

Rare Presentation of Synchronous Accessory Breast Carcinoma in Abdominal Wall - A Case Report and Review of the Literature

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Rezumat

Prezentare rară a carcinomului mamar accesoriu sincron în peretele abdominal - raport de caz și o trecere în revistă a literaturii de specialitate

Malignitatea primară în țesutul mamar accesoriu este un diagnostic rar, reprezentând mai puțin de 1% din totalul cancerelor de sân și se prezintă cel mai frecvent cu o masă palpabilă la nivelul axilei. În plus, un al doilea cancer mamar primar concomitent, găsit în țesutul mamar accesoriu, este un fenomen extrem de rar, deși rapoartele despre acestea sunt în creștere. Locurile extramamare în care au fost raportate tumori maligne mamare primare secundare includ pliul inframamar, abdomenul și chiar vulva. Atunci când se suspectează o malignitate mamară ectopică, evaluarea triplă este abordarea standard a investigației. Cu toate acestea, nu există ghiduri specifice de tratament și, prin urmare, se respectă aceleași ghiduri ca și în cazul cancerului mamar pectoral. Aici, prezentăm un raport despre o femeie în vârstă de 71 de ani, tratată pentru un cancer mamar stâng, diagnosticată cu un al doilea carcinom mamar primar concomitent în cadranul superior drept al peretelui abdominal, care a fost tratat chirurgical cu excizie locală largă și biopsie a ganglionului limfatic santinelă. Acest caz evidențiază importanța menținerii unui indice ridicat de suspiciune în timpul evaluării acestor pacienți pentru a stabili diagnosticul corect într-un stadiu incipient.

Cuvinte cheie: sân accesoriu, tumoră a peretelui abdominal, axilă, cancer mamar

Abstract

Primary malignancy in accessory breast tissue is an infrequent diagnosis representing less than 1% of all breast cancers and most commonly presents with a palpable mass in the axilla. Furthermore, a concurrent second primary breast cancer found in accessory breast tissue is an extremely rare phenomenon, although reports of these are on the rise. Extra-mammary sites where second primary breast malignancies have been reported include the inframammary fold, the abdomen and even the vulva. When an ectopic

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breast malignancy is suspected, triple assessment is the standard approach to investigation. However, there are no specific treatment guidelines, and it therefore follows the same guidelines as for pectoral breast cancer. Here, we present a report of a 71-year-old female being treated for a left breast cancer, diagnosed with a concurrent second primary breast carcinoma in the right upper quadrant of the abdominal wall, which was surgically managed with wide local excision and sentinel lymph node biopsy. This case highlights the importance of maintaining a high index of suspicion during assessment of these patients in order to establish the correct diagnosis at an early stage.

Keywords: Accessory breast, abdominal wall tumour, axilla, breast cancer

Introduction

Ectopic breast tissue is a common congenital anomaly characterized by the presence of accessory breast tissue along the embryonic mammary line, that develops between the fourth and sixth weeks of gestation extending bilaterally from the axilla to the groin on the ventral side of the body (1). It has been reported to occur in up to 6% of the population. The theory behind its development is widely explained by the failure of the remnant tissue at the milk line to regress during the formation and development of the human embryo, which usually occurs along the whole line except in the thoracic area, the site where the anatomic (pectoral) breast tissues will develop (1). Morphologically, it can present with any of the basic elements of the normal breast: the parenchymal tissue, the areola or the nipple. Accessory breast tissue has traditionally been classified according to the Kajava classification (2), which categorizes ectopic breast tissue based on the presence or absence of glandular tissue, nipple, and areola. Although this classification has limited impact on contemporary surgical management, it remains useful for describing the wide spectrum of accessory breast tissue presentations encountered in clinical practice.

Because accessory breast tissue contains normal mammary elements, it can undergo any of the changes seen in the anatomic breast as it remains hormonally responsive and is capable of undergoing the full spectrum of physiological changes associated with puberty, pregnancy, lactation, and menopause, as well as benign and malignant pathological transformation (3). Malignancy in ectopic breast tissue has been reported but is considered markedly rare, with a percentage less than 1% among breast cancer cases (4).

On review of the literature, the majority of the data pertaining to the presentation and treatment of carcinoma in accessory breast tissue are retrieved from case reports. Hence, there may be limited

awareness of the condition and a lack of distinct guidelines for the investigation and management of this entity. In this article, we present a case of primary breast invasive ductal carcinoma of upper abdominal wall diagnosed during the treatment of a distinct cancer in the contralateral breast and treated with wide local excision and sentinel lymph node biopsy.

