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.

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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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relatively
both
Fig.
age
hyperechoic
tumor
treatment.
increase
showed
presented
elliptical
in
needle
biopsy
mammary
immunohistochemistry
these
and
margins
anesthesia
both
mammography,
and
physical
history
not
mammogram,
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
hypoechogenic
and
hyperechogenic
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

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
over-
treatment.
In
this
patient,
both
mammogram
and
ultrasound
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
ultrasound
showed
a
high
depth
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 patient 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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