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CASE REPORT

Lymphoepithelioma-like carcinoma of the breast presenting as breast abscess

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Abstract

Lymphoepithelioma-like carcinoma (LELC) is a rare type of neoplasm in which only twenty cases have been reported in the breast. This type of tumor can be difficult to distinguish from other breast tumors particularly medullary carcinoma and lymphoma in the breast. We present a case of LELC of the breast presenting as an abscess along with a review of the literature. This is the 21st reported case of LELC of the breast and the first case to present as an abscess. Her clinical picture could have been mistaken for other infectious or inflammatory diseases. Given the potential for favorable outcome, early detection and general knowledge of this neoplasm are essential to expedite treatment for this rare tumor type.

Key words: Lymphoepithelioma-like carcinoma of the breast; Breast cancer; Breast abscess

Core tip: We present a case of lymphoepithelioma-like carcinoma (LELC) of the breast, which is a rare tumor type that can be difficult to diagnose. This particular case is the first case of LELC of the breast presenting as an abscess with radiologic and histologic studies as well as literature review of this rare tumor.

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INTRODUCTION

Lymphoepithelioma-like carcinoma (LELC) is an undifferentiated carcinoma composed of malignant epithelial cells with a lymphocytic background, which was first described in the nasopharynx by Regaud, Reverchon and Schminke. These cells have been described in other sites including the stomach, salivary gland, lung, thymus, skin and cervix[1]. LELC in the breast was first described by Kumar *et al*[2]. LELC of the breast is a rare disease with only 20 reported cases described in the literature[2-15]. Distinguishing LELC from medullary carcinoma and certain types of lymphoma has been a diagnostic challenge[3,14,15]. Making this distinction has profound impact on therapy and overall prognosis.

In this paper, we present a case of LELC of the breast in a 64-year-old female with an unusual presentation and clinical course.

CASE REPORT

A 64-year-old African American woman presented with a painful palpable lump in her left breast for four weeks.

