

Giant epidermal inclusion cyst with infection arising within the breast parenchyma: a case report

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Abstract

Epidermal inclusion cysts (EICs) of the breast develop in the deep breast parenchyma, and they are very rare. Only about 10 cases have been reported in the English-language literature to date. In this report, we present a rare case of a giant EIC with infection arising within the deep breast parenchyma. Unlike a typical EIC of the breast, the EIC in the present case was a cystic and solid lesion containing a large amount of liquid within the cyst and popcorn-like calcification in the wall. In this report, we describe the contrast-enhanced spectral mammography (CESM), ultrasonography, and computed tomography findings and provide a reference for the diagnosis of EICs. To the best of our knowledge, this is the first report of the CESM findings of an EIC. Our case illustrates that CESM has excellent performance similar to that of magnetic resonance imaging and is much more effective than conventional digital mammography. Additionally, our case indicates that precise correlation of CESM with ultrasonography findings contributes to the diagnosis of EICs. This rare case with multiple imaging findings will increase the awareness of EICs in the breast parenchyma.

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Introduction

An epidermal inclusion cyst (EIC) is a benign lesion lined by stratified squamous epithelium and filled with lamellated keratin and epithelial and keratin debris.¹ EICs may occur anywhere in the body, although they are more common in body areas containing hair such as the face, neck, trunk, extremities, and scalp.² An EIC of the breast is uncommon and typically occurs in the cutaneous or subcutaneous tissue of the periareolar region. Some EICs seem to grow deep inside the parenchyma of the breast, whereas part of the lesion in fact protrudes into the skin or connects with it as shown by ultrasonography (US). EICs that occur in the deep breast parenchyma and neither protrude into the skin nor adhere to it are very rare. Only about 10 such cases have been reported in the English-language literature to date.^{1,3–10}

In this report, we present a rare case of a giant EIC with infection arising in the deep breast parenchyma. In contrast to a typical EIC of the breast, the EIC in this case was a cystic and solid lesion with a large amount of liquid within the cyst and popcorn-like calcification in the wall. We describe the contrast-enhanced spectral mammography (CESM), computed tomography (CT), and US findings in this case and provide a reference for the diagnosis of EICs. To the best of our knowledge, this is the first report of the CESM findings of an EIC.

Case report

A 53-year-old postmenopausal woman presented with a slightly painful palpable mass

in the left breast. The mass had persisted for 20 years with no discomfort. It had begun to rapidly increase in size during the last 2 months. The patient had no history of trauma or surgical procedures involving the breast skin. Physical examination revealed an oval, well-circumscribed mass with a diameter of 8 cm occupying the upper region of the left breast. The lesion was firm, freely mobile, and non-adherent to the overlying skin. Systemic examination revealed no abnormalities.

US showed an oval, circumscribed, heterogeneously complex cystic and solid mass within the mammary parenchyma in the upper region of the left breast, measuring 7.0 × 7.5 cm (Figure 1). No color flow signal was detected inside the lesion. Mild edema was present in the perilesional parenchyma. The bilateral axillary lymph nodes were slightly enlarged, the largest on the left



Figure 1. Ultrasonography showed an oval, circumscribed, heterogeneously complex cystic and solid mass within the mammary parenchyma. Mild edema was present in the perilesional parenchyma.

measuring 1.9×0.8 cm. The lesion was classified as BI-RADS Category 4A.

CESM was performed using a digital mammography system (Senographe Essential; GE Healthcare, Chicago, IL, USA). Briefly, after the patient had undergone intravenous injection of a contrast medium (iohexol), mammography was performed with low-energy and high-energy images almost simultaneously. The obtained images were subtracted according to a specific algorithm and then evaluated to detect uptake of the iodine contrast agent in the enhancing lesions.^{11,12} A low-energy mammogram is equivalent to a standard mammogram.

The low-energy mammogram revealed an oval, circumscribed, high-density mass with popcorn-like calcification in the upper region of the left breast, measuring 7.5×7.8 cm. Several bilateral axillary lymph nodes were enlarged (Figure 2(a) and (b)). In the subtracted images, an oval circumscribed cyst with septa and rim enhancement was observed. Part of the wall and septa were thin and smooth, whereas the others were thickened and obscure. The intensity in the cyst was low compared with the background enhancement, which was mild (Figure 2(c) and (d)).

