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Ductal Carcinoma in situ of the breast in sclerosing adenosis encapsulated by a hamartoma: A case report

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ABSTRACT

INTRODUCTION: Ductal Carcinoma in situ (DCIS) of the breast can develop in areas of sclerosing adenosis. The radiographic finding of sclerosing adenosis is a spiculated mass and can look like invasive ductal carcinoma. We report a patient with DCIS in sclerosing adenosis encapsulated by a hamartoma, with imaging findings quite different from the typical findings of sclerosing adenosis.
PRESENTATION OF CASE: A 73-year old woman, with no previous mammography, presented with a palpable mass in the left breast. Mammography showed a 36 mm well-defined mass with fat density in the middle outer quadrant of the left breast. Ultrasonography showed a well-defined mass in the same area which was composed of hypoechoic and hyperechoic areas. The histological diagnosis by core needle biopsy was sclerosing adenosis. We considered the patient's age and tumor size and performed a partial mastectomy for both diagnosis and treatment. Final pathology showed DCIS in sclerosing adenosis in a hamartoma.
DISCUSSION: This patient had DCIS in an area of sclerosing adenosis, encapsulated by a hamartoma. DCIS can develop in areas of sclerosing adenosis, and can appear similar to invasive ductal carcinoma, so we must avoid misdiagnosis or over-treatment. Malignant transformation of a hamartoma is rare, but can occur since it contains epithelial tissue. Definitive biopsy should be performed due to the possibility of a malignancy inside the hamartoma.
CONCLUSIONS: When diagnosing a hamartoma, the presence of atypical findings on imaging studies, should suggest the possibility of malignancy. Although rare, a malignant tumor may be present inside the hamartoma.

1. Introduction

Sclerosing adenosis is a benign proliferative breast disease that presents with acinar, myoepithelial, and connective tissue changes in the terminal ductal lobular unit. It is known that ductal carcinoma in situ (DCIS) can develop in areas of sclerosing adenosis [1–3]. When DCIS is associated with sclerosing adenosis, accurate diagnosis becomes more difficult due to similarities between these conditions, which potentially leads to a misdiagnosis as invasive ductal carcinoma [4–6]. A hamartoma is a benign tumor consisting of a fibrous fatty stroma with various amounts of epithelial elements [7]. A hamartoma has a typical mammographic appearance of lucent lesions containing fat, varying dense fibrous and adenomatous elements, a sharp margin which is a thin radiopaque line, and sometimes a thin capsule [8]. Ultrasonographically, a well-defined mass with an echogenic rim and internal heterogeneity is shown displacing the adjacent normal breast tissue [9]. Carcinoma in a hamartoma has been reported [10], but is very rare. In the present patient, it was difficult to establish the diagnosis because a hamartoma covered an area of sclerosing adenosis which included DCIS. This pathologic condition has been never reported to the best of our knowledge. This work is reported in accordance with the SCARE criteria [11].

Abbreviation: DCIS, ductal carcinoma in situ.

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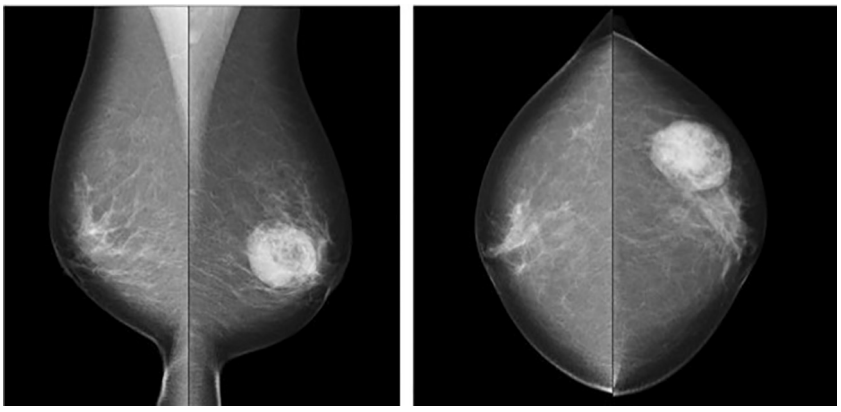

Fig. 1. Mediolateral-Oblique (MLO) and Cranial-Caudal (CC) views of the left breast, show a well-defined, fat-containing mass, suggesting a hamartoma. The mass has a relatively high density compared to surrounding breast tissue.