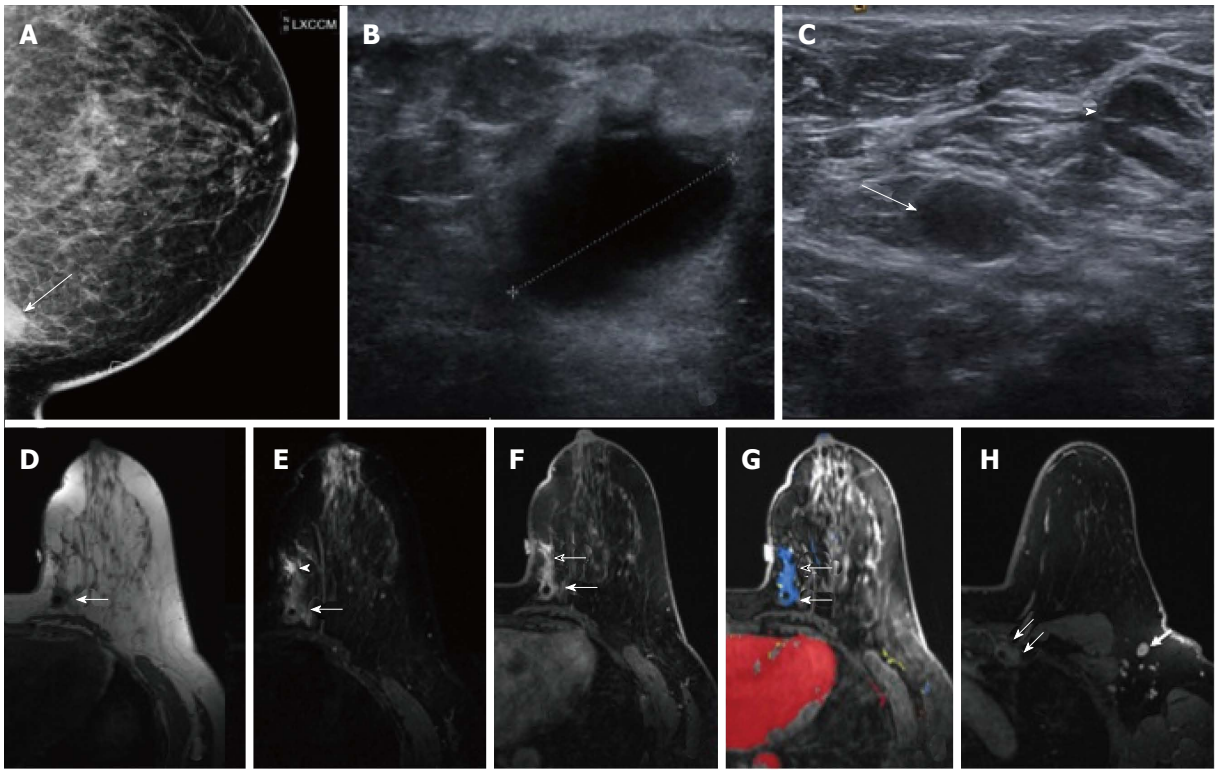

Figure 1 Imaging findings. A: Medially exaggerated cranio-caudal mammographic view of the left breast demonstrates the anterior aspect of the palpable mass that is a partially circumscribed and partially indistinct mass at 9 o'clock, posterior left breast (arrow); B: Ultrasound of the palpable lump demonstrates a 2.1 cm indistinct irregular hypoechoic solid mass with no posterior shadowing (caliper); C: Ultrasound of the left axilla demonstrates multiple oval and rounded lymph nodes measuring up to 1.4 cm. One oval 1.1 cm lymph node is diffusely hypoechoic (arrow) and is targeted for ultrasound guided Fine Needle Aspiration. The majority of the remaining lymph nodes demonstrate a thickened cortex (arrow head); D-H: Breast MRI: axial images of the left breast shown in multiple series as below; D: On pre-contrast T1W non-fat suppressed imaging the palpable mass is a 2 cm irregular mass with a central artifact from clip (arrow); E: On pre-contrast T2W fat suppressed imaging the mass is isointense to slightly hyperintense relative to the breast parenchyma (arrow). Similar signal is noted anterior to the mass with a higher intensity area more anteriorly (arrowhead); F: On first post-contrast fat suppressed T1W imaging the 2 cm irregular mass with a clip (arrow) is enhancing. Additional non-mass enhancement is noted anterior to the mass, measuring approximately 3 cm in the anterior-posterior plane (open arrow); G: Computerized color mapping demonstrates mostly persistent kinetic pattern of the mass and non-mass enhancement (area in blue, arrows); H: First post-contrast fat suppressed T1W imaging confirms the presence of multiple abnormal level 1 (arrow) and level 2 (not shown) axillary lymph nodes and abnormal left internal mammary chain adenopathy (double arrows).

She was initially seen by her primary care physician who prescribed antibiotics without any improvement. She subsequently underwent a mammogram, which demonstrated the anterior aspect of an ill-defined density located in the most posterior medial aspect of the left breast best seen on exaggerated cranio-caudal view (Figure 1A). This mass showed no associated focal architectural distortion or cluster of microcalcification. Her prior annual mammograms were all negative 4 years in a row. Ultrasound revealed an irregular markedly hypoechoic mass with indistinct margins at the 9 o'clock position of the left breast measuring approximately 2.1 cm in maximal diameter (Figure 1B). On color Doppler, no internal vascularity was detected. There were multiple enlarged left axillary lymph nodes detected on both mammogram and ultrasound (Figure 1C). The mass was classified as highly suggestive of malignancy according to the American College of Radiology Breast Imaging Reporting and Data System (ACR BI-RADS: 5). The patient subsequently underwent ultrasound-guided core needle biopsy of the left breast mass and a fine needle aspiration of the two dominant left axillary lymph nodes. Her initial biopsy specimen was described as an abscess-like necrotic tissue but there were atypical cells that were highly suspicious for necrotic malignancy. The fine needle aspirations of the two enlarging axillary lymph nodes were negative for malignancy. This suspicious mass demonstrated low T1 and high T2 signal intensity on the pre-contrast imaging on a subsequent breast MRI (magnetic resonance imaging) with the susceptibility artifact from a biopsy clip (Figure 1D-F). After intravenous administration of gadolinium (Omniscan, GE), the mass showed irregular margins and enhancement, initially rapid and subsequently persisted (type Ⅲ curve) (Figure 1F-H). Additional linear non mass-like enhancement with similar kinetic pattern was noted anterior to the mass, near the palpable marker, which could represent both tumor extension or post biopsy change. Her right breast showed no suspicious finding. Multiple abnormal enlarged and enhancing level 1 and level 2 left axillary lymph nodes and left inter-pectoral lymph nodes were detected, measuring up to 1.4 cm. There was also a 3.0 cm enhancing soft tissue mass with irregular margins in the left parasternal region, suggestive of left internal mammary adenopathy.