A chest CT scan was performed at the patient's request because her mother had died of lung cancer. The CT scan revealed a well-defined cyst behind the gland in the left breast with a diameter of 7.8 cm, compressing the anterior aspect of the mammary gland. The thickness of the cyst wall was 0.2 to 1.0 cm, and popcorn-like calcification was present in the left wall. Septa divided the cystic spaces into two parts. The density was homogeneous and the CT value ranged from -2 to 6 HU (Figure 3).

The subsequent US-guided core needle biopsy depicted a benign cystic and solid lesion containing a large amount of brown fluid. The mass was surgically excised, and the pathological diagnosis was an EIC with

infection. In the pathological examination, the lesion macroscopically comprised two closely adjacent cysts. The thickness of the cyst wall was 0.2 to 1.0 cm. Coarse calcification was present in the wall, and white, gray, and black substances were observed in the lumen. Microscopically, the cystic lesion was lined with benign stratified squamous epithelium and filled with lamellated keratin. The adjacent tissue exhibited infiltrations of inflammatory cells (Figure 4).

Discussion

An EIC of the breast typically presents as a single small lump in the cutaneous or subcutaneous tissue of the periareolar region. The EIC usually appears as a solid, well-circumscribed mass on US and as a homogeneous, well-circumscribed mass with increased density on mammography.¹⁻³

We have herein reported a very rare case of a giant EIC in the breast. The EIC was associated with infection and arose in the deep breast parenchyma; it was cystic and solid with a large amount of liquid in the cyst and popcorn-like calcification in the wall. Part of the cyst wall was thickened secondary to infection. This case is unusual in three aspects. First, the location is uncommon. The EIC occurred in the deep breast parenchyma and pushed against the anterior aspect of the mammary gland without connection to the skin. The exact pathogenesis of a cyst arising in the breast parenchyma is poorly understood. Of the 10 cases reported in the English-language literature, 5 EICs developed after reduction mammoplasty,^{6,8,9} possibly as a result of implantation of superficial epidermal tissue into the deep tissue after the surgical procedure.^{13,14} The patients in the other five cases had no history of trauma or surgery; two of these EICs were believed to have arisen from metaplasia of the ductal epithelium,^{1,7} and the pathogenesis of the remaining three were unknown. We presume that

