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Adenoid Cystic Carcinoma Of The Breast: Local Treatment And Sentinel Lodge: About A Case

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Summary:
Adenoid cystic carcinoma [ACK] is a rare tumor representing less than 1% of breast cancers and mainly affecting postmenopausal women. Its clinical, radiological and histological presentations are confused with other benign and malignant breast cancers. It has a good prognosis despite its triple negative profile and can be controlled by local surgical treatment and radiotherapy with a minimal risk of local recurrence and axillary or distant lymph node metastases. We report a case of localized adenoid cystic carcinoma of the breast without lymph node or distant metastases, treated by a wide tumorectomy with sentinel lymph node technique, the management of which was supplemented by local irradiation.

Keywords*: adenoid cystic carcinoma, breast, sentinel lymph node, indocyanine green*

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I. Introduction

Adenoid cystic carcinoma is a rare pathological entity and usually develops in the salivary glands and the upper airways but can be seen in other anatomical sites such as the external auditory canal, the Bartholin glands, the uterine cervix and rarely the breast where it represents approximately 0.1% of cancers.[1,2,3] Breast ACK is an insidious growing tumor with low malignant potential unlike other locations and has a good prognosis. [4, 5] The local recurrence rate is low and it exceptionally gives lymph node and distant metastases despite its triple negative phenotype similar to infiltrating ductal carcinoma of basal phenotype. [1.6]There is no consensus for its management because of its rarity. From a new observation and review of the literature, we discuss the anatomo-clinical and therapeutic particularities as well as the prognosis of this type of breast tumor.

II. Patient And Clinical Observation

Patient information: This is a 78-year-old patient, postmenopausal for 20 years, hypertensive for 2 years under antihypertensive medication, who presented with a nodule in the right breast discovered on self-palpation and progressively evolving for 2 months without other associated signs.

Clinical results: The breast clinical examination found a nodule measuring approximately 2.5 cm in diameterin the upper-external quadrant of the right breast, hard in consistency, with regular contours, painless and mobile in relation to the superficial and deep skin plane, without inflammatory signs or nipple discharge. The axillary and supraclavicular lymph node areas were free.

Timeline of the current episode: July 2022: The patient consulted our gyneco-breast cancer surgery department and was referred to radiology department for breast ultrasound and a biopsy of the breast nodule was performed using a tru-cut needle. Subsequently, she underwent an extension assessment [abdomino-pelvic ultrasound and chest X-ray]. August 2022: surgery was performed and then referred to radiotherapy for additional management.

Diagnostic evaluation:

The ultrasound-mammography performed revealed a nodule of 22 x 18 mm, well-limited in the upper-external quadrant of the right breast containing micro and macro-calcifications. Furthermore, the other quadrants of the right breast as well as the left breast were normal.

A microbiopsy of the right breast nodule at the level of the upper-external quadrant was performed and revealed the following morphological and immunohistochemical characteristics:

Microscopic examination showed tumor proliferation made of cribriform masses and clusters with small lumens lined with epithelial cells and filled with mucus; which masses show pseudo-lumens with a stromal matrix and a few fibroblasts; tumor cells with a basaloid appearance and scant cytoplasm. The nuclei are ovoid, dense, showing mild to moderate atypia and mitotic figures numbering 06 mitoses / 10 mitoses; absence of perineural sheathing or vascular embolus.

Immunohistochemistry

- Hormone receptors and herceptest came back negative with a ki67 at 50%.
-Anti Cytokeratin K7 antibodies: Diffusely expressed by epithelial cells
-Anti-cytokeratin 5/6 antibodies: Diffusely expressed by myoepithelial cells.
-Anti PSl00 antibodies and anti p63 antibodies: focal positivity.
-Anti-Soxl0 antibody: Mosaic nuclear positivity.
-Anti CDI l7 c-kit antibodies: Diffuse membrane positivity.
Conclusion: Morphological appearance and immunohistochemical profile in favor of a breast adenoid cystic carcinoma.

An extension assessment including an abdominopelvic ultrasound and chest X-ray came back normal.

Diagnosis: Based on the clinical, radiological and pathological examination and taking into account the characteristics of ACK of the breast, given the unifocality and the size of the lesion, a diagnosis of ACK was made, classified T1N0M0 and a partial mastectomy indicated.

Therapeutic intervention: The patient was summoned for hospitalization the day before the surgical procedure. The first surgical step consisted of searching for the sentinel lymph node with indocyanine green by injecting 10 ml of indocyanine at 4 periaurolar points, then locating the lymphatic channel by transcutaneous fluorescence up to its termination in the axillary hollow. [Figure 1 and 2].