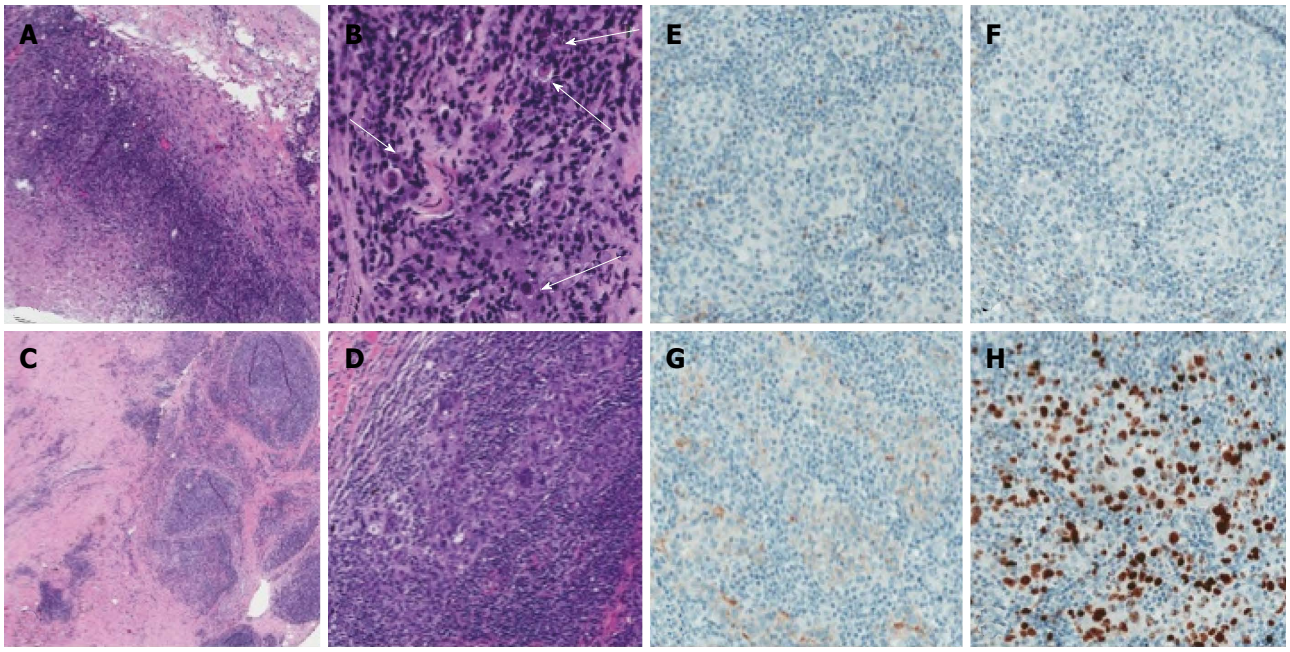

Figure 2 Pathology findings. A, B: Breast, core needle biopsy: large area of necrosis with chronic inflammation and fibrosis at periphery of lesion, 4X magnification (A); 16X magnification of A showing rare, atypical cells (arrows, B); C, D: Breast, surgical excision: poorly circumscribed lesion adjacent to previous biopsy site, 10X magnification (C); high grade carcinoma with marked pleomorphism and syncytical growth somewhat obscured by marked intra- and peri-turmoral inflammatory infiltrate (D) characteristic of LELC, 10X magnification; E-H: Immunophenotype: ER negative (E), PR negative (F), Her-2/neu 1+ (G), and Ki-67 27% (H). 10X magnification. ER: Estrogen-receptor; PR: Progesterone-receptor.

