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Case Report

Acquired cystic lymphangioma imitating breast cancer recurrence ☆,☆☆

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ABSTRACT

Cystic lymphangioma is a benign, congenital lymphatic malformation mainly encountered in infants during the first 2 years of life. It is rarely found in adults. Cystic lymphangioma of the breast is an extremely uncommon entity, and only a few cases have been described in the literature.

We present the case of a 52-year-old female who had a mastectomy and chemoradiotherapy for breast cancer 8 years ago and in whom we discovered a suspicious mass of the treated breast through an annual imaging check-up. A cancer recurrence was suspected, and the patient underwent surgical resection. Pathology results were consistent with a cystic lymphangioma.

Introduction

Cystic lymphangioma is a benign, congenital lymphatic malformation, mainly encountered in infants during the first 2 years of life [1,2]. Adult cystic lymphangiomas are exceedingly uncommon, as they are likely the consequence of a lymphatic obstruction secondary to an extra-lymphatic origin [3]. Cystic lymphangiomas are typically located in the neck, the axillary region, and the abdomen [1]. Breast cystic lymphangioma is an extremely rare entity, and only a few cases have been reported in the literature [4]. Imaging modalities may assist in the diagnosis of breast lymphangioma when they demonstrate classic imaging features [1,5]. However, excluding malignancy is often challenging, especially in patients with a history of treated breast cancer.

We report the case of a 52-year-old female who had a mastectomy and chemoradiotherapy for breast cancer 8 years ago and in whom we discovered a suspicious lesion through an annual imaging check-up.

Case presentation

A 52-year-old female was referred to the radiology department for a suspected breast cancer recurrence reported in her annual follow-up CT. The patient had a history of invasive ductal

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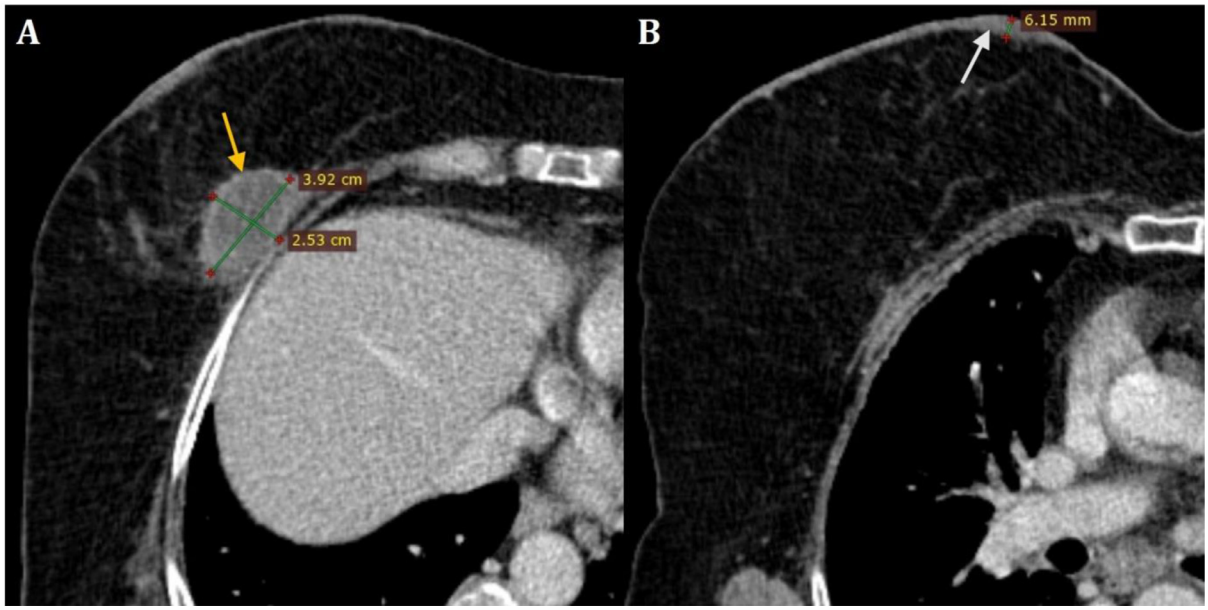

Fig. 1 – Axial chest contrast-enhanced CT scan images showing an irregular mass of mixed density and spiculated margins in the junction of the inferior right quadrants (yellow arrow) as well as skin thickening (white arrow).

carcinoma of the right breast. She completed her treatment 8 years ago, consisting of a partial right mastectomy as well as chemotherapy, radiotherapy, and hormone therapy. The patient didn't report any complaints in the past few years. Her physical examination didn't find any breast mass, skin modifications, nipple discharge, or abnormal lymph nodes. Blood tests didn't show any abnormalities. Her annual follow-up CT scan demonstrated an irregular mass of mixed density and spiculated margins in the junction of the inferior right quadrants. It was intimately adherent to the pectoralis muscle, far from the nipple (4 cm), and measured 25 × 39 mm (APxT). The right breast skin was slightly thick (6 mm) (Fig. 1). No other lesion was found. These findings suggested a cancer recurrence; hence, mammography and breast ultrasound were requested.

