

Metastatic Adenoid Cystic Carcinoma of the breast at diagnosis

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Abstract:

Introduction: adenoid cystic carcinoma of the breast is a rare lesion among breast carcinomas, representing less than 0.1% of breast cancer cases. Generally indolent behavior with rare lymph node involvement and metastases.

Case Report:

76-year-old patient complaining of a lump in her right breast that had appeared 8 months ago. Ultrasonography showed a heterogeneous image of 3.9/2.4/3.7cm. On palpation, there was an 8cm nodule and mobile and fibroelastic axillary lymph nodes. A core needle biopsy was performed with an anatomopathological result of triple-negative grade 2 adenoid cystic carcinoma. Screening demonstrated two 1.1cm pulmonary nodules with soft tissue density suggesting metastasis. Abdominal tomography demonstrated liver nodules suggestive of secondary implants. The PET scan confirmed uptake in the lung and liver nodules, in addition to lytic lesions in S3. Liver biopsy confirmed implantation of adenoid cystic carcinoma. Chemotherapy was started with a regimen of Carboplatin, Paclitaxel and Pembrolizumab. After the third cycle of chemotherapy, the breast was reevaluated without showing any tumor reduction. The metastases also did not show regression, and treatment was interrupted. The patient evaded the service until January 2024, when he returned complaining of pain in the right hip and difficulty walking. Chest and hip tomography showed an increase in lung lesions and the appearance of lytic metastasis in the iliac bone. The patient refused to undergo radiotherapy and in May 2024 she developed liver failure and death.

Discussion:

In the metastatic setting, the regimens of choice are based on anthracyclines and taxanes with reports of little response. Our report aims to contribute to the introduction of a new group of drugs (pembrolizumab) associated with the standard taxane regimen, demonstrating that this type of tumor really has a difficult response to chemotherapy agents and that there is little that can be done in the setting of multiple metastases.

Key words: adenoid; breast; metastasis; death

Introduction

Adenoid cystic carcinoma of the breast is a rare lesion among breast carcinomas, representing less than 0.1% of breast cancer cases [1]. Generally indolent behavior with rare lymph node involvement and metastases [2].

It predominantly affects the subareolar region of the breast in 50% of cases (3). Most are triple-negative tumors with basal-like characteristics, but have low Ki67 values, generally less than 20[2,3]. Unlike triple-negative tumors, triple-negative cystic adenoids have an excellent prognosis when confined to the breast. They present an epithelial and myoepithelial component similar to salivary gland tumors. They have architectural differentiations that are tubular-trabecular, cribriform and

solid [4]. Its main symptom at presentation is a palpable lump, rarely associated with breast pain [5,6]. Treatment mostly depends on the size of the lesion. Conservative surgery is indicated in cases of tumors that can be excised with free margins and mastectomy in cases of larger tumors with disproportion in the tumor/breast ratio. The axillary approach is generally not necessary and is indicated in cases of grade 2 or grade 3 tumors, in which there is a 2% chance of axillary involvement [7,8], with dissection of the sentinel lymph node being appropriate. Post-operative radiotherapy is a complementary treatment required in cases of conservative surgery to reduce the chances of local recurrence, which are common throughout the first 10 years of follow-up [9,10]. Adjuvant

chemotherapy is mostly unnecessary because it is an indolent tumor, and no gain in overall or disease-free survival has been demonstrated with the addition of chemotherapy [6]. There are reports in the literature from experts that indicate adjuvant chemotherapy in cases of tumors with axillary involvement, grade 3 tumors and tumors larger than 3cm [6,11].

Due to its rarity and peculiar characteristics, there is much to be learned about biological behavior, carcinogenesis and the best way to treat it. In the absence of clinical trials involving this histological type, case reports become important in acquiring knowledge to optimize treatments.

Case report:

Female patient, black, 76 years old, G3P3, complaining of a lump in the right breast that had appeared 8 months ago. He has a history of admission to the intensive care unit 10 months ago due to an ischemic stroke, presenting left hemiparesis. Presents hypertension, hypothyroidism and dyslipidemia. Family history of father with prostate cancer. He presents sclerocalcification in the mitral and aortic valve on the echocardiogram. He presented clinical signs of an initial dementia process. Ultrasonography showed a heterogeneous image of 3.9/2.4/3.7cm in the union of the upper quadrants of the right breast, axillary lymph nodes with proportionality between cortex and hilum bilaterally BI-RADS 4 B BI-RADS 4 B.