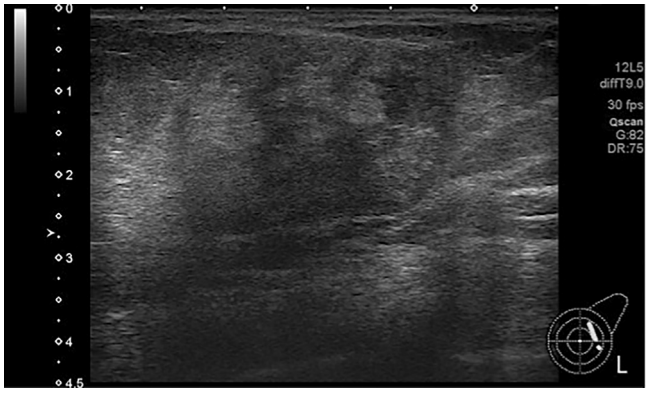

Fig. 2. Ultrasonography, showing a well-defined mass 36 × 24 mm in size containing both hypoechoic and hyperechoic lesions.

2. Presentation of case

A 73-year old woman, with no previous mammography, presented a palpable lesion in the left breast. She discovered it one month before presentation. She had a past medical history of insomnia and glaucoma. Her family history was not significant. Physical examination identified a mass which was elastic, 30 mm in size and located in the middle outer quadrant of the left breast. She underwent mammography and ultrasound. Mammography showed a well-defined mass, which included fat densities, in an elliptical mass in the same location found on physical examination (Fig. 1). Ultrasound examination showed a well-defined mass which was 36 × 24 mm in size and with mixed by hypoechoic and hyperechoic areas (Fig. 2).

Based on these findings, the differential diagnosis included a hamartoma or phyllodes tumor, in consideration of her age and a core needle biopsy was performed. Histopathology showed an increase in glandular elements with stromal proliferation and indistinct myoepithelial cells, which looked like invasive ductal carcinoma, but immunohistochemistry showed a normal two-layer structure of mammary glandular epithelial cells and myoepithelial cells which led to the diagnosis of sclerosing adenosis (Fig. 3). However, the diagnosis was not able to explain the imaging findings which showed a well-defined mass. We considered the patient's age and the lesion size and decided to perform a partial mastectomy under general anesthesia for both diagnosis and appropriate treatment. The incision was 5 cm long over the tumor and the surgical margins were 1 cm, taking into account the possibility that the tumor was malignant. Intraoperative findings showed no infiltration into surrounding tissue and the operation was uneventful (the operating time was about 1 h with minimal bleeding).

The postoperative course was uneventful and she was discharged from the hospital on postoperative day one. The postoperative pathological diagnosis was DCIS in an area of sclerosing adenosis, in a hamartoma (Figs. 4 and 5). The DCIS lesion measured 25 × 23 × 20 mm. DCIS proliferated in sclerosing adenosis encapsulated by a hamartoma, which contained normal breast tissue and fat tissue and separated from surrounding normal tissue by a thin fibrous capsule. The DCIS was papillary-cribriform, solid and flat. Tumor cells showed partial apocrine metaplasia. The immunohistochemical characteristics were estrogen receptor weakly positive (Allred score 3), progesterone receptor negative (Allred score 0), and human epidermal growth factor receptor type 2 positive (Immunostaining score 3). We recommended adjuvant treatment with radiotherapy. However, after extensive discussion with the patient, radiation therapy was omitted at the patient's request.

3. Discussion

DCIS in sclerosing adenosis is usually an indistinct lesion on imaging with architectural distortion on both mammographic and ultrasound imaging. These findings can appear similar to invasive ductal carcinoma [4–6], so we must avoid misdiagnosis or overtreatment. In this patient, both mammogram and ultrasonography findings showed a well-defined mass including fat densities which looked like a hamartoma. However, in retrospect, the fact that the mass was relatively hard, mammography showed high density, and ultrasonography showed a high depth to width ratio with poor compressibility, are not typical findings of a typical hamartoma. The core needle biopsy showed sclerosing adenosis, which was not expected from the result of imaging studies. Retrospectively, we might be able to explain the discrepancy between clinical presentation, imaging findings and pathological ones, because DCIS in an area of sclerosing adenosis was encapsulated by a hamartoma. At that time, we considered the age and the tumor size and decided to resect the lesion for both diagnosis and treatment, without a vacuum-assisted biopsy instrument. The postoperative pathological diagnosis was DCIS in sclerosing adenosis in hamartoma, with the hamartoma obscuring typical imaging findings of sclerosing adenosis. This combination of lesions (DCIS in sclerosing adenosis in a hamartoma) has not been reported to the best of our knowledge.