Subsequently, the patient underwent left lumpectomy with axillary node dissection. Positron emission tomograph scan showed no evidence of distant metastases. Her final pathology was consistent with stage IIA (pT1c, N1a, M0) lymphoepithelioma-like carcinoma (Figure 2A-D) with three out of twenty-three lymph nodes involved along with a small focus of extracapsular extension. Additional immunohistochemistry staining (Figure 2E-H) demonstrated that the tumor was negative for both estrogen-receptor and progesterone-receptor (ER 0%, PR 0%) and there was no overexpression of human epidermal growth factor receptor 2 (1+). The staining for proliferative index or Ki67 was 27%. The patient received adjuvant chemotherapy with dose dense doxorubicin and cyclophosphamide followed by weekly paclitaxel and a course of adjuvant radiation therapy.

Her clinical course was complicated by persistent inflammation and serosanguinous drainage from the surgical site. She was given antibiotics on multiple occasions for presumed cellulitis with minimal improvement. Follow-up punch skin biopsy and imaging done of the area showed no evidence of recurrence. These skin changes at the surgical site were thought to be secondary to post-surgical changes and radiation treatments. At her 3-year follow-up, the patient was doing well with no evidence of recurrent disease.

DISCUSSION

Lymphoepithelioma-like carcinoma of the breast is a rare tumor type characterized by epithelial neoplastic cells with a background of lymphocytic cells. Only twenty cases of this type of breast neoplasm have been described in the literature. The reported cases are summarized in Table 1. Making the diagnosis of LELC can pose a diagnostic challenge due to its morphologic similarities with medullary carcinoma and certain types of lymphoma on pathologic examination[5,14,15]. Making the distinction between LELC-B and other histologically similar tumors has significant impact on therapy and prognosis. Medullary carcinoma of the breast as described by Rapin and Ridolfi includes the following features: syncytial growth pattern > 75%, complete circumscription, diffuse mononuclear stromal infiltrate, moderate to marked nuclear pleomorphism and absence of microglandular features[16,17]. LELC of the breast has similar features, but specifically obscures the neoplastic cells[13]. Though poorly differentiated carcinomas, both medullary carcinomas and LELC-B consistently express cytokeratin markers[12,18]. In our case, the neoplastic cells expressed CK7 and CK5/6, which differentiate these entities from a large cell lymphoma, which may also be on the histologic differential. Additionally, the extensive lymphoplasmacytic infiltrate associated with the epithelial cells may raise concern for a lymphoepithelial lesion, indicating a small cell lymphoma. Special studies for kappa and lambda expression by the B lymphocytes and plasma cells associated with medullary carcinoma and LELC-B show polytypic cell population with expression of both kappa and lambda light chains. In situ hybridization for kappa and lambda light chains was performed in our case, showing a kappa-to-lambda ratio of approximately 2:1, ruling out a

Table 1 Summary of reported cases of lymphoepithelioma-like carcinoma of the breast