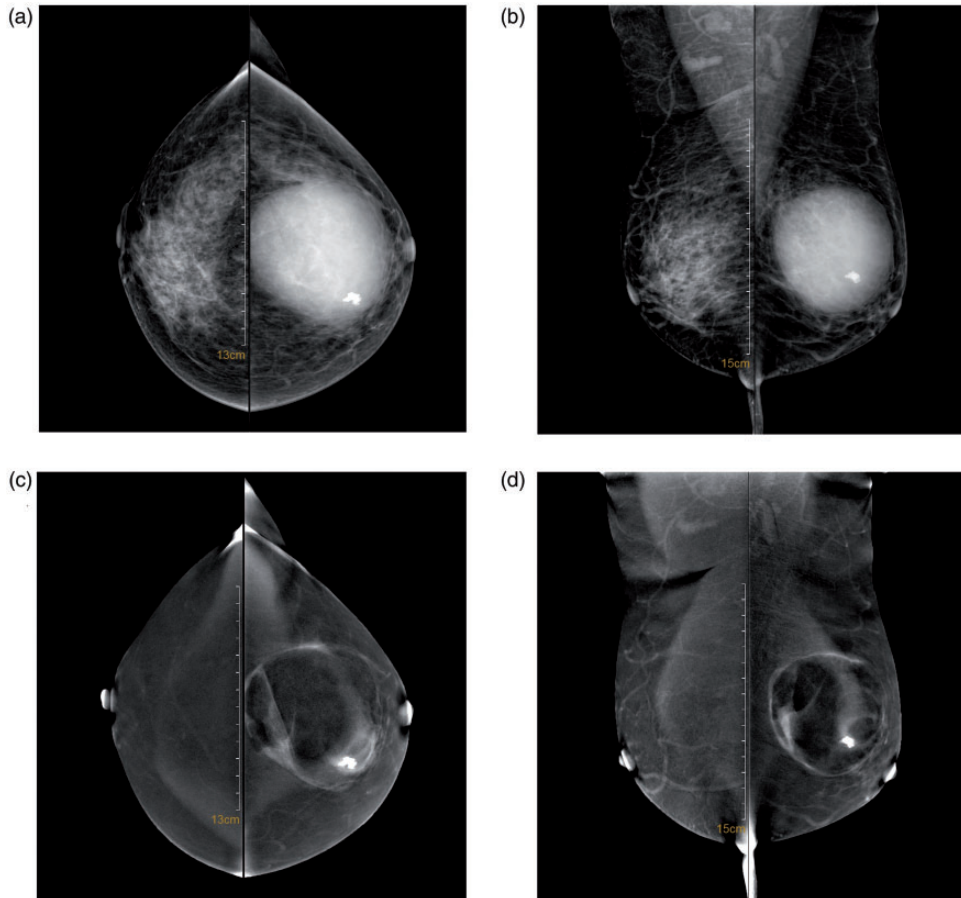


Figure 2. (a) Craniocaudal and (b) mediolateral oblique low-energy images (equivalent to standard mammogram) show an oval, circumscribed, high-density mass with popcorn-like calcification in the upper region of the left breast. (c) Craniocaudal and (d) mediolateral oblique subtracted images revealed an oval circumscribed cyst with septa in the upper region of the left breast. Peripheral contrast enhancement was observed. Part of the wall and septa were thin and smooth, and the others were thickened and obscure. The intensity in the cyst was low compared with the background enhancement, revealing the cystic nature of the lesion. The background enhancement was mild.

the EIC in our case was congenital, growing slowly from cell nests. Second, a cystic and solid EIC in the breast with a large amount of liquid is unusual. To our knowledge, only one case of an EIC in the breast containing a large amount of fluid from this origin has been reported in the literature to date.⁴ In that case, however, only mammogram features were described, and the possible causes of the fluid formation were

not analyzed. In our case, the infection of the cyst might have caused pus formation and liquefaction necrosis, resulting in the accumulation of brown fluid within the cyst. Third, popcorn-like calcification in the walls of EICs of the breast is rare. Calcification may occasionally be seen in the walls or within the keratin debris of EICs in the breast; such calcification has been described as heterogeneous

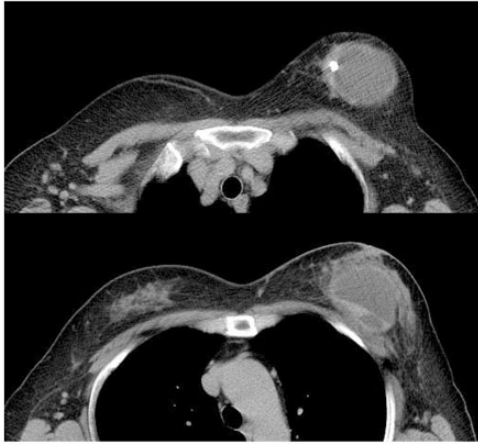


Figure 3. Computed tomography revealed a well-defined cyst behind the gland in the left breast with a diameter of 7.6 cm, compressing the anterior aspect of the mammary gland. The thickness of the cyst wall was 0.2 to 1.0 cm, with popcorn-like calcification in the left wall. Septa divided the cystic spaces. The cyst cavity had a homogeneous density, and the computed tomography value was -2 to 6 HU.

microcalcification, speckled calcification, and egg-shell peripheral calcification in literature.^{8,14} To the best of our knowledge, popcorn-like calcification in the wall has not been previously described. Such calcification might occur following necrosis of the epithelial lining cells.

