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A Rare bilateral breast tumor: Primary breast sarcoma and intracystic papillary carcinoma

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Abstract:

Bilateral synchronous breast cancer (BSBC) is not a rare entity. This bilaterality can be diagnosed at the same time or during an interval of six months. The bilaterality as well as the multicentricity of a breast cancer in fact reflects the capacity of the neoplastic transformation to appear in different places of the breast tissue independently, simultaneous or not. Bilateral synchronous tumors are considered independent when they have different histological and immunohistochemical characteristics.

We report the observation of a 53-year-old patient with concomitant bilateral invasive breast cancer: this is an intracystic papillary carcinoma of the right side and a primary high-grade sarcoma on the left side.

Introduction:

Women treated for unilateral breast cancer are at high risk of developing new cancer in the contralateral breast. This risk is estimated at five to seven times the risk of developing primary primary breast cancer in the general population [1]. Synchronous bilateral breast cancer was a fairly common pathology. Its incidence varies according to the authors from 1.5 to 3.5% and according to the diagnostic interval chosen by the authors [1].

Intra-cystic papillary carcinoma (ICPC) of the breast is a variant of intracanal carcinoma, it is a papillary lesion located in a dilated or cystic galactophore channel. ICPC is rare and represents 0.5 to 1% of carcinomas breast [2]. It is characterized by slow growth with a good prognosis. However, the therapeutic attitude is not well codified, hence the risk of abusively treating such a lesion.

Breast sarcomas are even rarer. All histological forms are possible with a predominance of tumors of the histiofibrocytoma type. The grade, reaching the margins and sometimes tumor necrosis are major elements of the prognosis. Among the different subtypes, angiosarcomas are distinguished by a more frequent occurrence on irradiated ground and by a very dark evolution with a high metastatic pulmonary potential. [3]

The treatment is most often based on a mastectomy without lymph node dissection given the exceptional axillary invasion. In certain situations, a conservative treatment can be discussed according to the tumor size, the grade and the breast volume. Locoregional radiotherapy will readily be offered to tumors larger than 5 cm or high grade. Systemic chemotherapy is not a standard but should be discussed in the forms at high risk of relapse. [3]

On the occasion of our observation, we describe the diagnostic and therapeutic aspects of these two synchronous rare tumor entities .

Observation:

This is a 53-year-old patient, menopausal for 7 years without hormone replacement therapy, who consults for left breast inflammation noted for a week. This patient does not have a history of neoplasias in the family.

On clinical examination, there is a voluminous well-limited breast mass left retroareolar movable relative to the deep planes of 16 cm in diameter associated with breast inflammation and ulceration of the skin at the level of the left upper quadrant. **(fig. 1).**