Case	Ref.	Year	Age	Presenting problem	Size (cm)	Lymph node (Y/N)	Surgery	Chemotherapy (Y/N) (agent listed if known)	Radiation therapy (Y/N)	Outcome (mo)[1]	ER status	PR status	Her2 status	EBV
1	Kumar *et al*[2]	1994	65		2	N	Mastectomy ALND	NR	NR	7	+	+	NR	NR
2	Cristina *et al*[3]	2000	54	Mass	1.5	N	Quadrantectomy ALND	Y	N	6	+	-	-	-
3	Dadmanesh *et al*[4]	2001	43	NR	1.9	Y	Quadranectomy	N	N	60	-	-	-	NR
4	Dadmanesh *et al*[4]	2001	53	NR	2	N	NR	N	N	72	-	-	-	NR
5	Dadmanesh *et al*[4]	2001	49	NR	1	N	Quadranectomy	N	N	[2]	-	-	-	NR
6	Dadmanesh *et al*-4[4]	2001	52	NR	2.7	N	Quadranectomy	N	N	36	+	-	-	NR
7	Dadmanesh *et al*[4]	2001	64	NR	2	N	Mastectomy	N	N	60	-	-	-	NR
8	Dadmanesh *et al*[4]	2001	69	NR	2.3	N	Mastectomy	N	Y	48	-	-	-	NR
9	Naidoo *et al*[10]	2001	50	Mass	2.5	Y	Wide local excision ALND	N	N	3	NR	NR	NR	-
10	Peştereli *et al*[13]	2002	56	Mass	1.9	Y	Modified radical mastectomy ALND	Y	N	12	+	+	-	-
11	Ilvan *et al*[6]	2004	59	Mass	3.5	N	Wide local excision ALND	Y (Tamoxifen)	Y	52	+	+	NR	-
12	Ilvan *et al*[6]	2004	67	Mass	1.1	N	Quadrantectomy ALND	N	Y	46	+	+	NR	-
13	Sanati *et al*[15]	2004	62	Mass	3	NR	NR	NR	NR	36	+	-	-	
14	Kurose *et al*[9]	2005	47	Mass	2.8		Total mastectomy ALND Tamoxifen	Y (CEF)	N	12	+	+	+	-
15	Saleh *et al*[14]	2005	51	Mass	2	Y	Lumpectomy ALND	N	N	NR	-	-	NR	-
16	Kulka *et al*[8]	2008	42	Mass	2.5	N	Lumpectomy	N	Y	NR	+	-	-	-
17	O'Sullivan-Meija *et al*[12]	2009	55	Abnormal mammo	2	N	Mastectomy	Y (Trastuzumab)	Y	22	-	-	+	-
18	Jeong *et al*[7]	2010	37	Mass	2.2	N	Modified mastectomy	Y	N	23	-	-	-	-
19	Dinniwell *et al*[5]	2012	55	Mass, tenderness	4	N	Excisional biopsy	N	Y	36	-	-	-	-
20	Nio *et al*[11]	2012	45	Mass	3	N	Quadrantectomy ALND	Y	Y	NR	-	-	-	NR
21	Present case	2012	64	Mass, tenderness	2	Y	Partial mastectomy ALND	Y (AC + paclitaxel)	Y	36	-	-	+	NR

[1]Time after initial surgical procedure; [2]Contralateral (right) LELC 3 years after initial diagnosis. ALND: Axillary lymph node dissection; NR: Not reported; CEF: Cyclophosphamide, epirubicin hydrochloride, 5FU; AC: Doxorubicin and cyclophosphamide; LELC: Lymphoepithelioma-like carcinoma; ER: Estrogen-receptor; PR: Progesterone-receptor; EBV: Epstein-Barr virus.

monotypic B cell population.

Although not tested in our patient, Epstein-Barr virus (EBV) and human papilloma virus (HPV) have been cited for their possible association with LELC of the breast. EBV has been linked to Burkitt's lymphoma and nasopharyngeal carcinoma. To date, EBV has not been shown to be associated with LELC of the breast; however, LELC in other anatomic sites namely salivary glands, sinonasal tract, stomach, thymus and lungs have been associated with EBV positivity[1,19]. In addition, HPV has been associated with two cases of LELC-B[8,11]. Another case has been seen with sclerosing lymphocytic lobulitis[10].

