

Case report

Carcinoma of the axillary tail of Spence: A rare case report

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ABSTRACT

Introduction: Carcinoma arising from the axillary tail of Spence (CATS) is a rare entity that requires a high level of clinical suspicion. The clinicopathologic, prognostic, and imaging features of CATS are poorly understood.

Case summary: A 46-year-old woman presented to our hospital with right axillary swelling associated with bleeding and foul-smelling discharge. She had initially presented to a different hospital with a similar presentation where she was diagnosed with metastatic carcinoma, favouring adenocarcinoma on the FNAC report, following which she received 3 cycles of chemotherapy. MRI and PET-CT scan workups at our hospital showed a soft tissue mass likely arising from the axillary tail of the right breast with a few enlarged axillary lymph nodes and an unremarkable right breast. She underwent excision of the mass with axillary lymph node dissection and reconstruction with a pedicled Latissimus dorsi flap. The final diagnosis was based on immunohistochemistry, with tumor cells positive for GATA3 and CK-7, negative for estrogen and progesterone receptors (ER, PR), positive for human epidermal growth factor receptor 2 (Her2 neu), and having a Ki-67 labelling index of 45 %.

Discussion: Carcinoma of the axillary tail of Spence (CATS) has a reported incidence of 0.3 %. There have been only few papers till date that have reported the clinicopathologic, prognostic, and imaging features of CATS. MRI is an important imaging modality to localize CATS. Histologic examination aids in the diagnosis by identifying the presence of a histologic pattern of primary breast carcinoma and immunohistochemical characteristics such as ER, PR, Her2neu, and gross cystic disease fluid protein (GCDFFP). Due to the rarity of CATS, no specific guidelines concerning management currently exist. Surgical management involves wide local excision of the axillary mass with axillary lymph node dissection.

Conclusion: CATS, even though rare, should be considered in the differential diagnosis of a patient presenting with an axillary mass. MRI is a valuable tool to distinguish CATS from other lesions. Immunohistochemistry is essential to confirm the diagnosis.

1. Introduction

Breast cancer is the most common malignancy in females worldwide [1]. However, carcinoma arising from the axillary tail of Spence (CATS) is an extremely rare entity that develops at a specific anatomical position of the breast, with a reported incidence of 0.3 % [2]. It requires a high level of clinical suspicion to diagnose CATS. The important differential diagnosis for an axillary mass includes occult breast cancer with axillary lymph node metastasis, ectopic breast cancer, subcutaneous soft tissue tumor, benign breast tumor of the ATS, or metastatic lymphadenopathy originating from nonbreast tissue. There is limited information in the literature about the clinicopathologic, prognostic, and imaging features

of CATS. In this article, we report an unusual presentation of carcinoma arising from the axillary tail of the Spence (CATS) in the right axilla and its clinicopathologic characteristics. This case has been reported in line with the SCARE criteria [3].

2. Case summary

A 46-year-old perimenopausal woman of Indian origin, school teacher by profession, presented in our hospital outpatient department with right axillary swelling of 4 months duration, which was insidious in onset and gradually progressive in nature. The patient was in extreme discomfort due to associated pain, foul-smelling discharge, and

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intermittent episodes of bleeding from the mass. Three months before visiting us, she attended a different hospital with the same complaints where she underwent fine needle aspiration cytology from the axillary mass, which revealed metastatic carcinoma, favouring adenocarcinoma, following which she received 3 cycles of chemotherapy (Inj Epirubicin 130 mg and Inj Cyclophosphamide 900 mg) at 3-week intervals. The disease progressed while on chemotherapy, as there was an increase in the size of the axillary swelling with associated bleeding and ulceration. She had no associated medical comorbidities and had not undergone any surgical procedures in the past. There is no family history of malignancy. Her age at menarche was 14 years with regular menstrual cycles. She has two living children, with her age at first live birth being 22 years.