A literature review was conducted using PubMed/MEDLINE and Google Scholar. English-language case reports, case series, and review articles on accessory breast carcinoma were included.

Case Report

A 71-year-old woman presented to the symptomatic breast unit at a tertiary referral center outpatient department with a palpable left breast mass for 3 weeks. It was not associated with any breast pain, skin changes or nipple discharge. There was no significant family history of breast or ovarian cancer. She was on hormonal replacement therapy for 5 years.

Clinical examination confirmed the presence of a suspicious mass in the left upper quadrant of the breast measuring about 20 x 30 mm in diameter, with no associated skin tethering or fixation and no nipple discharge or inversion. An incidental hard subcutaneous lesion was noted on the right upper abdominal region measuring approximately 30 mm. No palpable axillary lymphadenopathy was identified.

Radiological investigations included bilateral mammography and an ultrasound of the left breast and the abdominal wall lesion. The mammogram reported a 23-mm BI-RADS 5 lesion in the left breast, confirmed on ultrasound (*Fig. 1*). Ultrasound of the right upper abdominal wall reported a non-suspicious subcutaneous lesion.

Image guided core needle biopsy of the left breast lesion was performed which confirmed an invasive ductal carcinoma, grade 2, associated with ductal carcinoma in situ- cribriform subtype with immuno-

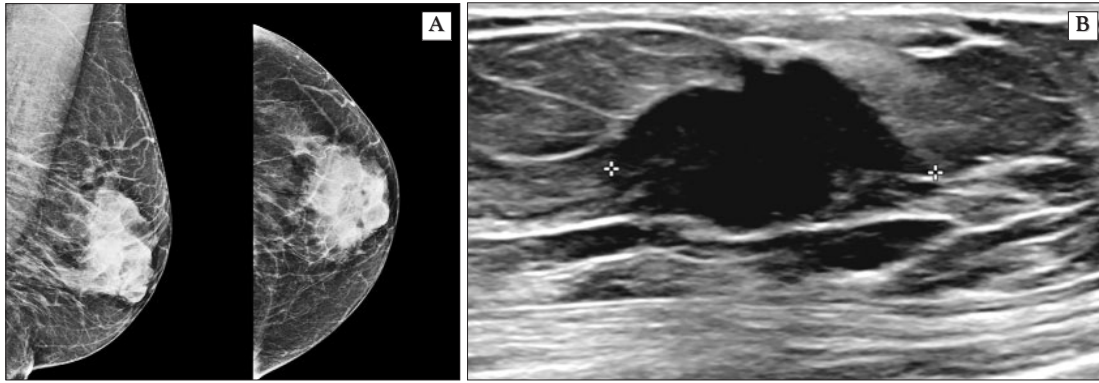


Figure 1. (A) Mammographic images of the left breast lesion, demonstrating the mediolateral oblique (MLO) view on the left and the craniocaudal (CC) view on the right. (B) Ultrasound (US) image of the left breast lesion.

histochemistry for receptors demonstrating positive ER and PR receptors and a negative HER2/neu receptor status.

Following multidisciplinary team discussion, the patient proceeded to primary breast conserving surgery with a left-sided wide local excision and sentinel lymph node biopsy (SLNB). The right-sided abdominal subcutaneous lesion was also excised at this time. As expected, left breast histopathology

confirmed a 25-mm invasive ductal carcinoma, grade 2, ER positive (8/8), PR positive (6/8), and HER2/neu receptor negative (score 1+). A 9.5-mm macrometastasis was identified in 1 of 2 sentinel lymph nodes.

Histological assessment of the subcutaneous lesion excised from the right abdominal wall revealed an invasive carcinoma of no special (ductal) type, grade 2, arising from an encapsulated papillary carcinoma within accessory breast tissue (*Fig. 2*). The overall