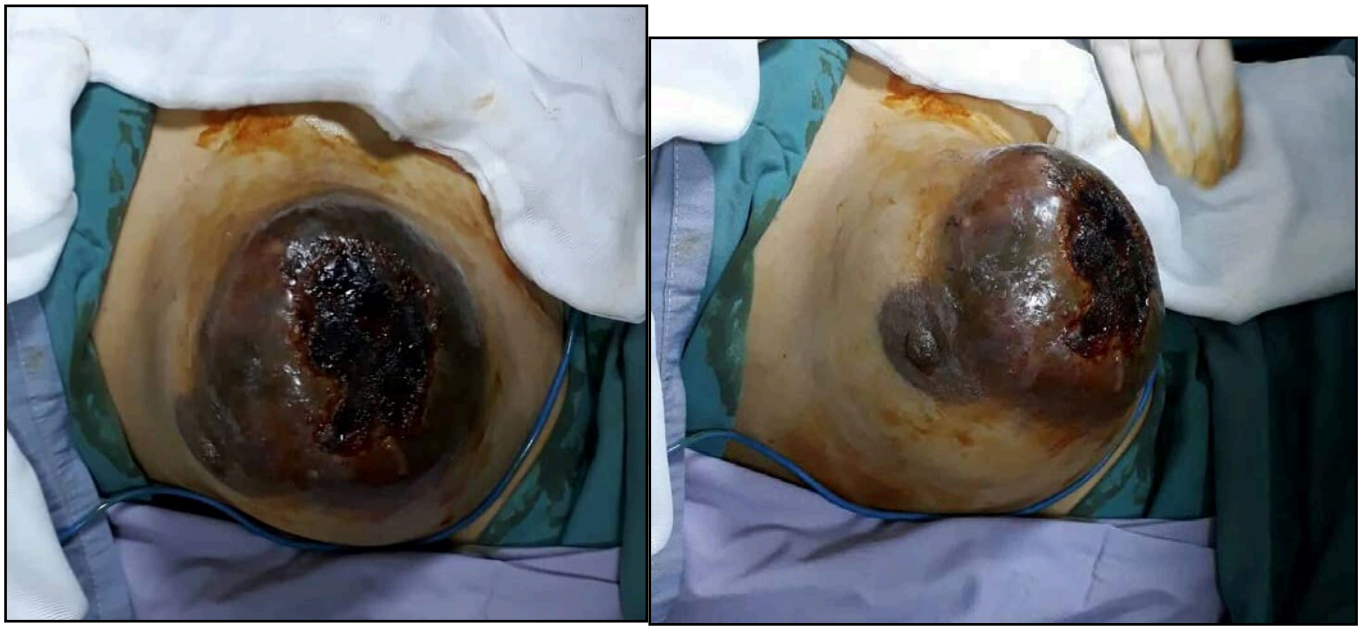

Figure 1: Left breast: necrotic ulcerative aspect of neglected breast cancer

The right breast is occupied by a nodule of 10 cm ill-defined mobile and without associated skin or nipple abnormality.

A bilateral echo-mammography is performed. It highlights two bulky necrotic breast tumor masses, suspect, responsible on the left for a loss of the cutaneous substance with suspicious axillary lymphadenopathy (fig. 2).

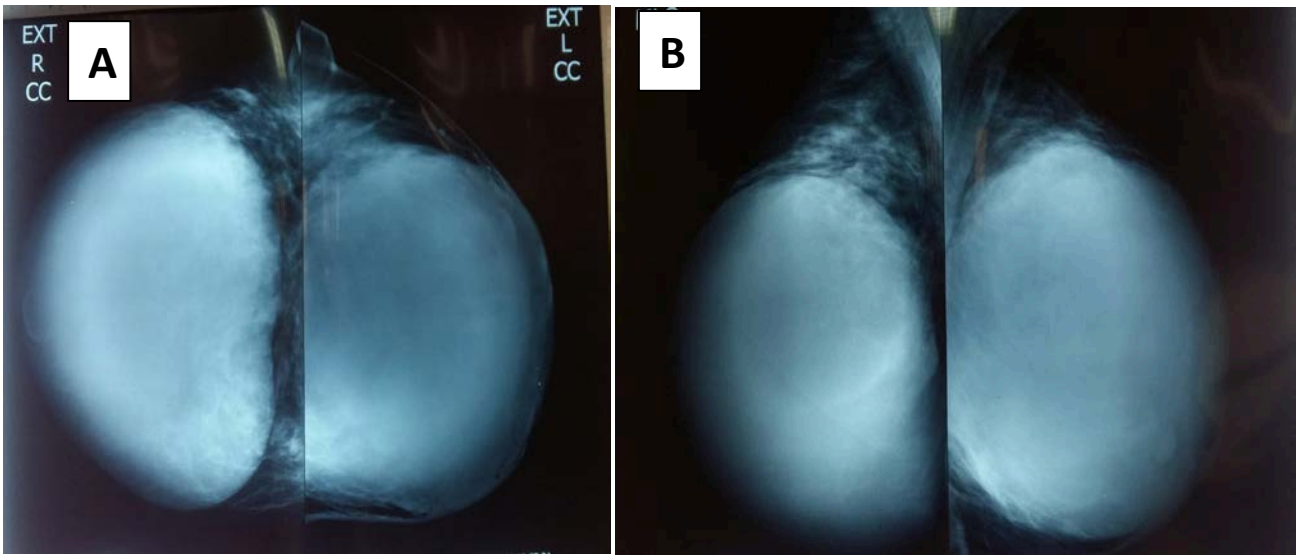

Figure 2: Mammographic appearance of two tumor masses: A / craniocaudal cut B / external oblique cut

The color Doppler study shows on the right breast an increased vascularization of the cystic wall and of this heterogeneous portion which extends into the adjacent glandular parenchyma thus signifying its tissue nature and its extracystic extension .

In the left breast, it is therefore a complex left retroareolar cyst with a fleshy vascular portion.

The balance sheet is classified according to the BIRADS classification of the ACR, ACR4b on the right and ACR5 on the left.