On general examination, the patient's performance score was ECOG = 1, vital signs were stable, and pallor was present. On local examination, the bilateral breasts and left axilla were normal with no palpable mass. A 7 cm × 6 cm ulceroproliferative firm, tender mass was palpable in the right axilla with limited mobility. There was overlying ulceration and bleeding. There was a limitation in the movement of the right shoulder. A 2-cm single, mobile, firm, non-tender level 1 axillary lymph node was also palpable. Other systemic examinations were normal.

Laboratory investigations revealed a haemoglobin level of 6 g/dl. We reviewed her previous slides in our institution, which also revealed metastatic carcinoma favouring adenocarcinoma. A whole body FDG-PET scan was performed to look for the primary site, showing an FDG avid large lobulated heterogeneously enhancing soft tissue mass (8.8 cm × 9.5 cm × 7 cm, SUV max 27.9) likely involving the axillary tail of the right breast, infiltrating the overlying skin and subcutaneous tissue of the right proximal arm, and FDG avid multiple discrete right axillary and subpectoral lymph nodes (largest: 2.4 cm × 1.4 cm, SUV max 13.6). Both breasts were normal, with no abnormal uptake elsewhere in the body. An MRI of the breast and axilla with contrast was performed to look for any additional lesions and the relationship of the axillary mass with the nearby major vessels, which showed a well-defined lobulated heterogeneously enhancing hyperintense lesion (10.6 cm × 9.7 cm × 7.6 cm) epicentered in the right axillary recess, probably arising from the axillary tail of the right breast, closely abutting the lateral chest wall muscles and axillary vessels, with few enlarged axillary lymph nodes. The breast was unremarkable [Fig. 1].

The patient underwent surgery after a multidisciplinary tumor board discussion, due to excessive discomfort and associated bleeding from the mass. Preoperative biopsy was not considered due to intermittent bleeding. To prepare the patient for surgery, 4 units of packed red blood cells were administered. The surgery involved a wide local excision of

the axillary mass with level 1, 2, and 3 axillary lymph node dissection. During the procedure, the mass was found to be adherent to the axillary vein, which made dissection difficult. The intercostobrachial nerve was sacrificed as it was densely adhered to the mass. However, the medial pectoral vessels, long thoracic nerve, thoracodorsal nerve, and thoracodorsal vessels were preserved. After excision, a pedicled Latissimus dorsi flap was used for reconstruction due to the large size of the defect. A drain was placed at the end of the procedure. The patient had an uneventful postoperative period and was discharged five days after the surgery when the drain was removed.

Histopathologic examination of the specimen showed tumor cells arranged in sheets, nests, and trabeculae, suggesting a poorly differentiated carcinoma, most likely an adenocarcinoma. The final diagnosis was based on immunohistochemistry, which revealed that the tumor cells were positive for GATA3 and CK-7, negative for estrogen and progesterone receptors (ER, PR), positive for human epidermal growth factor receptor 2 (Her2 neu) and had a Ki-67 labelling index of 45 %. These findings confirmed the diagnosis of a poorly differentiated invasive breast carcinoma, no special type, in the right axillary tail [Fig. 2]. The patient is planned for 6 cycles of adjuvant chemotherapy with paclitaxel, carboplatin, and trastuzumab, followed by radiotherapy and maintenance trastuzumab.

3. Discussion

Carcinoma of the axillary tail of Spence (CATS) has a reported incidence of 0.3 % [2]. The axillary tail of Spence (ATS) is considered a narrow extension of the upper quadrant of breast tissue that enters the axilla through a defect in the clavipectoral fascia, known as the foramen of Langer [4]. The axilla is also a common location for ectopic breast tissue, which is residual breast tissue that persists from normal embryologic development and is found in up to 6 % of the population [5]. In the 4th week of human development, a pair of mammary ridges derived from the ectoderm form. These ridges, often referred to as the "milk line," run bilaterally along the ventral surface of the body from the anterior axillary folds to the medial aspect up the inguinal folds. Normally, these ridges regress except at the site of the breasts [6]. Ectopic breast tissue represents the failure of regression in the milk line. Ectopic breast tissue does not show continuity with normal breast parenchyma, which differentiates it from ATS.