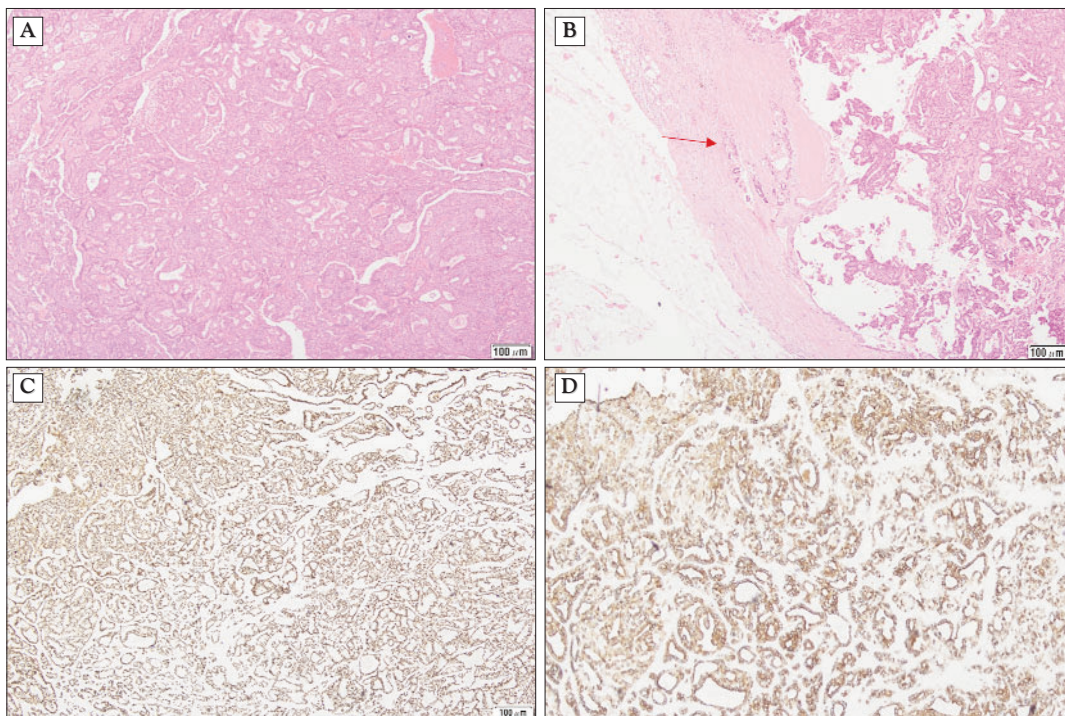


Figure 2. Histopathological and immunohistochemical features of the lesion. (A) Low-power photomicrograph ($\times 10$) showing a partially encapsulated atypical glandular proliferation consistent with encapsulated papillary carcinoma. (B) Low-power photomicrograph ($\times 10$) demonstrating a focal area of stromal invasion (red arrow). (C) Immunohistochemical staining for GATA3 ($\times 10$) showing diffuse nuclear positivity in the atypical cells. (D) Immunohistochemical staining for CK7 ($\times 10$) demonstrating diffuse cytoplasmic positivity in the atypical glandular cells

tumour size was 19 mm and extended to all margins. It was ER positive (8/8), PR positive (7/8), and HER2 negative (score 1+).

Following discussion of this histopathology with the multidisciplinary team, the patient proceeded to further investigation with an MRI of both breasts and axillae – which did not identify any further pathology, followed by a re-excision of margins and sentinel lymph node biopsy. The final histopathology of the accessory breast carcinoma demonstrated adjacent glandular breast parenchyma with fibrocystic changes. There was no evidence of residual or recurrent malignancy. Three sentinel lymph nodes were identified and were negative for metastatic carcinoma.

Adjuvant radiotherapy was administered to the left breast and level I axillary lymph nodes at a dose of 40 Gy in 15 fractions, followed by a boost of 11.25 Gy in 5 fractions. No radiotherapy was delivered to the accessory breast cancer site. Oncotype DX testing was performed on both tumours with a result of 13 for the left-sided breast cancer and 0 for the accessory breast cancer. Adjuvant anti-estrogen therapy was commenced. The postoperative course was uneventful, and at 24 months of follow-up, the patient demonstrated no clinical or radiological evidence of local recurrence or distant disease. Surveillance comprised annual bilateral mammography and regular clinical examination of the previous accessory breast cancer site, while endocrine therapy was continued without significant adverse effects.

Discussion

Although accessory breast tissue is most commonly located in the axilla, several other locations have been reported in the literature, some of which are positioned outside of the milk line. These include the face, posterior neck, chest, middle back, buttock and the vulva in females (1). Of note, the diagnosis of cancer in ectopic breast tissue can be extremely difficult even in close proximity regions such as the axilla. The diagnostic challenge is impacted by the rarity of the disease, a lack of clinical awareness and the broad range of differentials. Typically, a patient will present with a palpable lesion and other pathologies must be considered in the differential including primary lymphoma and a metastatic tumour arising from an occult primary lesion. In this particular case, as the palpable lesion was located in the subcutaneous tissue of the abdominal wall, the differential also included cutaneous and subcutaneous nodules including dermoid and epidermoid cyst.

