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Case Report

Axillary Supernumerary Breast, an Atypical Location for Invasive Lobular Carcinoma, A Case Report

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Abstract

The frequency of breast tissue outside the mammary gland, or supernumerary breast, is rare, with a reported incidence in women of 0.3 to 6% [1]. This tissue is typically located in the armpit [1,2], ectopic tissue can be the site of benign or malignant physiological variations similar to normally located breast tissue. The axilla is the most common site, while the sternal, infra-clavicular, epigastric, and vulvar regions have also been described.

We report a case of infiltrating lobular carcinoma of the axillary supernumerary breast managed at the Tidjini HADDAM Cancer Center in Sidi bel abbes in ALGERIA.

Keywords: Supernumerary Breast; Axillary Breast; Infiltrating Lobular Carcinoma; Breast Cancer

Introduction

Extra-mammary or supernumerary breast tissue is rare, with a reported incidence of 0.3 to 6% [1]. Breast embryonic development begins during the 4th week of gestation and is located on the ventral surface of the body. The «milk line» forms a ridge from the bilateral mammary ectodermal tissue and extends from the base of the axillary region to the inguinal region.

From the 5th week of gestation, the «milk line» gradually disappears but continues to proliferate and differentiate into mammary lobules specifically at the level of the 4th intercostal space [2]. Failure and/or abnormality of this involution process can lead to ectopic breast tissue presenting as polymastia (accessory breast glands) or polythelia (accessory nipples) [3].

Ectopic breast tissue has the same anatomical and physiological characteristics as breast tissue in the «normal» position and is subject to the same hormonal repercussions, vulnerable to the same disorders, including malignant transformation [4].

Indeed, ectopic breast cancer accounts for 0.2 to 0.6% of all breast cancers [5]. The axilla is the most common location (70-90%), with the glandular tissue located in the subcutaneous tissue and deep dermis [6,7]. Its diagnosis is mostly delayed, estimated at 40.5 months [8] due to misdiagnoses, especially with other subcutaneous masses: lymphadenopathy, sebaceous cysts, nevi, lipomas, or hidradenitis [4].

Case Report

We report the case of a 61-year-old female patient who presented to our clinic with an unlabeled axillary retraction for 12 years, but who recently presented with unusual paroxysmal axillary pain and a change in coloration with the appearance of pseudo-pustular erythema without any breast signs. Clinically, the patient had a red, 5 cm major axis, axillary cutaneous mass, adherent to the cutaneous and deep planes, polylobulated, with palpable, round, 3 cm

diameter adenopathies fixed to the deep plane. The homolateral breast is normal in appearance, with congenital nipple umbilication which is also bilateral (Figure 1).

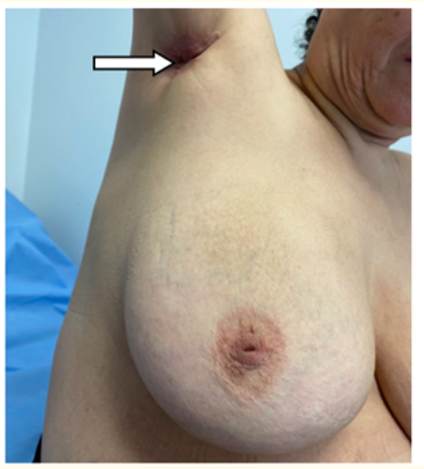

Figure 1: Clinical aspect of the axillary mass corresponding to an infiltrating lobular carcinoma of the supernumerary breast.

An initial axillary ultrasound was performed and revealed a heterogeneous anechoic axillary cutaneous mass with deep extension measuring 38x17mm, in close contact with the axillary vascular axes; associated with a 34x24mm axillary lymph node mass.

A breast MRI confirmed the presence of a neoplastic process on a supernumerary breast and ruled out any breast lesions in both breasts.

A microbiopsy revealed breast parenchyma with diffuse carcinomatous proliferation arranged in «Indian files», composed of medium to large round cells with marked cytological atypia and high mitotic index. This appearance corresponds to a grade II invasive lobular carcinoma according to SBR, on supernumerary breast. Immunohistochemistry confirmed an invasive lobular carcinoma of the Luminal B type with 80% positive hormone receptors, HER2 negative and Ki67 at 10%.

After an extension assessment, namely a thoraco-abdomino-pelvic scan and bone scintigraphy, the tumor was classified as T4dN2M0, and neoadjuvant chemotherapy followed by surgery and external radiotherapy was decided after a multidisciplinary consultation meeting.

The patient was placed on neoadjuvant chemotherapy, protocol 04 AC 60 – 12 Paclitaxel, i.e. 04 cycles combining adriamycin and cyclophosphamide followed by 12 cycles of paclitaxel at 80mg/m^2. A minor clinical response was observed after the 04 AC60 cycles, but the redness disappeared and the mass became mobile relative to the deep plane after the 12 paclitaxel cycles (Figure 2).

