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The Challenge of Distinguishing Cylindroma of the Breast from Adenoid Cystic Carcinoma: A Case Report and Review of this Rare Tumor

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ABSTRACT

Background: Cylindroma of the breast is a rarely described tumor that closely resembles its dermal counterpart.

Case presentation: We report a case of a 55-year-old woman with an incidental finding of a left breast mass on screening mammography. On histologic examination, the tumor was found to be consistent with cylindroma of the breast.

Conclusions: Generally considered to be a benign lesion, cylindroma of the breast may be challenging to differentiate from morphologically similar tumors, such as solid type adenoid cystic carcinoma, and this uncertainty may lead to a dilemma in treatment options. These patients should be followed closely, as long-term outcome data is not available given the uncommon occurrence of these lesions within the breast.

Introduction

Cylindroma of the breast (CB) is a particularly rare lesion. It has been considered to be a benign lesion, similar to its dermal counterpart, and is often referred to as a dermal analogue tumor. Dermal cylindromas are more common, usually found as a solitary lesion, but may occur as multiple lesions in patients with Brooke-Spiegler Syndrome (BSS). Individuals with BSS develop multiple skin tumors such as cylindromas, spiradenomas, and trichoepitheliomas due to a mutation in the CYLD1 tumor suppressor gene that results in an error in apoptosis regulation. These tumors usually originate in hair follicles or sweat glands and are most often benign but may be malignant.[1-7] While dermal cylindromas are usually found on the head and neck, CBs arise from the breast parenchyma without involvement of the overlying skin, which is not surprising as breast tissue originates from the primordium that gives rise to the cutaneous apocrine glands.[1-7] Cylindromas may also be found in the salivary glands, lungs and kidneys. We report an individual case of an asymptomatic breast tumor discovered on screening mammography and discuss the challenge of differentiating CB from adenoid cystic carcinoma. We review the current literature and the difficulty of establishing a definitive diagnosis and treatment plan.

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Case Presentation

A 55-year-old female with a history of bilateral subglandular saline breast implants presented with an asymptomatic abnormal finding on initial screening mammography classified as BIRADS 4 (Figure 1-A). On exam, she was found to have a well-circumscribed 8mm palpable mass located in the 10 o'clock position, approximately 5 cm from the nipple without involvement of the overlying skin. There was no lymphadenopathy.Ultrasound confirmed a 0.8 cm lobulated hypoechoic mass in the left upper inner quadrant with irregular borders and increased vascularity (Figure 1-B). An excisional

biopsy was recommended due to the close proximity of the lesion to the implant.

The patient had a significant maternal family history of cancer, including a perimenopausal aunt with breast cancer. Her mother and other maternal aunts, uncles, and cousins were diagnosed with colon, lung, prostate, brain, and unknown primary cancers between the ages of 40-60 years. Hereditary risk and genetic testing were reviewed and genetic screening revealed a PMS2 gene mutation, which is linked to Lynch syndrome. This mutation results in dysfunction of a DNA mismatch repair gene that increases the lifetime risk of colorectal, gynecologic, gastrointestinal, brain, sebaceous and various other types of cancer.

An ultrasound guided excisional biopsy revealed a low grade basaloid tumor of uncertain malignant potential with an involved margin, consistent with but not definitively, a cylindroma (Figure 2). The receptor status was ER-neg, PR-neg, Her2/neu-neg. The cells stained positive for p63 at the periphery. Due to the histologic similarity to adenoid cystic carcinoma (ACC), the decision was made to return to the operating room for re-excision. No residual tumor was seen on pathology.

In considering evaluation and/or treatment of the axilla, a review of the literature describes CB as a benign lesion treated with wide local excision alone. However, some authors consider CB as a variant of low-grade ACC with relatively indolent behavior.[8] Reports of low-grade ACC noted lymph node involvement in less than 2% of patients, usually in tumors greater than 2 cm. Therefore, a sentinel lymph node biopsy was not performed. Medical and radiation oncology consultations were completed postoperatively. Further testing included a colonoscopy that was normal as well as a CT scan of the chest, abdomen and pelvis with no evidence of synchronous lesions or metastasis. Systemic therapy was not indicated. However, as decreased local recurrence with postoperative radiation in the ACC population has been demonstrated, and because recurrence may be difficult to detect early on mammography due to shielding from the breast implant, adjuvant radiation therapy was recommended in the face of the patient's gene mutation. The patient tolerated 50 Gy of external beam radiation to the tumor bed and remains well 4 months after treatment. Close follow-up is planned with physical examination and yearly mammogram.

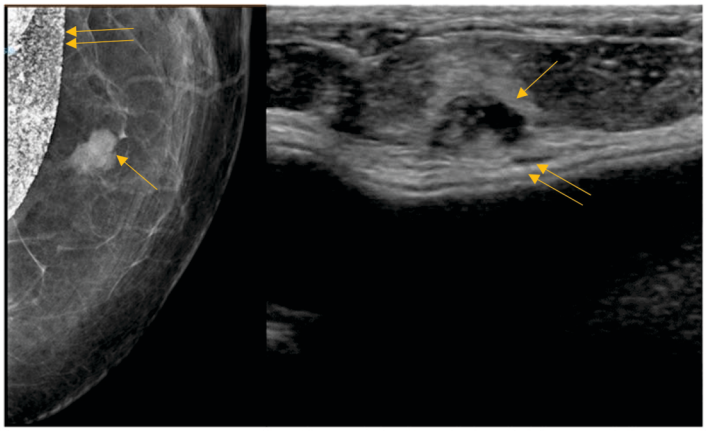

Figure 1. A) Spot compression craniocaudal view shows a lobulated mass (arrow) in the upper inner quadrant adjacent to the silicon implant (double arrow) B) Ultrasound shows a lobulated hypoechoic solid mass (arrow) with irregular borders closely adherent to the implant capsule (double arrow)