The case presented here, to the best of our knowledge, is the first case of lymphoepithelioma-like carcinoma of the breast to present as an abscess. Many of the prior case reports of LELC of the breast focus on the distinguishing histopathologic features of the disease and how to distinguish it from morphologically similar entities. Our case illustrates an unusual clinical presentation of this rare tumor type. Our patient presented with a painful breast lump and tenderness that could have easily been mistaken for an infection or other inflammatory processes which would have lead down a completely dif-

ferent diagnostic path and ultimately a significant delay in appropriate treatment. The patient's tumor also appeared to have necrotic and abscess-like features on initial pathologic examination, which has not been described in other cases of LELC. Initial clinical presentations were reported in 13 of the 20 cases. Twelve patients (60%) presented with a palpable mass while only 1 patient had abnormal findings on mammography. Her clinical course was also complicated by recurrent cellulitis and inflammation of the involved site minimally responsive to antimicrobial therapies. These changes may have been secondary to surgery and radiation therapy but did not appear to be consistent with recurrence of her primary disease.

All reported cases of LELC of the breast (Table 1) were found in women, ranging in age from 37 to 69 years. The median age was 54 years at time of presentation. The tumor sizes ranged from 1 to 4 cm and lymph node involvement seen in 25% of the patients including our patient. ER and PR status were evaluated in all but one patient (19 patients). ER was positive in nine patients (47%) and PR was positive in five patients (26%). Her-2 receptor status was reported in fifteen patients and was found to be overexpressed in three patients (20%).

Our patient regularly had annual mammography. Her last screening mammogram was performed six months prior to the detection of the palpable mass which showed no suspicious findings. However, the mass was probably not included in the field of view because of its very posterior location in the medial breast. This type of breast neoplasm appears to have overall favorable prognosis but early detection is imperative for proper treatment. After review of the literature, management of our patient went in line with most of the prior LELC-B cases. Seventeen of the reported cases mentioned some type of surgical intervention including mastectomy (complete or partial) in ten patients (59%) and axillary node dissections in nine patients (53%). Seven patients (34%) received some type of adjuvant chemotherapy and radiation therapy. Our patient underwent partial mastectomy and axillary node dissection followed by adjuvant chemotherapy along with radiation therapy. Disease-free outcome duration ranged from 2 to 72 mo in the cases reviewed while our patient was doing well without evidence of recurrent 36 mo after primary surgery.

In conclusion, this case and other cases of LELC-B demonstrate the diagnostic challenges for this particular type of tumor mainly due to its morphologic similarities with other neoplasms and rarity of the tumor. This is the first reported case of LELC of the breast presented with abscess-like clinical and pathologic features. Early recognition and appropriate diagnostic workup is the key for optimal management for this particular type of tumor.

COMMENTS

Case characteristics

A 64-year-old African American woman presented with a painful palpable lump.

Clinical diagnosis

Palpable mass of left anterior breast.

Differential diagnosis

Breast cancer, breast cellulitis/abscess.

Imaging diagnosis

Ultrasound showed an irregular markedly hypoechoic mass with indistinct margins at the 9 o'clock position of the left breast measuring approximately 2.1 cm in maximal diameter.

Pathological diagnosis

Her final pathology was consistent with stage IIA (pT1c, N1a, M0) lymphoepithelioma-like carcinoma. Additional immunohistochemistry staining demonstrated that the tumor was negative for both estrogen and progesterone receptors (ER 0%, PR 0%) and there was no overexpression of human epidermal growth factor receptor 2 (1+). The staining for proliferative index or Ki67 was 27%.

Treatment

The patient received adjuvant chemotherapy with doxorubicin and cyclophosphamide followed by weekly paclitaxel and a course of adjuvant radiation therapy.

Related reports

Lymphoepithelioma-like carcinoma (LELC) of the breast is a rare tumor type and the case presented is the first case of LELC of the breast presenting as a breast abscess.

Term explanation

LELC is an undifferentiated carcinoma composed of malignant epithelial cells with a lymphocytic background.

Experience and lessons

This case report emphasizes the importance of having knowledge of rare tumor types, particularly those in which early recognition can have profound impact on treatment outcome.

Peer review

It is an interesting and rare case of overlapping features where the diagnosis hangs between abscess and a tumour. The diagnosis is critical for appropriate management.

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