CATS is commonly found in patients over the age of 45 years. It is usually detected in the early stages and is characterized by a lack of hormone receptors [4,7,8]. Compared to upper outer quadrant (UOQ)

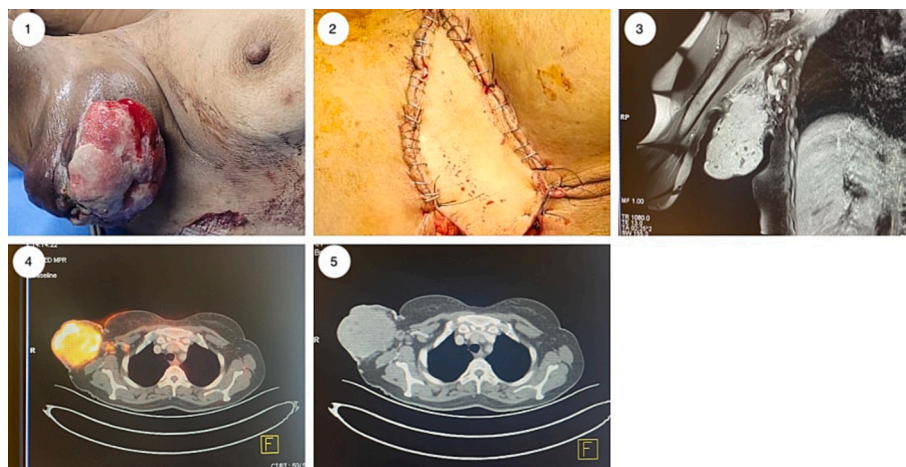


Fig. 1. 1. Preoperative image of right axillary mass; 2. Intraoperative image showing pedicled Latissimus dorsi flap reconstruction; 3. MR image showing a well-defined lobulated lesion in the right axillary recess with locoregional lymphadenopathy; 4. PET scan showing a large metabolically active heterogeneously enhancing soft tissue mass likely involving the axillary tail of the right breast with multiple right axillary and subpectoral lymph nodes; 5. CECT scan showed a soft tissue mass likely involving the axillary tail of the right breast.