Abdominal wall masses often have a broad differential diagnosis, with benign conditions including

lipomas, sebaceous cysts, and various inflammatory processes being more common in clinical practice. Unfortunately, these factors might lead to delayed diagnosis in such patients (5). This fact about confounding differential diagnoses has previously been emphasized in a case of a primary breast cancer in an ectopic breast reported by Joana Marques-Antunes and colleagues, where a 49-year-old woman had an invasive lobular carcinoma in an accessory breast on the left axilla which was initially diagnosed as a sebaceous cyst, hence the lesion was excised under local anesthesia. Only upon final histopathological assessment it was found to be breast tissue widely infiltrated by an invasive carcinoma which necessitated further excision of the remnant tissue with axillary lymph node dissection. The authors emphasized the importance of careful assessment and attention to the presence of any abnormal mass along the mammary line (6).

Invasive ductal carcinoma is the most common malignancy arising in accessory breast tissue, although other histological subtypes, including lobular, medullary, mucinous carcinoma and phyllodes tumour, have also been reported (1). Our patient had the uncommon finding of an invasive ductal carcinoma arising within an encapsulated papillary carcinoma, a rare entity accounting for only 0.5–1% of all breast carcinomas (7). Although the abdominal wall lesion appeared benign on ultrasound and was contralateral to the known breast cancer, its location along the embryonic milk line prompted surgical excision. The distinct histopathological features compared with the left breast invasive ductal carcinoma confirmed that these represented two separate synchronous primary breast malignancies.

The presence of an accessory breast cancer concurrent with bilateral invasive breast cancer is extremely rare but has been documented in literature, where two cases from China were reported by Jin-yan Hao and colleagues in 2012. Both patients had bilateral invasive breast cancers with another primary tumour found in the accessory breast tissue of the axilla (8). In 2021, a case from the USA highlights the importance of bilateral assessment, where Addae and his colleagues reported on a 60-year-old female patient who presented five years after her initial diagnosis and treatment of left breast cancer. She had a new mass in the contralateral axilla which histologically proved to be a second primary breast cancer (5).

Inflammatory breast cancer has been also observed in upper abdominal accessory breast tissue in a woman with a personal history of ipsilateral breast cancer by Miles et al (9). The patient presented 9 years after her treatment of breast cancer with localized

erythema that failed to respond to antibiotics. She was treated with neoadjuvant chemotherapy followed by surgical resection and SLNB.

Recent reports further illustrate the heterogeneity of accessory breast carcinoma. Zhong et al. (10) described a primary invasive ductal carcinoma arising in accessory breast tissue of the upper abdominal wall in a male patient, while Eguchi et al. (11) reported an inframammary accessory breast carcinoma successfully managed with wide local excision and sentinel lymph node biopsy. More recently, Chen et al. (12) described synchronous ipsilateral accessory breast carcinoma and ductal carcinoma in situ of the pectoral breast. Despite differences in anatomical location and tumour biology, these reports, together with the present case, support management according to established breast cancer principles. Because of the rarity of accessory breast carcinoma, comprehensive molecular characterization remains limited. Immunohistochemical assessment of estrogen receptor (ER), progesterone receptor (PR), and HER2 status is therefore performed using the same standards as pectoral breast cancer, allowing molecular classification and guiding systemic therapy according to established breast cancer guidelines (13).

Table 1 summarises the published cases identified in our literature review. The present case represents a rare combination of synchronous contralateral breast cancer and primary accessory breast carcinoma of the upper abdominal wall.

Surgical management of breast cancer has evolved; treatment has shifted from radical mastectomy to wide

local excision (14). The same surgical principles apply to accessory breast cancers. With the accumulation of increasing clinical evidence from reported cases, ipsilateral mastectomy is no longer recommended, as it has not been shown to improve prognosis. It is proposed by Madej et al, that unless the accessory breast cancer is very close to or connected with the breast there is no requirement for ipsilateral mastectomy (15). In our case, the abdominal wall tumour was at least 3 cm caudal to the inframammary fold and so surgical wide local excision was deemed to be sufficient after the breast had thorough radiologic assessment with MRI.