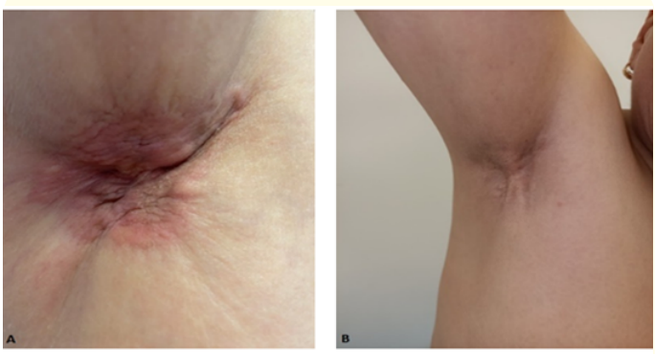

Figure 2: Progression of tumor mass during chemotherapy. A: Assessment after 04 AC60 – B: Assessment after 12 Paclitaxel.

The patient underwent surgery, which consisted of a lenticular resection of the entire tumor mass and a lymph node dissection, without any procedure on the homolateral breast.

The histological and immunohistochemical study on the surgical specimen found at gross description a tissue formation measuring 10/07/03 cm covered by a skin. slice of section: presence of a well-defined whitish nodule of 1.5 cm.

Microscopic description found a Tumor cells arranged in sheets, single files and large nests.Tumor cells are discohesive, small, monomorphic with scant cytoplasm and low grade nuclei. The tumor stroma is abundantly fibrous and inflammatory. Peri-neural invasion is present. lymph node dissection found 10 lymph nodes infiltrated by the same neoplastic process out of the 19. The histological response was classified as RCB1 (Residual Cancer Burden1) and the tumor was classified as pT1c N3a (Figure 3).

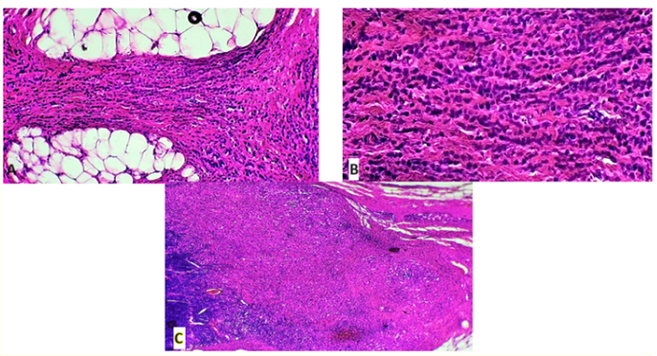

Figure 3: A- Tumor cells arranged in single files and single cells (HE X10). B- Tumor cells are discohesive, small, monomorphic with scanty cytoplasm and lacking marked atypia (HEX20). C- Lymph node matastasis (HEX 10).

Immunohistochemistry: Estrogen and progesterone receptors was positive, respectively 70% and 80%, KI-67 (60%) and HER2 score 2 with FISH test negative.

The disease progression was marked on the 17th postoperative day by the onset of homolateral mastitis of the lower quadrants with significant inflammatory signs, paroxysmal pain, and a fever of 39°C without signs of parietal sepsis. We requested a breast MRI, which confirmed the homolateral mastitis with extensive dermo-hypodermic cellulitis of the entire breast and post-operative fibro-scarring without signs of malignancy. The patient responded to a 10-day antibiotic treatment with Pristinamycin and Metronidazole with complete clinical and radiological response.

The patient received external beam radiation therapy to the axillary region at a dose of 45Gy.

Discussion

Polymastia is a congenital condition characterized by the persistence of breast tissue in an abnormal location. It is located along the primitive mammary line, which extends from the armpit to the inguinal fold. It is estimated that 0.3% to 6% of women and 1% to 3% of men have a supernumerary breast. There are two types of ectopic breast tissue: supernumerary breast tissue and aberrant breast tissue [9,10]. Supernumerary breasts include a nipple, areola, and mammary gland, either isolated or associated [11].

Teir reported that the frequency varied between different ethnic groups, ranging from 0.6% to 6% [11,12]. Regarding aberrant breast tissue, they have neither nipple nor areola, and they generally do not involve a secretory system [11]. They are usually located near the breasts [13]. The armpit is the most common location, while the sternum, infra-clavicular region, epigastrium, and vulva have also been described [9,11,14]. Evans., *et al*. found ectopic breast tissue in the armpit in 71% of cases [4] and in 58% of cases for Marshall., *et al*. followed by the sternal region in 18.5%, sub clavicular in 8.6%, sub mammary in 8.6%, and vulvar in 4% [11]. Overall, they are associated with a frequency that varies between 1.7% and 6%. In 13% of cases, an axillary supernumerary breast is present on both sides [15].