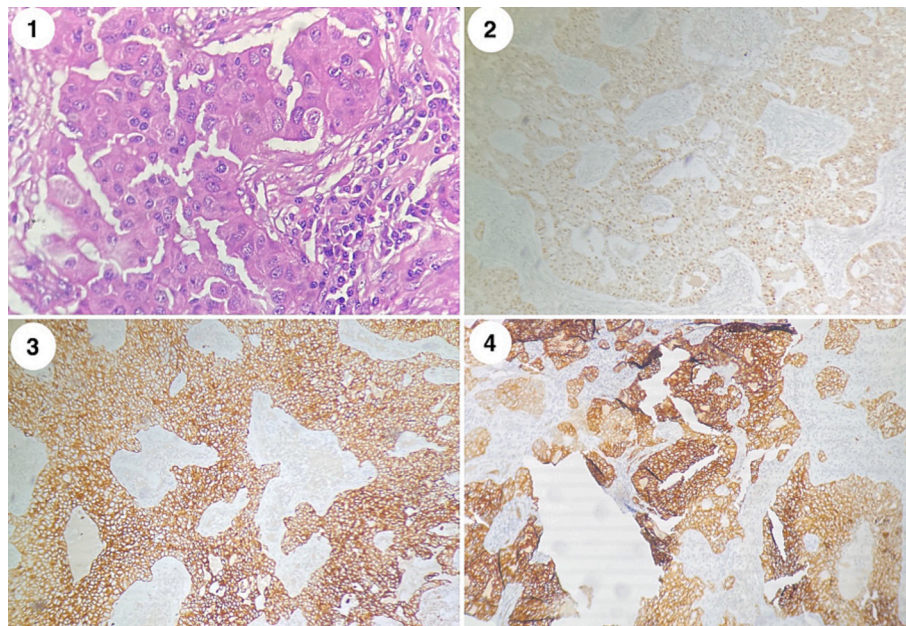


Fig. 2. 1. Tumor cells arranged in sheets, nests, and trabecular pattern (hematoxylin and eosin stain, $\times 400$); 2. GATA3 diffuse nuclear uptake in the tumor cells ($\times 400$); 3. Diffuse CK7 expression in the tumor cells ($\times 400$); 4. Strong membranous Her2-neu staining of the tumor cells ($\times 400$).

breast cancer, CATS has a higher grade, a higher rate of negative hormone receptors, and a greater tendency to spread to the lymph nodes, leading to a poorer prognosis [9]. In our case, a 46-year-old woman presented with advanced stage of CATS with bleeding and ulceration. Furthermore, her hormone receptor status was negative. As per our literature review, there are no reported cases of CATS presenting with ulceration and bleeding.

Diagnostic modalities include radiologic imaging followed by biopsy and histopathological diagnosis. MRI is a vital imaging method that can help in identifying CATS. A sagittal oblique reconstructed MRI can locate CATS by showing the continuity of the mammary tissue from the UOQ of the breast to the axilla [10]. An MRI performed at 1.5 T reveals a poorly defined mass that is restricted to the subcutaneous tissues of the axilla. The mass presents with varying signal intensities on T1-weighted images, composed of hyperintense and hypointense tissues. On fat-suppressed fast spin-echo T2-weighted images, the parenchyma is moderately hyperintense with regions of signal intensity consistent with fat. After IV contrast administration, they exhibit moderate enhancement on fat-suppressed T1-weighted images, similar to the parenchyma of the ipsilateral breast [11]. A PET scan can also aid in the preoperative diagnosis of CATS by demonstrating a metabolically active lesion in the axilla without any evidence of a primary breast malignant lesion [12]. In our case, contrast-enhanced MRI suggested a lesion in the right axilla with continuous mammary tissue from the UOQ breast to the axilla, confirming the location of ATS.

CATS is best diagnosed through histologic examination, which is considered the gold standard. This test helps identify the presence of a histologic pattern of primary breast carcinoma and immunohistochemical characteristics such as ER, PR, Her2neu, and gross cystic disease fluid protein (GCDFFP). In our case, we were able to confirm the diagnosis of CATS only after an immunohistochemical study of the specimen that revealed diffuse reactivity to GATA3, CK-7, and Her2Neu. The Ki67 labelling index was found to be 45 %, while both ER and PR tested negative.

Although the diagnosis of CATS was made based on the immunohistochemical study of the final specimen in our case, it's important to keep in mind the potential differential diagnoses when dealing with an axillary mass. CATS should be distinguished from ectopic breast cancer, occult breast cancer with axillary lymph node metastasis, subcutaneous