A solid part aspiration puncture was performed on the right breast but it is not very cellular and the material is hemorrhagic and inflammatory. However , the percutaneous echo-guided biopsy of the contralateral breast, performed at the same time, had shown a histological aspect of a malignant tumor with spindle cells which can evoke a poorly differentiated sarcomatous tumor.

The decision of the multidisciplinary committee was to perform a bilateral mastectomy.

Histological analysis of the operating room, on the left, shows a poorly differentiated high-grade sarcoma, largely necrotic of the left breast, measuring 17 cm from the major axis, ulcerated in the skin over 8 cm from the major axis. It infiltrates adjacent breast tissue and spares the nipple.

Among others, on the right, it is an intracystic papillary carcinoma with invasion of the right breast tissue, measuring 14 cm from long axis, located in retro-nipple, of grade SBR II. Presence of vascular emboli at the level of the cystic wall. the tumor is associated at the level of the axillary extension with simple intraductal papillomas and with intra-lobular neoplasia.

The extension assessment, including a thoraco-abdomino-pelvic scanner and a scintigraphy, was negative.

After discussion of the case with multidisciplinary staff, axillary dissection is decided, followed by chemoradiotherapy with an immunohistochemical study of hormone receptors.

Discussion

Bilateral breast cancer

Breast cancer is considered bilateral when both breasts have a malignant lesion simultaneously or at different times. The bilaterality as well as the multicentricity of breast cancer actually r reflects the ability of the malignant

transformation to appear in different places of the breast tissue independently, simultaneously or not [1]. This ability is certainly linked to a number of risk factors. Cancer discovered in the contralateral breast must be distinguished from a metastatic location at this level. Several criteria, in fact, suggest that it is a real second primary breast cancer. These criteria are: [1]

• the two cancers are of different histological types; as is the case in our observation; otherwise, other criteria must be sought;

• the presence of an in situ component in contralateral breast cancer;

• the degree of differentiation of the 2nd cancer is higher compared to the 1st cancer;

• the absence of evidence of any metastasis during the treatment of the first cancer with a time interval between the two cancers of several years

However, there is ambiguity as to the definition of simultaneous bilateral breast cancer, which makes the study of bilateral breast cancer sometimes difficult. For some, contralateral breast cancer is simultaneous if it is discovered during the treatment of the first cancer or in the month which follows; others, when discovered within the first six months or 12 months after treatment for the first cancer [4] .

The data concerning the prognosis and the impact of the occurrence of the second cancer in the contralateral breast are contradictory. For some authors, the survival rate after bilateral breast cancer is, at the same stage, the same as that of patients treated for unilateral breast cancer, while for others it is worse. [5].

In fact, the prognosis of patients treated for bilateral breast cancer depends on the prognostic elements of both the first and second cancer. [5].

The therapeutic management of synchronous bilateral breast cancer was controversial and variable depending on the teams and the periods. The local surgical procedure offered to patients was often radical (mastectomy), to the detriment of conservative treatment [5].

Intracystic papillary carcinoma

Intracystic papillary carcinoma is a special feature of breast cancer. It is a rare malignant ductal tumor, representing 0.5 of all breast carcinomas [2]. It usually occurs after 40 years. In about 50% of the cases it is of central seat and more precisely in the retroareolar region . The tumor size varies from 1 to 14 cm, as is the case in our observation. It is generally characterized by slow growth with a good prognosis compared to other ductal carcinomas. The overall five-year survival is 83 to 96% according to the authors [2].

Clinically the tumor is revealed by a breast swelling; conventionally old, having recently increased in volume, which can lead to skin ulcers or painful tension [6]. In our observation, the anteriority of this mass was unknown and was revealed following an inflammation of the contralateral breast. This tumor can also manifest itself in a bloody nipple discharge that is reported in 22 to 34% of cases [6].

On mammography ICPC generally appears as a round, oval or lobular opacity. The contours are generally sharp, generally well circumscribed, but sometimes they can be hidden or indistinct in places [7]. On the mammogram of our patient, the tumor mass is observed as an opacity with limit and clear outline. Spiculated contours are rare [7].

The ultrasound-guided puncture with the fine needle of the cyst brings back a bloody or old blood color liquid and the cyst reproduces quickly after the puncture [7]. Liquid cytology should be interpreted with caution due to the frequency of false negatives, as in our observation [7]. The biopsy of the lesion involving the solid portion is generally more informative.