On palpation, there was an 8cm nodule in the right breast and mobile and fibroelastic axillary lymph nodes. There were no palpable nodules in the cervical or supraclavicular region bilaterally. A core needle biopsy was performed with an anatomopathological result of triple-negative grade 2 adenoid cystic carcinoma. Staging demonstrated two 1.1cm pulmonary nodules with soft tissue density suggesting metastasis. Abdominal tomography demonstrated liver nodules suggestive of secondary implants. The PET scan confirmed uptake in the lung and liver nodules, in addition to lytic lesions in S3. Liver biopsy confirmed implantation of adenoid cystic carcinoma. The patient was staged as T2N0M1. Chemotherapy was started with a regimen of Carboplatin, Paclitaxel and Pembrolizumab. After the third cycle of chemotherapy, the breast was reevaluated without showing any tumor reduction. The metastases also did not regress, and treatment was interrupted. The patient evaded the service until he returned in January 2024 complaining of pain in his right hip and difficulty walking. Chest and hip tomography showed an increase in lung lesions and the appearance of lytic metastasis in the iliac bone (figure 1, 2, 3).

At this point the breast was reevaluated with ultrasound showing a small tumor evolution from 3.9cm in largest diameter to 4.3cm. The patient refused to undergo radiotherapy and in May 2024 she developed liver failure and death.

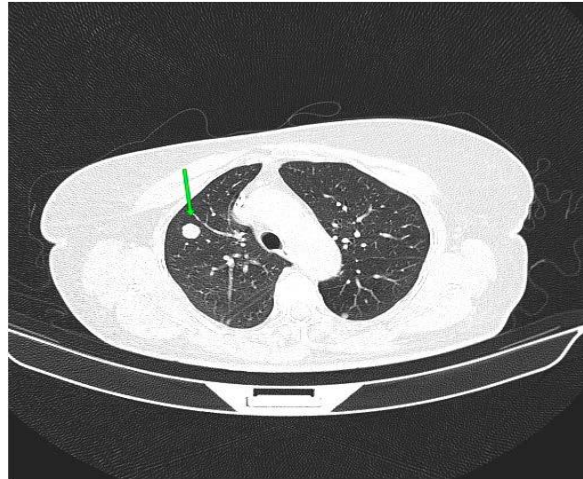


Figure 1:



Figure 2:

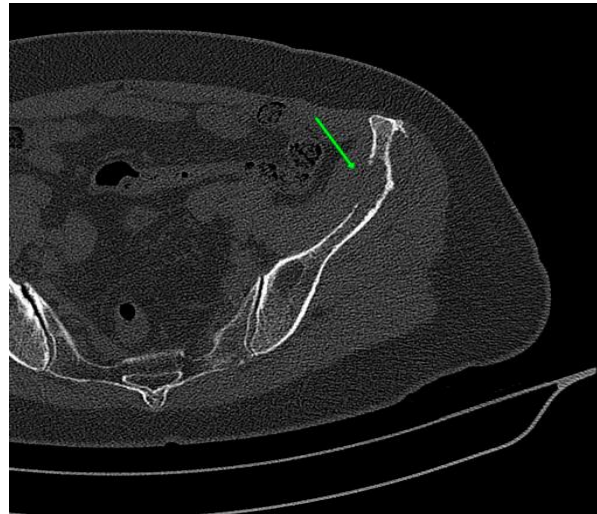


Figure 3:

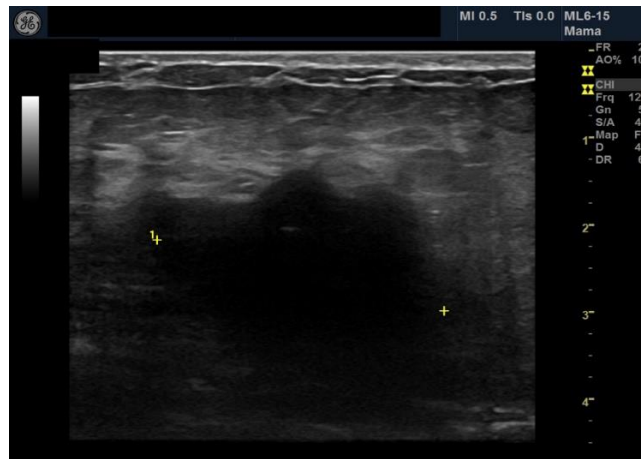


Figure 4:

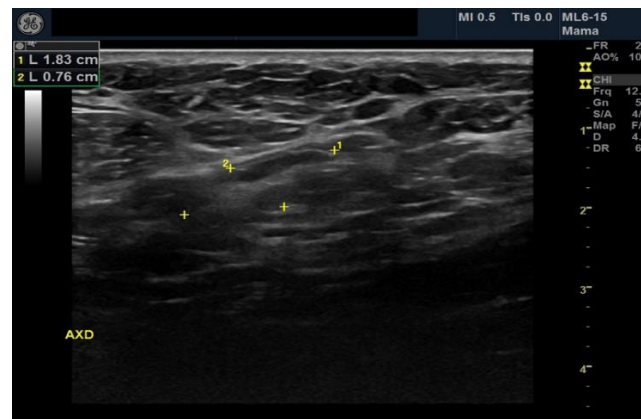


Figure 5:

Discussion:

This case describes a rare metastatic involvement of an adenoid cystic tumor of the breast that did not present axillary involvement. In the literature, the possibility of metastatic involvement without the necessary axillary involvement in this type of tumor is described [12,13]. The main site of metastasis involvement is the lung and few cases are described in the literature with involvement of multiple organs as in our case [14].

Due to the rarity of metastatic involvement of adenoid cystic carcinoma, treatments are based on experiences with cystic adenoids of the salivary gland. For metastatic disease, vinorelbine, doxorubicin, and 5-fluorouracil have been used. Mitoxantrone was used in a breast metastatic study obtaining stability of lung metastases [8]. There is no established recommendation in cases of advanced disease. The use of chemotherapy based on doxorubicin and taxanes, the basic regimen for breast cancer,

was applied in cases of metastatic disease with variable results of stable disease, progressive disease and partial response [14]. Currently, in salivary gland tumors, the expression of Trop-2 (Trophoblast Cell Surface Antigen 2) has raised the possibility of using Trop-2 inhibitors also for breast disease. Thus, sacituzumab govitecan has been described as a possibility in metastatic triple-negative disease as an antibody drug conjugate with action against TRop-2 and an antitopoisomerase 1 drug conjugate [15].

Our patient was categorized as grade 2 because she had a solid component in less than 30% of the tumor extension. Despite being a grade 2 tumor, it is a case of severe presentation with multiple foci of metastases, corroborating the idea of Arpino et al to categorize these tumors into low grade and high grade. Due to this idea of classification, our tumor would fit into a high-grade tumor in which a worse prognosis is described, when it presents basal cell characteristics. The literature describes that the larger the solid component, the worse the patient's prognosis [12]. Local treatment was not planned as the patient had multiple foci of metastases. In these cases, metastasectomy would not be indicated, with chemotherapy being the standard treatment [14]. Our patient died within 1 year after diagnosis, in line with reports of survival in 5, 10 and 15 years of 98.1%, 94.1% and 91.4%, respectively [3,16,17]. The literature describes stability of tumor size for 1 to 2 years, corroborating our case in which the tumor was practically stable during 1 year of follow-up [13]. There are cases in the literature of slow tumor growth over a period of 9 years in a patient who denied treatment [13]. A. heterogeneous image presentation on ultrasound is compatible with the aspect described in the literature [13].

the mortality rate described is 3.3% [2], being a tumor with an excellent prognosis, showing rare cases of evolution with metastases, with treatment being based on local excision with margins, radiotherapy, sentinel dissection in tumor chaos high-grade, without the need for chemotherapy [18]. Our case goes completely against the evolution of adenoid cystic tumor, showing a case of presentation with multiple metastases and death within 1 year, despite the use of a standard regimen for advanced triple-negative tumors. This lack of response demonstrates that this tumor responds poorly to chemotherapy regardless of the regimen used and despite being a triple-negative tumor, it behaves differently from triple-negative tumors in terms of the performance of pembrolizumab and platinum.

Conclusion:

We conclude that despite cystic adenoids being an indolent tumor, in cases of metastatic presentation there is difficulty in the chemotherapy approach, with low response even with the addition of new chemotherapy regimens.

Disclaimers:

no conflict of interest in this case description

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