A mammogram demonstrated an irregular mass of increased density and spiculated margins in the posterior third of the right breast, adherent to the pectoralis muscle (Fig. 2).

Sonography showed a hypoechoic, partially attenuating mass with spiculated margins in the junction of the inferior right quadrants. The lesion measured 49 × 21 mm (TxAP) (Fig. 3).

No other lesions or lymphadenopathies were found. The left breast showed no abnormalities.

The right breast lesion was then classified as BI-RADS 4, and a core needle biopsy was requested.

The patient underwent 2 core needle biopsies; however, the 2 of them demonstrated equivocal pathologic findings. Therefore, a complete surgical excision of the mass was performed.

The histopathological examination findings were consistent with a cystic lymphangioma with negative surgical margins and no signs of malignancy.

Discussion

Cystic lymphangioma is defined as a vascular malformation rather than a true neoplasm. Congenital weakness of the vascular wall, proliferation of lymphatic vessels, and blockage of the lymphatic channels result in dilated lymphatic tissue that fails to communicate with the normal lymphatic system [1,6]. In addition, some instigating events, such as trauma or infection, may promote the development of cystic lymphangiomas [7]. These malformations are rarely encountered in adult patients, even less in the breast area [2,4]. Breast cystic lymphangiomas can be either diffuse or localized, and they are usually located in the upper outer quadrant or the subareolar space [1,2,4–8].

A few cases of secondary lymphangiomas after mastectomy and thoracic wall irradiation have been reported, although they described a cutaneous form known as lymphangioma circumscriptum [9,10].

Cystic lymphangiomas are usually asymptomatic. However, they may present with pain or discomfort, especially if bleeding or infected [1,5].

Radiologic evaluation of breast cystic lymphangiomas is no different from other breast lesions; it is based on a combination of mammography and sonography [5].

Mammographic presentation is commonly a round or lobulated, unique, or multiple masses with equal- to low-density compared to the underlying breast, which are usually interspersed between the normal parenchyma without obvious micro or macrocalcification [5,6,11].

Sonographic appearance can vary from the typical cystic multiloculated mass with multiple septations and clear con-

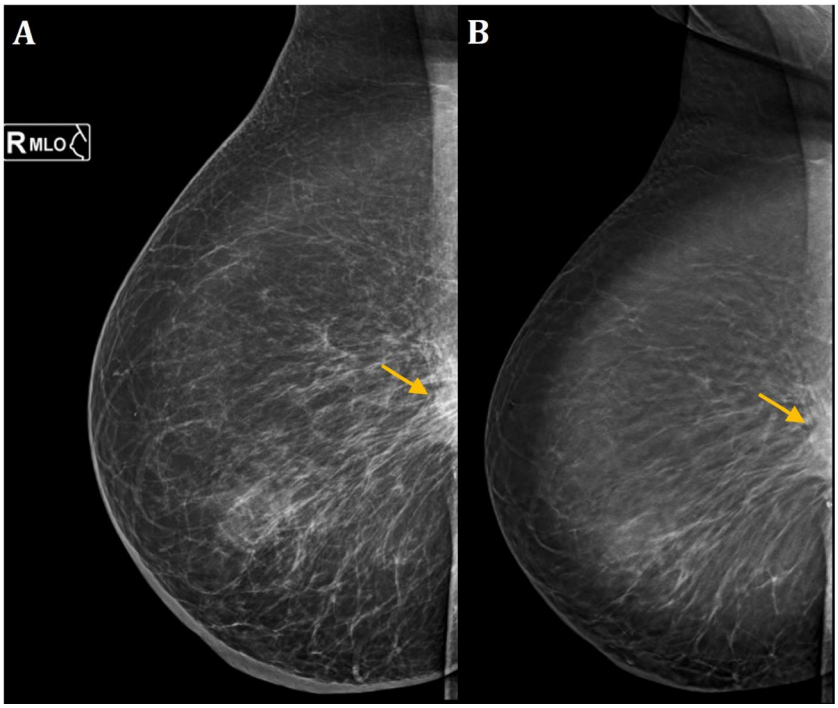

Fig. 2 – Conventional right breast mammogram (A) and tomosynthesis (B) images demonstrating an irregular mass of increased density and spiculated margins in the posterior third of the right breast, adherent to the pectoralis muscle (yellow arrow).