Like normal breast tissue, ectopic breast tissue is hormone-dependent. The anomaly is present at birth but is usually only discovered during pregnancy or lactation, when it may increase in size, become cyclically painful, or cause discomfort. Suspected abnormal ectopic breast tissue follows the same diagnostic principles as normal tissue. This includes anamnesis, physical examination, and conventional paraclinical tests [16]. It is important to note that the presence of a supernumerary breast may go unnoticed on standard mammograms due to its location. The tumor may be located in a place that does not appear on standard radiographic images. Ultrasound should be used to complement mammography. MRI may be performed to exclude ipsilateral or contralateral breast cancer. Since ectopic breast tissue can be bilateral, contralateral evaluation should be strongly considered [7]. In our case, an MRI was recommended and performed due to the lobular histological type of the carcinoma [17,18]. The reported incidence of the lobular histological subtype among ectopic breast carcinomas is 3 to 9.5% [1,19]. A review of the Japanese literature showed that medullary, mucinous, and apocrine carcinomas were more common. Additionally, Marshall., *et al*. noted that the distribution of histological types was as follows: 79% invasive ductal carcinomas, 9.5% invasive lobular carcinomas, and 9.5% medullary carcinomas [11].

Histologically, the absence of lymph node tissue and the presence of normal breast ducts and lobules adjacent to the tumor are necessary to confirm the diagnosis of ectopic breast carcinoma and to exclude the possibility of a metastatic tumor from an occult primary lesion [20]. Ectopic breast cancer is classified using the same TNM staging system as for primary breast cancer. Due to its subcutaneous location, skin involvement may be early and lead to T4 stage cancer [19]. In half of the cases, there is extension of the cancer to the axillary lymph nodes at the time of diagnosis. This early nodal extension is explained by the proximity of the malignancy to the nodes, but also by the delay in diagnosis, which can be 3 to 24 months after the onset of symptoms, especially in the absence of a nipple and areola.

The diagnosis and management of ectopic breast cancer follow the same therapeutic approach as breast cancer. Due to its rarity, there is no acceptable level of evidence to support a different approach. A high index of suspicion is necessary to diagnose carcinoma of an ectopic breast. Once the diagnosis is established, it is important to assess the extent of tumor spread, which includes a bone scan, chest-abdominal CT scan, and breast MRI to ensure the absence of distant metastases and a second breast cancer.

Due to its incidence and the scarcity of data, surgical management has been controversial in recent years. Historically, ectopic breast cancer has been treated with radical mastectomy and chest irradiation [19]. Later, Cogswell and Czerny [15] demonstrated that ipsilateral mastectomy in these patients does not lead to a better prognosis. In a series of autopsied patients with ectopic breast cancer who died of metastatic lesions in the skeletal system, pleura, and liver, there was no infiltration of the ipsilateral breast. No breast involvement was observed in radical mastectomy specimens in patients with distant metastases. Evans and Guyton [7] concluded that there was no additional benefit of mastectomy compared to wide excision with axillary lymph node dissection in the presence of sentinel node positivity. These aggressive techniques are no longer an option, and local excision is the recommended procedure. Mastectomy should only be considered in patients with a second primary breast lesion.

Regarding adjuvant treatment, it has the same indications as breast carcinoma in its anatomical position [20]. This treatment

can be supplemented with hormone therapy when the tumor is positive for hormone receptors (positive estrogen receptors).

It is difficult to accurately assess the prognosis of breast cancer on ectopic breast tissue because the diagnosis is often delayed and because of the lack of long-term follow-up data for this rare tumor.

A Japanese retrospective study of 98 patients reported in the journal Surgical Oncology that, of 64 evaluable patients, all but one were alive after a median follow-up of 28 months [21]. A 2015 Chinese retrospective study published in Oncology Letters included 11 cases of breast cancer on ectopic breast tissue [22]. The patients, aged 27 to 48 years, were followed for a median of 20 months. Axillary lymph node dissection was performed in six of them. Four had distant metastases. The three-year overall survival rate was 54.5% (6 of 11 patients) due to the occurrence of late metastases. In 2011, Strasbourg surgeons reported in the journal Gynecology Obstetric and Fertility a case of cancer on ectopic breast tissue that occurred several years after its surgical removal. No cases of cancer after excision of an accessory breast gland had been previously published. The tumor had developed on the scar of the ectopic breast gland excision.

Breast cancer on ectopic breast tissue can also occur in male patients. In a literature review published in 2011 in the Journal of Plastic, Reconstructive and Aesthetic Surgery, Dutch and Italian surgeons reported six cases in men among 171 cases of axillary breast carcinomas (with an average delay of 40 months before diagnosis) [8] and its incidence on ectopic breast tissue is higher than in the usual form of male breast cancer [24].

Conclusion

Ectopic or supernumerary breast carcinoma is a rare and difficult diagnosis due to the lack of information and awareness among clinicians.

Furthermore, supernumerary axillary breast tissue is not detected by most screening and diagnostic tests. Its management is based on a multidisciplinary approach following the same recommendations as for breast cancer. It can also occur in men and does not seem to have a worse prognosis when diagnosed at similar stages of the disease.

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