It is well known that DCIS develops in areas of sclerosing adenosis [6]. The frequency of occurrence of this combination is unknown because there are few reports. We previously reported that DCIS in sclerosing adenosis more frequently presents with architectural

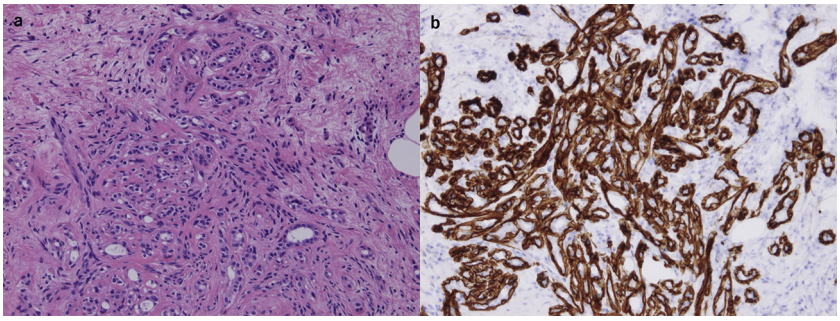

Fig. 3. Two core biopsies were performed showing similar results. Hematoxylin and eosin stained sections (a, ×100.) show an increase in glandular elements plus stromal proliferation and indistinct myoepithelial cells, which looked like invasive ductal carcinoma, but immunohistochemistry (b, ×100.) show a normal two layer structure of mammary glandular epithelial cells and myoepithelial cells, diagnosed as sclerosing adenosis.

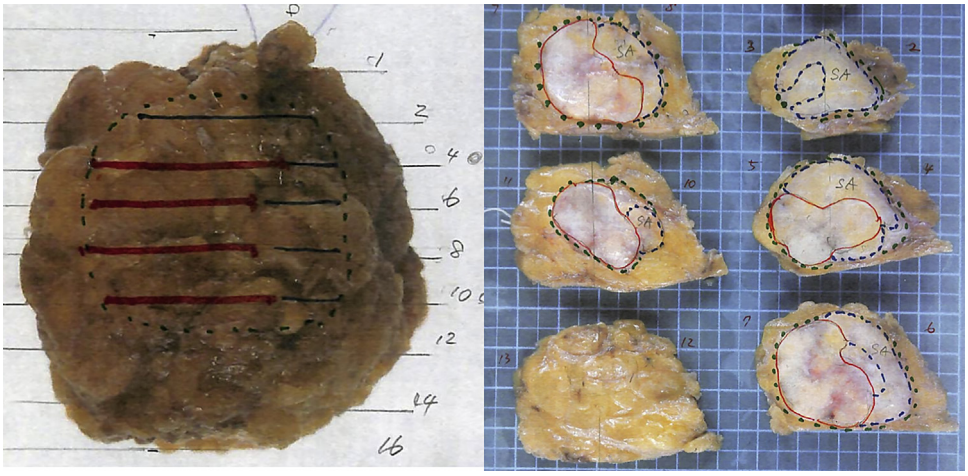

Fig. 4. Specimens from a partial mastectomy. The tumor was elastic and did not appear to infiltrate surrounding tissue. Ductal carcinoma in situ (the black line) in an area of sclerosing adenosis (the red line), in a hamartoma (the dotted line).

