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A Case with Suspected Breast Malignancy: A Rare Skin Appendage Tumor Spiradenoma

Meme Malignite Şüpheli Olan Olguda: Nadir Bir Deri Eki Tümörü Spiradenom

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Abstract: Spiradenomas are rare and benign cutaneous tumors originating from sweat glands. Eccrine spiradenoma cases in the breast are quite rare and there are only a few cases in the literature. In our case, a 24-year-old female patient who underwent surgery due to her mother's breast cancer presented with complaints of pain and swelling in her left breast. The patient's breast ultrasonography, breast magnetic resonance imaging and tru-cut assisted results are being investigated for suspicious findings for malignancy. Surgical excision complications occurred for definitive diagnosis and spiradenoma was reported as a skin appendage tumor in the pathological examination. Spiradenoma has not been reported in the literature in cases with high suspicion of breast malignancy. In such cases, therapeutic treatment options of surgical resection, considering the negative margins, are emphasized.

Keywords: Spiradenoma, Breast, Malignancy Suspicion

Özet: Spiradenomlar, ter bezlerinden kaynaklanan nadir ve iyi huylu kutanöz tümörlerdir. Memedeki ekrin spiradenom vakaları ise oldukça nadirdir ve literatürde yalnızca birkaç vaka bildirilmiştir. Olgumuzda, annesi meme kanseri nedeniyle cerrahi müdahale geçiren 24 yaşında bir kadın hasta, sol memede ağrı ve şişlik şikayetleriyle başvurmuştur. Hastanın meme ultrasonografisi, meme manyetik rezonans görüntüleme ve tru-cut biyopsi sonuçlarında malignite açısından şüpheli bulgular saptanmıştır. Kesin tanı için cerrahi ekzisyon biyopsisi yapılmış ve patolojik incelemede deri eki tümörü olarak spiradenom ön planda raporlanmıştır. Literatürde, meme malignite şüphesi yüksek olan vakalarda spiradenom daha önce rapor edilmemiştir. Bu tür vakalarda, sınır negatifliği göz önünde bulundurularak yapılan cerrahi rezeksiyonun hastanın küratif tedavisini sağladığı vurgulanmaktadır.

Anahtar Kelimeler: Spiradenom, Meme, Malignite Şüphesi

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1. Introduction

Spiradenoma is one of the benign neoplastic tumors of the sweat glands, usually located in the upper layers of the skin, superficial and deep dermis. This tumor, which is mostly seen on the trunk and extremities, rarely occurs in the breast region (1). Spiradenomas, which appear as breast masses, may be similar to other benign breast lesions in terms of clinical and histological aspects. Therefore, biopsy and histopathological examination are of great importance for the correct diagnosis. Spiradenomas usually present as hard, painful and mobile masses, and the size of the lesion usually varies between 1-2 cm (2). These lesions can be confused with other benign lesions such as fibroadenoma, lipoma, epidermoid cyst and dermoid cyst, and with malignant lesions. Therefore, in patients with breast lesions, the differential diagnosis of spiradenoma, which is a skin appendage tumor, should be considered, although rarely.

In this case report, a patient with a family history of breast cancer who underwent excisional biopsy due to a breast mass, which was evaluated as spiradenoma as a result of the biopsy, is presented.

2. Case Report

A 24-year-old female patient presented to an external center due to complaints of pain and swelling in the upper outer quadrant of her left breast and her mother's history of breast cancer. Breast ultrasonography performed at the external center revealed a hypoechoic solid lesion of 11x6 mm with regular borders and distinct lobulated contours at the 1 o'clock position in the right breast. It was determined that this lesion was continuous with the ducts and could be significant in terms of intraductal space-occupying pathologies causing fibroadenoma and ductal ectasia in the differential diagnosis. In addition, an anechoic lesion of 20x10 mm with regular borders and lobulated contours and venous and arterial flow was observed in the skin and subcutaneous tissue at the 11 o'clock position in the left breast. It was stated that vascular pathologies such as arteriovenous malformation (AVM) or hemangioma should be considered as a priority in the differential diagnosis of this lesion, but that histopathological diagnosis was necessary. Thereupon, the patient underwent a dynamic contrast-enhanced breast magnetic resonance (MRI) examination. In breast MRI examination, a 16 mm lobulated contoured and enhancing BI-RADS-4 lesion area limited to the skin and subcutaneous areas in the upper outer quadrant of the left breast was observed and histopathological correlation was suggested. In the right breast, an 8 mm sized, well-enhancing benign lesion was observed (Figure 1).