soft tissue tumor, benign breast tumor of the ATS, or metastatic lymphadenopathy originating from non-breast tissue. The significant differentiating factor between CATS and ectopic breast tissue is the continuity with the normal breast parenchyma. However, it can be difficult to identify the exact boundaries of the ATS and clavipectoral fascia on imaging. Therefore, MRI is a vital imaging modality that can help to localize CATS. It is essential to differentiate CATS from other diagnoses, such as UOQ breast tumor infiltrating into the axillary tail of Spence. In Okubo et al.'s study [10], a core needle biopsy of the axillary mass showed invasive ductal carcinoma, and a subsequent vacuum-assisted biopsy from the UOQ breast showed intra-ductal carcinoma, leading to the final diagnosis of CATS with extended intra-ductal spread. In our case, since the primary location of the tumor was epicentered in the axilla with no imaging abnormalities in the UOQ on imaging, our diagnosis of CATS was confirmed. It is crucial to eliminate the possibility of axillary lymph node metastasis, which can result from various primary tumors such as the breast, lung, gastrointestinal, haematological, thyroid, ovarian, and accessory breast cancer. To prevent misdiagnosis, a comprehensive and systematic physical and imaging examination, including a whole-body PET scan, should be done to determine the primary origin. In our case, since we already had a histologic diagnosis of metastatic carcinoma, we performed a PET scan to rule out other sources. The scan showed only an FDG-avid lesion in the axilla with axillary lymphadenopathy, thus ruling out metastatic axillary lymphadenopathy from other sources. It should be noted that another crucial differential diagnosis is metastatic axillary lymphadenopathy from an occult breast primary. The clavipectoral fascia can be used to differentiate between the two, with any histologically confirmed axillary malignancy superficial to the clavipectoral fascia being CATS, while any mass beneath it being defined as axillary lymphadenopathy [13]. Although we couldn't confirm the presence of clavipectoral fascia on MRI in our case, the mass's superficial location suggested CATS instead of metastatic axillary lymphadenopathy.

Given the rarity of CATS, there is no clear consensus on the optimal treatment. The management of CATS comprises of a multi-disciplinary approach. The best approach is to follow the recommendations for histologically similar breast cancers of the same disease stage [4]. Patients with locally advanced breast cancer, positive axillary lymph nodes, and high-risk triple-negative or Her2Neu overexpressing tumors are offered

neoadjuvant therapy [14]. Surgical management involves the wide local excision of the axillary mass with axillary lymph node dissection or modified radical mastectomy, based on the surgeon's discretion. Adjuvant treatment is similar to typical breast carcinoma patients and includes chemotherapy, anti-Her2 therapy, radiotherapy, and endocrine therapy. In our case, the patient had already received 3 cycles of anthracycline-based chemotherapy with a poor response. Therefore, the patient was planned for taxane-based chemotherapy, anti-Her2 therapy and radiotherapy.

The prognosis for CATS depends on the stage when diagnosed. Compared to UOQ breast cancer, CATS has a lower 10-year breast cancer-specific survival (BCSS) rate, particularly in the metastatic lymph node and hormone receptor-negative subgroups [9]. The estimated five-year disease-free survival rate for CATS is 67 %. The rates of local failure, regional recurrence, and distant metastasis are 0 %, 10 %, and 30 % respectively [4].

4. Conclusion

CATS, though rare should be considered in differential diagnosis for a patient presenting with an axillary mass. MRI is a valuable tool to distinguish CATS from other lesions by demonstrating its continuity with the breast tissue and its contrast enhancement pattern. Immunohistochemistry is essential to confirm the diagnosis by showing the expression of specific markers, such as estrogen receptor, progesterone receptor, and HER2, in the tumor cells. The optimal treatment is surgery which requires a wide excision with microscopically negative margins and axillary lymph node dissection. Adjuvant therapy options include chemotherapy, anti-Her2 therapy, radiotherapy, and endocrine therapy. However, further studies are necessary to elucidate the clinical and pathological characteristics, prognosis, and optimal treatment modalities of CATS.

Ethical approval

Exempted from Institutional Ethics Committee of Saroj Gupta Cancer Centre and Research Institute, Kolkata, India.

As this is a case report, with no experiment done on the patient and the treatment followed standard treatment guidelines, it has been exempted from ethical approval by Institutional Ethics Committee.

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Author contribution

Dr. Arnab Gupta- study concept
 Dr. Pranjit Kalita- study design, data collection, analysis and Interpretation
 Dr. Samir Bhattacharyya- conceptualisation
 Dr. Piyas Sengupta- analysis and interpretation
 Dr. Soumendra Mishra- contributor
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Guarantor

Dr. Arnab Gupta, Dr. Pranjit Kalita

Research registration number

None.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest statement

None.

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