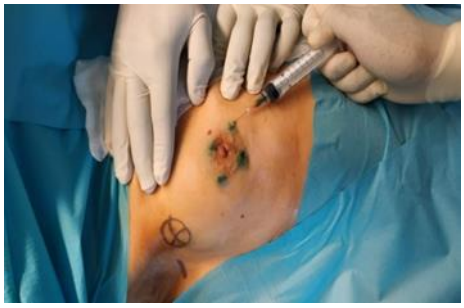

Figure1: injection of indocyanine green;

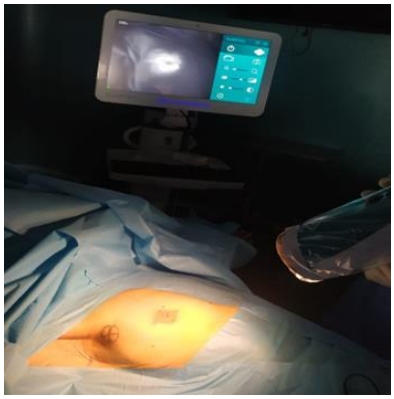

Figure2: location of the lymphatic channel by transcutaneous fluorescence;

An axillary skin incision was made opposite to the lymphatic canal, followed by the dissection, identification and removal of the fluorescent lymph node.[Figure 3].

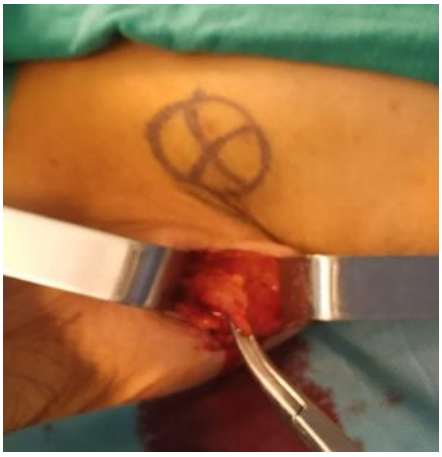

Figure3: Sampling of the fluorescent sentinel lymph node

The second surgical step was a wide tumorectomy after a peritumoral radial incision, subcutaneous detachment at the level of the Duret ridges and retroglandular detachmen t[Figure 4]. A wide excision of the tumor was performed, removing the aponeurosis of the pectoralis major muscle and the skin opposite the tumor. Glandular remodeling was performed after checking hemostasis.

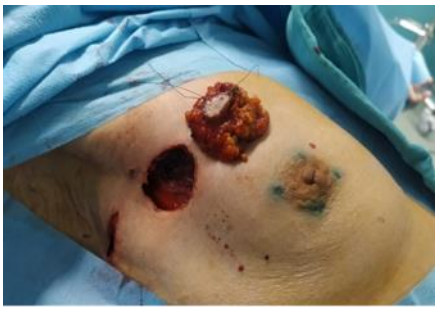

Figure4: Operating part with reference mark

The anatomopathological examination of the tumorectomy specimen confirmed the morphological and immunohistochemical appearance of an adenoid cystic carcinoma in its solid variant measuring 1.6 cm in long axis with healthy excision limits.
The sentinel node returned unharmed.

Monitoring and results of the surgical intervention: No surgical complications were noted during or after surgery. The patient was discharged the day after the procedure with painkillers and a prescription for local care. At the follow up appointment in 3 weeks, the clinical examination was unremarkable with a wound in the process of healing.

Given the final result of the anatomopathological study on the surgical specimen, the decision of the Multidisciplinary Consultation Meeting was to complete the surgery with a course of local radiotherapy.

Informed consent: The patient has given her full consent for the publication of her case.

III. Discussion And Literature Review

Breast ACK is a rare and unusual tumor and represents approximately 0.1 to 1% of breast cancers. [1, 6] It is a tumor that mainly affects women, but a few cases have been reported in men.[7] The age of onset is generally similar to that of invasive ductal carcinoma. [2] However, Arpino et al. found in their series a higher mean age (66 years) compared to invasive ductal carcinoma (62 years). Breast ductal carcinoma is exceptional before menopause.[1] The age range is 19-84 years. [2, 4]In our observation, the patient was 78 years old.

Clinically, CAK of the breast is revealed by a nodule usually located in the centromammmary region, sometimes bilateral or multifocal without predilection for one side.8] Classically the nodule is well circumscribed, firm and mobile, most often suggesting a fibroadenoma. [5, 2]In the literature, cases of CAK of

the breast presenting with skin infiltration, nipple retraction and discharge or invasion of the pectoral muscle are very rarely reported. It is also unusual to find axillary adenopathy.[1,9]

The iconographic diagnosis (mammography, ultrasound and magnetic resonance imaging) of breast CAK is difficult.10.11] The mammographic aspects are not specific and these lesions often appear in the form of more or less well-circumscribed lobulated nodules of homogeneous density.12] Sometimes, more or less architectural distortions are observed but microcalcifications are rarely encountered during a mammogram.13] The differential diagnosis is difficult with fibroadenoma, mucinous carcinoma or even infiltrating ductal carcinoma (IDC).14]

