inhomogenous, low echoic lesion suggests infarction, and combined high echoic lesions indicate hemorrhage. The radiologic features of these tumors depend on the stage of infarction. The imaging findings of our patient were similar to those previously reported, and consisted of a huge, inhomogeneous mass lesion with an internal anechoic portion histopathologically related to necrosis.

Typical MR findings of infarcted breast tumors are yet to be established. For infarcted juvenile fibroadenomas, MRI findings of infarcted tumors include solid portions that exhibit less pronounced enhancement, and edema, which is sometimes associated with hemorrhage and progressive enhancement (6). In one case of an infarcted phyllodes tumor in a non-pregnant woman, the MRI revealed a 3 cm sized slightly irregular mass of predominantly cystic appearance due to massive infarction (8).

The MRI findings need to be correctly analyzed to enable the differentiation of benign and malignant phyllodes tumors, since the histologic features of the phyllodes tumor influences the treatment decision making and prognosis. Yabuuchi et al. (9) reported equal to lower signal intensity of tumor than normal breast tissue on T2WI, cystic change with an irregular wall, and low

ADCs on DWIs as features of histopathologically malignant phyllodes tumor. Another study reported that larger tumor size, non-enhanced T2 low signal intensity septations, and silt-like patterns in enhanced images, are correlated significantly with the malignant histologic grade (10).

In our case, the MRI depicted a huge, heterogeneous, well-enhancing mass with cystic change and non-enhanced T2 low signal intensity septations mimicking malignant phyllodes tumor. However, high signal intensity of the mass on T2WI was comparative with normal breast tissue, cystic change with a smooth thin wall, delayed persistent kinetic patterns of the solid portion during the dynamic contrast enhancement study, and no significant diffusion restriction of the mass, indicating a benign nature.

MRI is a useful imaging modality to help differentiate an infarcted benign tumor from a malignant tumor with hemorrhagic necrosis, in pregnant patients presenting with a rapidly growing well circumscribed mass with cystic features on US examination. Here, we report the first case of spontaneous infarction of a benign phyllodes tumor in a postpartum woman.

Acknowledgments

This study was supported by Wonkwang University in 2015.

REFERENCES

1. Murthy SS, Raju KV, Nair HG. Phyllodes tumor in a lactating breast. *Clin Med Insights Pathol* 2016;9:13-17
2. Behrndt VS, Barbakoff D, Askin FB, Brem RF. Infarcted lactating adenoma presenting as a rapidly enlarging breast mass. *AJR Am J Roentgenol* 1999;173:933-935
3. Kim JY, Kim KS, Lee Y, Kim JH. Spontaneous Infarction of benign breast lesion during pregnancy: ultrasonographic and pathologic findings. *J Korean Soc Radiol* 2015;73:259-263
4. Likhitmaskul T, Asanprakit W, Charoenthammaraksa S, Lohsiriwat V, Supaporn S, Vassanasiri W, et al. Giant benign phyllodes tumor with lactating changes in pregnancy: a case report. *Gland Surg* 2015;4:339-343
5. Kim SJ. Spontaneously infarcted fibroadenoma of the breast in an adolescent girl: sonographic findings. *J Med Ultrason (2001)* 2014;41:83-85
6. Jesinger RA, Lattin GE Jr, Ballard EA, Zelasko SM, Glassman LM. Vascular abnormalities of the breast: arterial and venous disorders, vascular masses, and mimic lesions with radiologic-pathologic correlation. *Radiographics* 2011;31:E117-E136
7. Oh HJ, Kim SH, Kang BJ, Lee AW, Song BJ, Kim HS, et al. Ultrasonographic features of spontaneous breast tumor infarction. *Breast Cancer* 2015;22:596-601
8. Verslegers I, Tjalma W, Van Goethem M, Colpaert C, Biltjes I, De Schepper AM, et al. Massive infarction of a recurrent phyllodes tumor of the breast: MRI-findings. *JBR-BTR* 2004;87:21-22
9. Yabuuchi H, Soeda H, Matsuo Y, Okafuji T, Eguchi T, Sakai S, et al. Phyllodes tumor of the breast: correlation between MR findings and histologic grade. *Radiology* 2006;241:702-709
10. Tan H, Zhang S, Liu H, Peng W, Li R, Gu Y, et al. Imaging findings in phyllodes tumors of the breast. *Eur J Radiol* 2012;81:e62-e69

산후 여성에서 유방 엽상 종양의 자발적 경색: 증례 보고

차유지¹ · 김혜원^{1*} · 김헌수² · 원태완³

엽상 종양은 임신한 여성에서 드문 유방 종양이며 이러한 종양의 자발적 경색은 현재까지 보고된 바 없다. 경색은 주로 유방의 악성 종양에서 발생하나, 이러한 합병증의 발병 기전과 병인은 아직 정확히 알려지지 않았다. 임신한 여성에서 유방 종양의 경색은 섬유선종의 경우를 제외하면 드문 것으로 알려져 있다. 이에 저자들은 30세 여성의 유방에서 임신 후기에 빠른 성장과 산후 자발적 경색을 보였던 양성 엽상 종양의 증례를 영상 소견의 고찰과 함께 보고한다.

원광대학교 의과대학 원광대학교병원 ¹영상의학교실, ²병리학교실, ³외과학교실