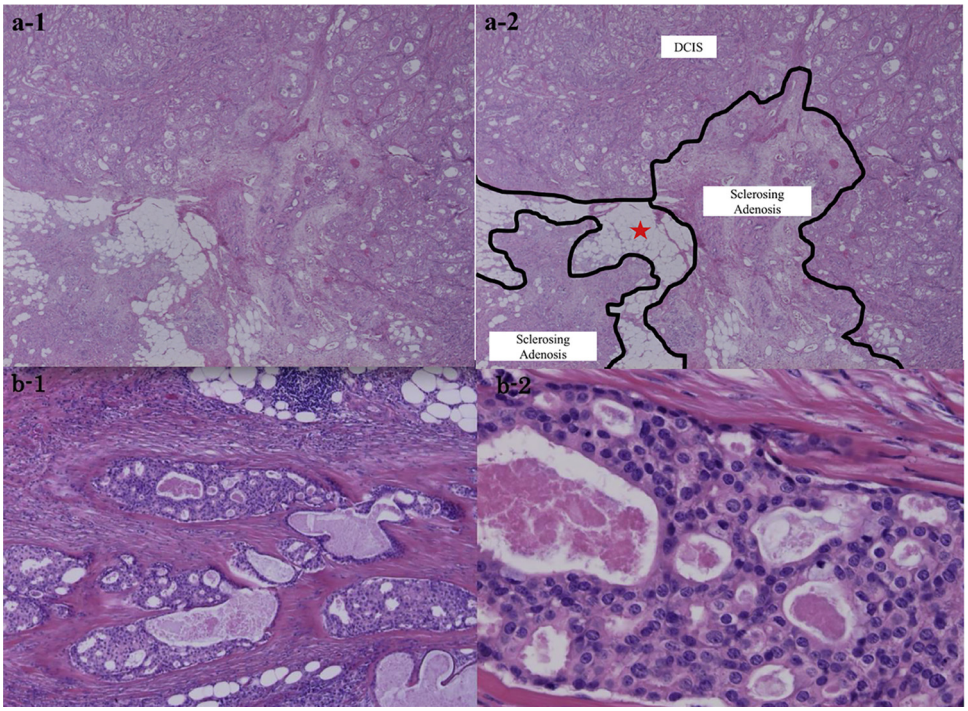

Fig. 5. Photomicrograph of the center of the excised specimen at low magnification (a-1:×1, a-2: a-1 scheme) showing ductal carcinoma in situ in sclerosing adenosis in a hamartoma (fatty tissues marked by the red star). Photomicrograph of the center of the excised specimen at low and high magnification (b-1:×4, b-2:×20) showing ductal carcinoma in an area of sclerosing adenosis.

distortion on mammography and ultrasonography compared with DCIS that is not associated with sclerosing adenosis and has a higher risk of bilateral breast cancer, which was seen in 38% of the patients with DCIS in sclerosing adenosis [6]. DCIS in sclerosing adenosis within a hamartoma, as in this patient, might not display these features.

Malignant transformation of a hamartoma is rare, but can occur since it contains epithelial tissue [12]. A recent review of the literature described 15 cases of carcinoma associated with hamartomas [10]. In the majority of cases, the diagnosis was on mammographic or ultrasound findings of suspicious features within an otherwise typical hamartoma (specifically, microcalcifications or a speculated lesion on mammography, irregular hypoechoic lesions on ultrasonography). There are no microcalcifications or speculated opacities on mammography in the present patient, but the lesion had irregular hypoechoic lesions on ultrasonography. These findings are not typical of DCIS in a hamartoma. In fact, this patient's findings are not typical for DCIS in an area of sclerosing adenosis or DCIS in a hamartoma. The hamartoma may have been present for a long time, after it began transforming into sclerosing adenosis, and then DCIS developed in this area of sclerosing adenosis.

A hamartoma is usually diagnosed based on typical imaging findings, but definitive biopsy should be performed due to the possibility of a malignancy inside the hamartoma. This is particularly true if the imaging findings are atypical or if there are discrepancies between the clinical presentation, imaging findings and pathological findings.

4. Conclusion

This patient presented with DCIS in an area of sclerosing adenosis encapsulated by a hamartoma. When diagnosing a hamartoma, the presence of atypical imaging findings such as a dense opacity on the mammogram and hyper-vascularity on ultrasound, suggest a malignancy. Although rare, it is necessary to consider the possibility of a malignant tumor inside a hamartoma.

Conflicts of interest

There are no conflicts of interest to be declared.

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Ethical approval

The ethical approval has been exempted as it was not necessary in this case report by our institution.

Consent

Informed consent for the publication of this work was given by the patient.

Author contribution

SF and AY gathered the patient's data and wrote the manuscript. SF, KM participated in the surgery. KM was responsible for the inpatient optimization. FA and KS were responsible for pathological diagnosis of this case. FA, AY, JK, TS, HT, AKL, KS and KM reviewed manuscript. All authors approved the final manuscript.

Guarantor

Shota Fukai MD.

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