For the axilla; sentinel lymph node biopsy can be considered in early stage with no lymphatic involvement and axillary dissection is performed if found positive (6), although upfront axillary dissection was elected by some surgeons in cases of BC in axillary tissue due to the proximity of the malignancy to the axillary node and was also performed in case of the presence of a suspicious node in preoperative imaging (16). Sentinel lymph node biopsy was successfully performed in our case, with no evidence of axillary nodal disease.

Further treatment is individualized according to tumour characteristics and patient factors, with chemotherapy, radiotherapy, and endocrine therapy determined following multidisciplinary team discussion. In patients with early-stage, ER-positive, HER2-negative disease, the use of the Oncotype DX assay may assist in guiding adjuvant chemotherapy decision-

Table 1. Clinicopathological characteristics of published cases of accessory breast carcinoma included in the present literature review

Author	Year	Country	Age/Sex	Accessory breast location	Histology	ER/PR/HER2	Synchronous pectoral breast cancer	Treatment
Hao et al.	2012	China	Case 1: 43/F Case 2: 53/F	Axilla	IDC	Case 1: ER+, PR+, HER2+. Case 2: ER-, PR-, HER2+	Yes (bilateral invasive breast cancers)	Case 1: Surgery. Case 2: Neoadjuvant chemotherapy + Surgery + adjuvant chemotherapy.
Zhong et al.	2018	China	62/M	Upper abdominal wall	IDC	ER+, PR+, HER2-	No	Surgery + radiotherapy + endocrine therapy
Miles et al.	2017	USA	58/F	Upper abdominal wall	IDC (Inflammatory breast cancer)	ER+, PR-, HER2-	Previous ipsilateral breast cancer	Neoadjuvant chemotherapy + surgery + radiotherapy + endocrine therapy
Addae et al.	2021	USA	60/F	Axilla	IDC	ER+, PR+, HER2-	Previous contralateral breast cancer	Surgery + radiotherapy + endocrine therapy
Eguchi et al.	2021	Japan	68/F	Inframammary fold	IDC	ER+, PR+, HER2-	No	Surgery + endocrine therapy
Marques-Antunes et al.	2022	Portugal	49/F	Axilla	ILC	ER+, PR+, HER2-	No	Surgery + radiotherapy + endocrine therapy
Chen et al.	2024	China	64/F	Axilla	Invasive carcinoma with medullary features	ER-, PR-, HER2-	Yes (ipsilateral)	Surgery + chemotherapy
Present case	2026	Ireland	71/F	Upper abdominal wall	IDC arising in encapsulated papillary carcinoma	ER+, PR+, HER2-	Yes (contralateral IDC)	Surgery + endocrine therapy

IDC = invasive ductal carcinoma; ILC = Invasive lobular carcinoma .

making by estimating recurrence risk and potential chemotherapy benefit. Regarding surveillance, routine follow-up with annual mammography was performed in our case for the left breast. However, as the right-sided accessory breast carcinoma was located outside the standard mammographic field, follow-up should primarily include regular clinical examination, with imaging tailored to the anatomical location and clinical findings.

Prognosis appears to depend primarily on established breast cancer prognostic factors rather than the ectopic location itself. Tumour size, lymph node status, histological grade, hormone receptor expression, HER2 status, and stage at diagnosis remain the principal determinants of recurrence risk and survival (14,17). Our patient's tumour demonstrated favourable biological characteristics, including ER/PR positivity, HER2 negativity, and negative sentinel lymph nodes, supporting the decision to administer adjuvant endocrine therapy and contributing to her disease-free status at 24 months of follow-up.

Conclusion

A cutaneous lesion in the milk line requires judicious assessment as it could be accessory breast tissue that can develop any of the pathologic conditions that develop in the breast, including an invasive carcinoma. Any notable worrisome changes require appropriate further evaluation with imaging and histopathological studies for definitive diagnosis and appropriate treatment. Due to the infrequent occurrence, many physicians are unfamiliar with the clinical presentation of accessory breast cancer. This reported case highlights the importance of maintaining a high index of suspicion for another primary cancer in ectopic breast tissue in patients who present with a subcutaneous lesion at the milk line and clinicians should be aware of the possibility of malignancy in this tissue. Wide local excision and sentinel lymph node biopsy are advised if there is no lymph node involvement, otherwise axillary clearance is performed. Adjuvant treatment is usually directed by tumour characteristics and follows the same principles as those for breast cancer. Surveillance imaging may need to be tailored based on the location of the accessory breast cancer.

Conflicts of Interest

The authors confirm that there was no conflict of interest.

Consent

Informed consent was obtained from the patient prior to publication of this case report and accompanying images.

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