In the present case, we performed multiple imaging examinations including CEM, CT, and US. To the best of our knowledge, this is the first report of the CEM findings of an EIC. CEM is a new mammographic technique. It helps to detect lesions by highlighting enhancement after injection of a contrast agent and acquisition of the weighted logarithmic subtracted image. CEM provides morphological and functional information, and its performance is similar to that of breast magnetic resonance imaging (MRI) in the diagnostic setting.¹⁵⁻¹⁷ Compared with breast MRI, the cost of CEM is lower and the examination time is shorter (around 7 minutes).^{18,19} The

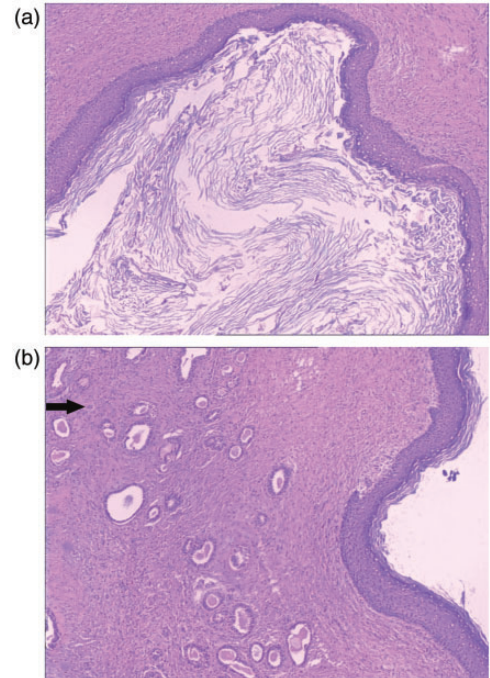


Figure 4. Histopathologic examination demonstrated two cysts filled with lamellated keratin and epithelial and keratinous debris. The cystic lesions were lined by stratified squamous epithelium (hematoxylin and eosin staining, $40\times$). The adjacent tissue exhibited infiltration of inflammatory cells (black arrow). The diagnosis was an epidermal inclusion cyst with infection.

low-energy images in our case revealed a well-circumscribed, hyperdense homogeneous density, consistent with previous reports in the literature.^{8,14} These features are similar to those of a fibroadenoma or phyllodes tumor; they are also similar to the benign features of a malignant lesion such as a mucinous carcinoma. In addition to the conventional mammographic features, CEM revealed more details of the lesion. Subtracted images showed that the cyst had peripheral rim enhancement, which has been previously shown on MRI,^{2,10,20} and part of the cyst wall was thickened, which was consistent with the CT findings. The intensity in the cyst was low compared

with the background enhancement, revealing the cystic nature of the lesion.²¹ CESM revealed more details than conventional digital mammography.

Diagnosis of EICs in the deep breast parenchyma is more difficult than diagnosis of EICs in the cutaneous or subcutaneous tissue. In particular, the imaging findings of the EIC with infection in the present case were atypical. We found that precise correlation of CESM and US findings can provide more details of the mass components than either imaging technique alone. The combined US and CESM imaging features illustrated that the mass was a cyst with solid or thick components without angiogenesis. In recent years, dual-energy CT has been used to characterize breast lesions.²² This technique exploits the same physical principle of CESM. It can not only evaluate the effective uptake of contrast medium by the lesion but can also characterize tissue using z-analysis.^{23,24} This technique might have high diagnostic value for EICs with a complex composition.

In conclusion, we have presented a rare case of an EIC with infection arising within the breast parenchyma. This case shows that CESM has excellent performance similar to that of MRI and is much more effective than conventional digital mammography. The precise correlation of CESM and US findings with the patient's clinical history can provide a more meaningful and accurate interpretation. This case will increase the awareness of EICs in the breast parenchyma and highlight the diagnostic value of associating CESM with US.

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Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Declaration of conflicting interests

The authors declare that there is no conflict of interest.

Ethics and consent

All the authors consent to publication of the manuscript in this journal. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal. This case report followed the CARE guidelines. The study was reviewed and approved by the Institutional Review Board of Qingdao University Medical College Affiliated Yantai Yuhuangding Hospital (approval no. 2019-393).


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Authors' contributions

YZ drafted the manuscript and contributed to the revision of the manuscript. LS and HZ helped to draft the manuscript. FL, HG, JL, and HX contributed to the critical revision of the manuscript. HS conceived of the study and participated in its design and coordination. All authors read and approved the manuscript.

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