The diagnosis is usually made by microbiopsy.15] The histological appearance is comparable to that of adenoid cystic carcinomas observed in other locations which is essentially based on the biphasic nature of the population but the primary breast location usually has a favorable prognosis.[16]Architectural proliferation is variable: glandular, basaloid tubular, trabecular, solid, cribriform with pseudocysts; solid foci represent less than 10% of tumor tissue.[8,12,17]Thus, Ro et al, based on the classification of salivary gland CAKs, suggested that breast CAK could be classified according to the proportion of this solid component: (grade I: no solid elements, grade II: less than 30% solid elements, grade III: greater than or equal to 30%).Thus, they propose three therapeutic strategies: a tumorectomy without curettage for grades I, a simple mastectomy for grades II, and a radical mastectomy and axillary lymph node curettage for grades III.18] Furthermore, in the series of Defaud-Hénona F. et al, three out of four patients with very massive architecture developed local or general relapses without having a particular immunohistochemical profile.[19]

The immunohistochemical profile of mammary CAK shows that basaloid cells resembling myoepithelial cells (p63, smooth muscle actin, vimentin, calponin, etc.) exhibit variable positivity for high molecular weight keratins, whereas glandular cells resembling luminal cells express pancytokeratins but generally do not express hormone receptors.[6,19] On the other hand, about twenty cases of positive estrogen receptors have been reported in the literature, but the diagnostic criteria and the method of studying estrogen receptors raise reservations and rereading the slides could allow other differential diagnoses to be ruled out such as collagenous spherulosis, adenomyoepithelioma, cribriform ductal carcinoma and cylindroma. [1,20,21,22] In our observation, the hormone receptors are negative and this excludes hormone therapy as adjuvant treatment.
HER-2neu expression has been little studied for CAKs, but it is negative in our observation. [23.24]

CAK meets the same diagnostic criteria as triple-negative carcinoma of basal phenotype [ER-, RP-, HER-2-neu-, CK5/6+, cKIT+] but differs from the latter by the presence of a dual glandular and myoepithelial cell population.[24,25] Nevertheless, their prognosis remains clearly good compared to that of triple negative ICC. This would be linked to an underexpression of genes related to migration, proliferation and the immune responsein these tumors. However, the literature reports other metachronous or synchronous tumors of adenoid cystic carcinoma, such as ICC, adenomyoepithelioma, Paget's disease of the nipple and adenosquamouscarcinoma. [2,10.24]

Therapeutic management is not yet standardized because of the rarity of breast CAK. Nevertheless, some authors recommend conservative treatment such as tumorectomy followed by radiotherapy. [26] In the series of Arpino et al, no patient treated by tumorectomy who received post-operative radiotherapy developed recurrences..[1]

Overall, the recurrence rate is low, varying from 7.7 to 31% according to the literature. This would be linked either to incomplete surgical excision or to conservative treatment without radiotherapy.[1, 2]
Other authors prefer a mastectomy from the outset to ensure local control.[1.6]

In the literature, lymph node metastases are rare, they are reported in 0.8% to 6.7%.[1,5,18]In the series of Defaud-Hénona F. et all, no lymph node curettage was positive[19]. Lymph node curettage would therefore be of no benefit; however, examination of the sentinel lymph node remains beneficial.

In our observation, we performed a wide tumorectomy with search and examination of the sentinel node, which returned negative.

Unlike other extramammary adenoid cystic carcinomas, distant metastases are rarely found with a rate of 8.1% after lumpectomy and 8.3% after mastectomy. This shows that local surgical treatment would not have impact on the risk of metastasis or survival.27.28]

The contribution of adjuvant chemotherapy in the management of breast CAK is not significant; there would be no difference in survival between recipients and non-recipients of adjuvant chemotherapy.[1]Distant metastases can be found in the absence of lymph node involvement and they can occur after more than 15 years, hence adequate monitoring is recommended.29] Given the more frequent pulmonary involvement and metastases from other sites [liver, brain, kidney, pleura, spine, femur and skull], a chest X-ray, a physical examination for local risk and an annual mammogram are recommended annually, especially in the case of conservative treatment.30] Considering the longevity of survival and the occurrence of local recurrences and metastases, Arpino et al. suggest a long-term follow-up of up to ten years.[1]

Overall survival is close to that of the general population, even after conservative treatment with survival rates of 100% at 5 years and 93.8% at 10 years in the series of Arpino et al.[1,31]

IV. Conclusion

CAK of the breast is a rare tumor with a good prognosis. Its diagnosis is anatomopathological based on morphological study coupled with immunohistochemistry. Therapeutic management is still poorly established. Local treatment with examination of the sentinel lymph node is suggested, supplemented by postoperative radiotherapy which improves the local control rate. Chemotherapy has not proven any significant benefit in management. However, standardization of therapeutic management is necessary for a better understanding of their prognosis.

Conflicts of interest

The authors declare no conflict of interest.
All authors approved the final version of the manuscript.

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