Macroscopic examination finds, in a cyst with a thick and fibrous wall, a rounded, or multi-lobed, brittle and haemorrhagic formation limited by a fibrous and thick capsule [8].

Histologically, the tumor architecture is papillary, the lesion is usually localized in a cystic duct, it is characterized by a small fibro-vascular tree devoid of a layer of myoepithelial cells, and a neoplastic epithelial proliferation exhibiting the morphological characteristics of a Low nuclear grade ductal carcinoma [8].

The therapeutic strategy remains variable given the rarity of this type of breast carcinoma. In general, the prognosis of ICPC in its isolated form appears excellent regardless of the type of intervention. Breast conserving surgery remains the most used. The absence of axillary lymph node metastases in the study by Baron et al. and that of Harris et al. combined with the absence of recurrence, suggest that the treatment of choice for an isolated ICC is an enlarged lumpectomy. [2] However, in some cases, a mastectomy may be offered (large tumors, insufficient margins, recurrence and patient preferences) which is the case for our patient who initially benefited from a mastectomy. [2]

Lymph node metastases remain exceptional. The sentinel node biopsy may be an excellent alternative for lymph node assessment in the case of associated invasive carcinoma [8].

The role of adjuvant therapy remains controversial. However, numerous articles and published data recommend radiotherapy in young women under 50, and in the forms associated with ductal carcinoma in situ [8]. Chemotherapy is not compulsory. Hormone therapy is mainly prescribed to reduce the risk of local recurrence in the event of positive hormone receptors.

Primtive breast sarcomas

Breast sarcomas are a rare heterogeneous set of neoplasms, accounting for about 1% of breast tumors and less than 5% of all sarcomas [3]. The average age of onset is 50-55 years. [3].

Among the different subtypes, we mainly find the included sarcomas, angiosarcomas, fibrosarcomas, liposarcomas, leiomyosarcomas and more rarely, stromal sarcomas, osteosarcomas, protuberant dermatofibrosarcomas, soft tissue sarcomas and rhabdomyosarcomas [3]. The assessment of the histological grade, which remains a major prognostic factor, is most often done using the classification of the National Federation of Cancer Control Centers (FNCLCC) , which takes into account the differentiation, the number of mitoses and necrosis [3]. Angiosarcomas are most often grade III [3]. For the other histologies, it is mainly grades II and III [3].

Sarcomas of the breast often present as large, palpable tumors. The median size varies, depending on the series, between 5 and 6 cm with extremes of up to 30 cm. [3] In our series, the size of the sarcoma was 17 cm. They are sometimes rapidly progressive and painful. The presence of clinical lymphadenopathy is exceptional. There does not seem to be a correlation between tumor size and histological type [9]. Cutaneous signs are possible, in particular in the case of angiosarcomas: edema, ulceration, appearance of pigmented or unpigmented papules, vesicles, eczematized appearance, change in coloring of scars of lumpectomy [3].

In standard imagery, there are few discriminating elements. Mammography may show the presence of spiculated microcalcifications, especially in the case of osteosarcomas. In ultrasound, we can find a well defined mass with a cystic component. [3].

The classic prognostic factors of soft tissue sarcomas have also been highlighted for breast sarcomas: the main ones are tumor size, histological grade and margins of excision [10].

Using a different grade assessment from that of the FNCLCC, McGowan et al. [10] also found specific survival rates significantly in favor of grade I / II tumors vs grade III / IV tumors. For this team, there is also a clear difference in survival without local relapse at five years depending on the invasion of surgical margins . [10].

As with phyllodes tumors, surgery is the standard treatment. All the authors agree on the need for broad surgery with negative margins to obtain the best local control. Mastectomy thus remains the “gold standard”.

Furthermore, in the absence of a carcinosarcoma-type epithelial component with a higher risk of invasion, axillary dissection is not indicated due to the low risk of involvement [3].

Few data are available on radiotherapy and they are mainly retrospective. In the majority of studies, radiotherapy has no impact on overall or recurrence-free survival, but it is a small number [3].

In a neoadjuvant situation, chemotherapy does not seem to bring any benefit in view of the low chemosensitivity and the fact that the majority of breast sarcomas are operable from the start [3].

We could therefore discuss chemotherapy for sarcomas of high grade breast, larger than 5 cm or incomplete excision.

Conclusion:

Intrakystic breast carcinomas and primary breast sarcomas represent two particular breast cancer entities whose association is also rare. The prognosis and management of these two simultaneous tumors depends on the prognostic elements of both the first and second cancer.

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