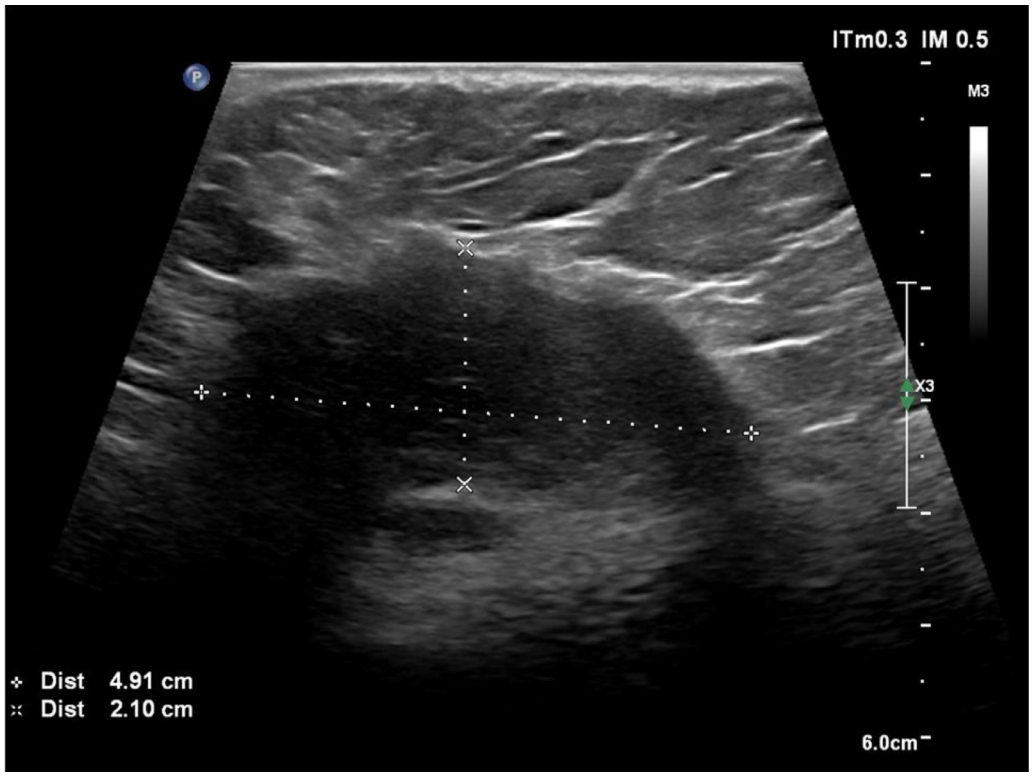

Fig. 3 – Right breast ultrasound image showing a hypoechoic, attenuating mass with spiculated margins in the junction of the inferior right quadrants.

tent to an ill-defined complex and irregular mass [5,6]. The latter presentation is due to the surrounding chronic fibrotic changes that appear hypoechoic and result in spiculated margins [6,11]. Such changes are often seen after surgery or radiation therapy, which explains the radiologic presentation of our patient's lesion.

MRI is performed to rule out an underlying malignancy in cases of suspicious mammographic or sonographic features. Moreover, it helps determine the lesion's extent [5,11]. On MRI, cystic lymphangiomas classically present as well-defined, multiseptated masses with fluid signal intensities and enhancing septa [2,5]. The fluid content is usually hypointense on T1-weighted images and hyperintense on T2-weighted sequences. However, it may demonstrate hyperintensity on both T1-weighted images and T2-weighted images when hemorrhagic or containing increased protein levels [5].

Fine-needle aspiration may assist in the diagnosis when it demonstrates lymphocyte-rich fluid. On the other hand, a core needle biopsy may be equivocal given the cystic nature of cystic lymphangiomas. Thus, the definitive diagnosis requires histopathology examination after complete resection with safe margins to avoid recurrences [1,4].

Conclusion

Despite being an extremely rare entity, cystic lymphangioma of the breast should be included in the differential diagnosis of a breast mass that appears after surgery. It is a benign condition; however, prompt diagnosis and complete resection are necessary to prevent recurrence and complications.

Patient consent

Written informed patient consent for publication has been obtained.

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