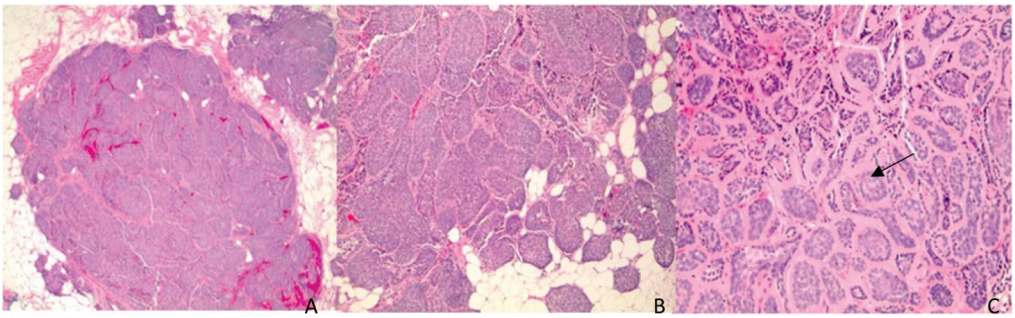

Figure 2. A) Low power image shows the relatively well circumscribed nature of the tumor composed of small, uniform basaloid cells B) Jigsaw pattern of growth with focal infiltration into fat C) Prominent hyaline basement membrane material (arrow) surrounding nests of basaloid cells

Discussion

Cylindroma of the breast often has borderline histologic features similar to those of adenoid cystic carcinoma, posing a dilemma for the clinician in predicting the benign or malignant behavior of these tumors, resulting in the potential for under or over-treatment. The rare nature of these tumors makes it difficult to accumulate adequate clinical data for physicians to help patients make informed decisions on the best course of treatment[8]

A cylindroma originating in the breast is rare with only 15 reported cases in the literature. Accurate identification can be challenging. Distinguishing CB from solid variant ACC, basaloid type is paramount to guiding treatment. CB classically displays well circumscribed, non-encapsulated nests and trabeculae of central basaloid cells and peripheral myoepithelial cells arranged in a characteristic jigsaw pattern surrounded by a thick periodic acid Schiff positive basement membrane that is immunoreactive for collagen IV. Immunohistochemical analysis typically demonstrates central basaloid cells that stain positive for cytokeratin 7 (CK 7) , eccrine ducts that stain positive for CEA, peripheral myoepithelial cells that stain positive for p63, and Langerhans cells that stain positive for S100 protein. ACC similarly demonstrates nests and trabeculae of basaloid cells, as well as negative receptor status for estrogen, progesterone and Her2/neu receptors. However, it lacks the thick, continuous basement membrane and Langerhans cells. It also shows nuclear atypia, mitotic figures, invasive growth patterns, and possible mucin production.[1-8]

While ACC has an excellent prognosis, it may manifest an aggressive and malignant course. Patients with local recurrence and distant metastasis have been reported; therefore, it is generally treated by mastectomy or lumpectomy with adjuvant radiation therapy. CB, however, is thought to be a benign tumor, treated with wide local excision alone.[6] Fusco *et al*. recently proposed an additional method for differentiating CB from ACC using molecular analyses in addition to histological and immunohistochemical evaluations. They utilized whole-exome sequencing to confirm a clonal somatic CYLD site mutation, solidifying the diagnosis of CB as opposed to ACC.[9] This added diagnostic evaluation may play a key role in selecting the proper treatment for patients where tumor identification is in doubt, such as in our case where, after multi-specialist review and discussion with our patient, the tumor was treated as ACC by the radiation oncologist.

Most authors agree that cylindroma of the breast was first described by Gokaslan *et al*. in 2001.[1] Our review of this and all subsequent case reports found the mean follow up period was 22 months (6 months-5 years, although several (n=8) of the reported cases do not specify follow up data). Aside from a single report of a CB malignancy published in 1939 by Nayer (likely not a cylindroma), no metastasis or recurrence has been reported for confirmed CB.[2,3,5-7,10]

Interestingly, of the 15 cases reviewed, 5 women were found to have synchronous invasive breast cancer (2 invasive lobular, 3 invasive ductal), more than triple the population incidence for synchronous breast cancer. While the cylindroma was an incidental finding on final pathology in these cases, it does raise questions about a possible association of this rare tumor with other malignant lesions. Bilateral breast MRI screening was considered but in light of limited data in these uncommon cases and plans for close follow-up, it was not recommended in this setting.

In conclusion, breast cylindroma is a rare tumor, and there is limited data to predict the long-term behavior and malignant potential of this tumor. Similarities with adenoid cystic carcinoma make the correct diagnosis challenging in some cases. Further investigation and follow up of women with cylindroma of the breast will be necessary to provide the best treatment and prognosis for patients.

Conflict of Interest

We have no financial or personal relationships to disclose. There is no funding source. Consent was not required for this paper.

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