Thereupon, the patient applied to our clinic for treatment planning after the tru-cut biopsy performed by an external center for the lesion in the left breast was evaluated as adenomyoepithelioma and intraductal papilloma. First, the patient's biopsy preparations were re-examined by us. In the material taken, tumor foci were observed in fragmented tissues showing high cellularity but no nuclear atypia and mitoses. The inner part of the tumor consisted of cells with pale wide cytoplasm and outer basaloid cells with narrow cytoplasm. In immunohistochemical examinations, outer basaloid cells were observed to be positive for p63 and inner cells were observed to be positive for high molecular weight cytokeratin (HMWCK) (Figure 2 a-d).

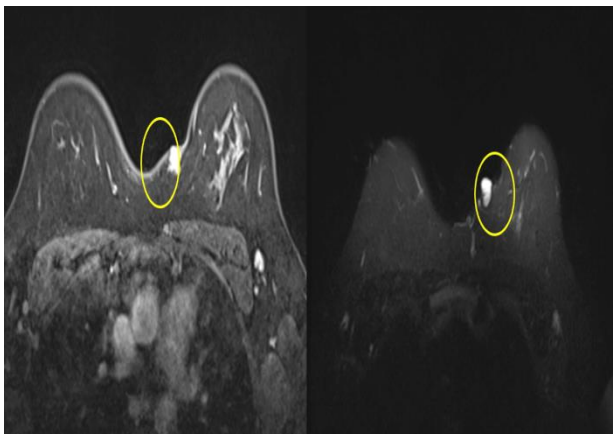

Figure 1. *Breast MRI Image (The 16 mm lobulated contour and contrast enhancement BI-RADS-4 lesion area limited to the skin and subcutaneous areas is circled in yellow)*

Morphological and immunohistochemical findings suggested a tumor composed of epithelial and myoepithelial cells. Although the needle biopsy result was evaluated as adenomyoepithelioma as a benign epithelial tumor, excisional biopsy was recommended for definitive diagnosis. Thereupon, the patient was decided to undergo excisional biopsy.

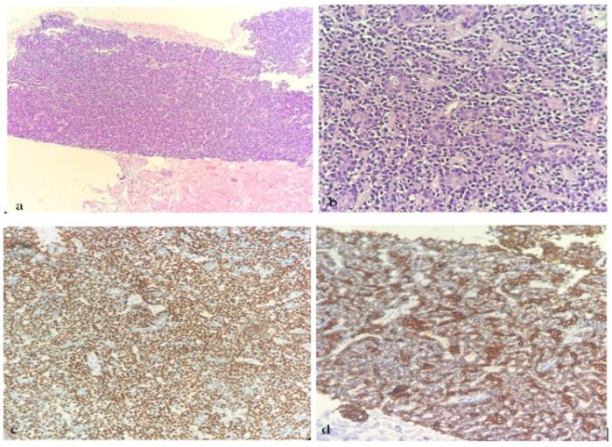

Figure 2 a-b. *Tru cut needle biopsy (x100, H&E) (H&Ex400), 1c: p63 positivity in dark outercells (x200) 1d: HMWCK positive in inner pale cells (x200)*

After obtaining detailed surgical consent for the surgery, the patient underwent a segmental mastectomy with an elliptical incision from the lesion located at the 11 o'clock position on the left breast under general anesthesia and was sent for frozen examination.A well-differentiated tumoral lesion with cribriform and solid pattern was observed in the frozen examination and solid papillary carcinoma, encapsular papillary carcinoma and neuroendocrine tumor were considered in the differential diagnoses. It was recommended that the definitive diagnosis be made after formalin follow-up. Since there was no tumor in the surgical margins, the reconstruction was completed and the operation was terminated. The patient, whose postoperative follow-up was unremarkable, was discharged on the 2nd day.In the final pathology of the segmental mastectomy material removed after formalin follow-up; no macroscopic lesion was observed except for a slight brown color change on the skin. In serial sections, a gray-white solid tumor measuring 10x8 mm with a multinodular pattern was detected under the skin. The entire tumor was sampled and its cellular characteristics were found to be compatible with needle biopsy. In

immunohistochemical examinations, more widespread positivity was detected with p63 and HMWCK. (Figure 3-a, b). While calponin and S100 staining is expected in the myoepithelial cell layer in adenomyoepithelioma cases, these two markers were found to be negative. In addition, a ductal staining pattern indicating eccrine differentiation was observed in the examination performed with epithelial membrane antigen (EMA) (Figure 3-c). When the mild color change described in the skin, the localization of the tumor in the subepidermal area, the multinodular pattern and immunohistochemical findings were evaluated together, it was thought that the case was compatible with a skin appendage tumor and primarily spiradenoma.

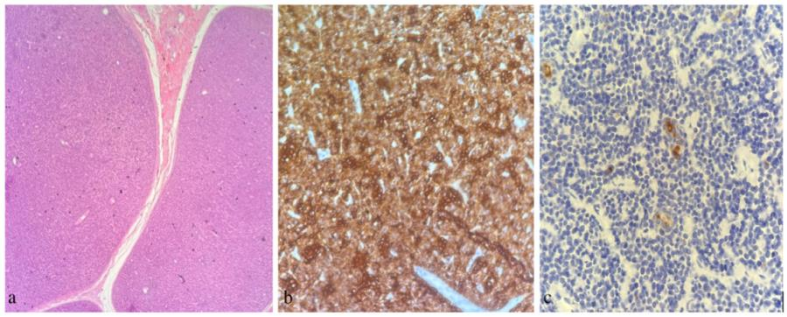

Figure 3a. Multinodular tumor (H&E, x100), 3b: HMWCK diffuse positive (x200), EMA ductal patterns positivity (X200)

3. Discussion

Spiradenomas are benign tumors that usually occur in the head, neck and trunk, and involve subcutaneous and dermal tissues. They are exceptionally uncommon in the breast; to date, only six such cases have been documented. (3, 4). These tumors can develop at any age in both sexes, with a higher prevalence in individuals between 20 and 40 years old. The likelihood of malignant transformation increases after the age of 50 (5, 6). Clinically, spiradenomas are solitary in approximately 97% of cases, although multiple lesions can occur, particularly in women (7). Their sizes can vary between 1 and 10 cm, but they are usually smaller than 1 cm (8). Although pain is an important symptom in patients, it is not seen in all patients. On physical examination, pathognomonically, there are typically hard swellings of blue or gray color under 1 cm (9). Although malignant changes of spiradenoma are extremely rare, systemic metastasis has been reported (1, 10).

Because spiradenoma is quite rare in the breast, it can be clinically confused with other superficial masses. Therefore, both benign and malignant superficial soft tissue tumors and other skin appendage tumors should be excluded from the differential diagnosis. Important options include hemangioma, vascular leiomyoma, lymphoma, metastatic masses, epidermal inclusion cyst, angiomyomyoma, glomus tumor, schwannoma, cutaneous endometriosis, dermatofibroma, and basal cell carcinoma. In addition, rare cutaneous adnexal tumors such as cylindrical tumors and hidradenoma should be included in the differential diagnosis. (11-14).

The fact that these lesions are usually small and unifocal provides a clue to the diagnosis; however, definitive diagnosis is made by surgical excision and histopathological examination.

Primary cancers originating in the breast tissue and malignant tumors involving the skin can be confused clinically and radiologically. In cases where differential diagnosis is difficult, definitive diagnosis is achieved by surgical excision of the lesion (15). Spiradenomas are rare, and due to the limited number of studies conducted on this subject, their likelihood of detection with imaging methods is low. However, their

spread to the dermis and superficial subcutaneous fat is a diagnostic finding (16).

In our case, the initial diagnosis was a hemangioma on ultrasound. Hemangiomas can be confused with breast cancer on breast ultrasound; therefore, some studies have suggested that breast MRI may be helpful in making the diagnosis before surgical excision in such cases (17). Epidermal inclusion cysts and eccrine spiradenomas are the lesions most often confused with each other. MRI findings are distinct, and these findings can be used in differential diagnosis.(18).

The curative treatment option for spiradenomas is surgery. Early surgical excision immediately after the lesion is identified is recommended for both diagnostic and therapeutic purposes due to the risk of malignant transformation. A high risk of local recurrence has been reported in cases with incomplete surgical excision (19). Negative surgical margins have been shown to contribute positively to prognosis (20).

Primary breast cancers and skin appendage tumors can be confused pathologically. Although such lesions can be distinguished clinically and radiologically, pathological recognition and correct interpretation are very important. Since skin appendage tumors of the breast are rare, making a correct diagnosis requires great care. In our case, adenomyoepithelioma diagnosis was considered to be the primary diagnosis among skin appendage tumors of the breast. Adenomyoepithelioma is a rare benign breast tumor in which myoepithelial cells predominate. The most obvious difference between spiradenoma and adenomyoepithelioma is the differences in their histological structures. Adenomyoepithelioma exhibits a structure in which epithelial and myoepithelial cells coexist, and myoepithelial cells show a clear distinction between the basal layer and epithelial cells. However, in spiradenoma, the tubes of the apocrine sweat glands and the density of basal cells are more pronounced.

It has also been reported in the literature that spiradenomas, similar to adenomyoepithelioma, can be confused with epidermal inclusion cysts (21). Cases diagnosed as epidermal inclusion cysts on tru-cut biopsy results and spiradenomas on final pathology reports are reported in the literature. These findings emphasize the difficulty of differential diagnosis of skin appendage tumors and the importance of accurate pathological evaluations (4).

Although the preoperative tru-cut biopsy in our case was consistent with adenomyoepithelioma, the primary pathological evaluation was consistent with spiradenoma. Immunohistochemical markers such as calponin, actin, p63, and S100, which help identify myoepithelial cells, play an important role in the diagnosis of these tumors (22). While S100 positivity may increase the likelihood of adenomyoepithelioma due to involvement of the myoepithelial layer, S100 was reported negative in our case (23). However, p63 and calponin positivity are among the strongest findings supporting the diagnosis of spiradenoma (24-26). Furthermore, immunohistochemical studies of cytokeratins suggest that spiradenomas are of eccrine origin (27).

Conservative surgical excision is recommended for the treatment of spiradenomas. During excision, it is important to carefully define surgical margins, taking into account the malignant potential. Current literature emphasizes that surgical margins should be at least 1 cm, which aims to minimize the possibility of tumor recurrence (28). In addition, in cases of multiple spiradenomas, additional treatment with CO2 laser is recommended after surgical reduction (25).

In conclusion, skin appendage tumors, although rare, should be considered in the differential diagnosis of patients presenting with a palpable breast mass and pain, and in whom imaging and tru-cut biopsy fail to provide a definitive diagnosis. Histopathological and immunohistochemical evaluation are crucial for reaching a correct diagnosis. In our case, because clinical, radiological, and histopathological suspicions persisted, the lesion was surgically removed; final pathology revealed a rare spiradenoma, thus preventing disease progression and enabling curative surgical excision. It is noteworthy that breast spiradenomas are

extremely rare in the literature. Although malignancy was initially suspected in our patient, whose mother had recently been treated for breast cancer, our literature search did not reveal any cases of spiradenoma diagnosed with a family history of cancer. This case demonstrates that this very rare skin tumor should be considered in the differential diagnosis even in patients suspected of malignancy.

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