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A Rare Cutaneous Lesion Mimicking Breast Carcinoma: Pilomatrixoma: Case Report

Meme Karsinomunu Taklit Eden Nadir Bir Kutanöz Lezyon: Pilomatrikoma

ABSTRACT Pilomatrixoma is a rare benign cutaneous appendage tumor that has characteristic histopathological features. It is originated from the hair follicle. It usually arises in the head and neck region but rarely occurs in the other sites. Only a small number of breast pilomatrixomas have been reported up to date in the literature. Herein, a case of 71 year-old female with a mass detected in the left breast and diagnosed as pilomatrixoma is reported. Mammography showed a nodular lesion with ill-defined margins that contained coarse calcifications. In addition, the breast ultrasound revealed a hypoechoic lesion with irregular borders that showed posterior acoustic shadowing and internal echoes. Thus, the ultrasound findings were reported as BI-RADS 4, suspicious for malignancy. Although pilomatrixoma is a rare tumor that may occur in breast, it should be noted in the differential diagnosis of breast masses that contain calcifications by the radiologists and the clinicians.

Key Words: Breast; carcinoma; pilomatrixoma

ÖZET Pilomatrikoma karakteristik histopatolojik özellikler taşıyan nadir bir benign deri eki tümörüdür. Kıl follikülünden köken almaktadır. Genellikle baş ve boyun bölgesinde ortaya çıkmaktadır ancak nadiren diğer bölgelerde de görülebilmektedir. Literatürde bugüne kadar az sayıda meme yerleşimli pilomatrikoma bildirilmiştir. Burada, sol memesinde kitle saptanan ve pilomatrikoma tanısı alan 71 yaşında kadın hasta sunulmuştur. Mammografide kaba kalsifikasyonlar içeren düzensiz sınırlı hipoekoik nodüler bir lezyon gözlenmiştir. Ek olarak, meme ultrasonografisinde posterior akustik gölgelenme gösteren ve internal ekolar içeren, düzensiz sınırlı hipoekoik lezyon saptanmıştır. Nitekim, ultrasonografik bulgular malignite şüphesini belirten BI-RADS 4 şeklinde rapor edilmiştir. Pilomatrikoma memede gelişebilen nadir bir tümör olmakla birlikte, kalsifikasyonlar içeren meme kitlelerinin ayırıcı tanısında radyologlar ve klinisyenler tarafından akılda tutulmalıdır.

Anahtar Kelimeler: Meme; karsinom; pilomatrikoma

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Pilomatrixoma (pilomatricoma, trichomatrioma, calcifying epithelioma of Malherbe, benign calcifying epithelioma) is a rare benign tumor of skin that originates from the hair follicle and differentiates towards the matrix, inner sheath, and hair cortex. It was initially described by Malherbe and Chenantais in 1880, as a tumor arised from sebaceous glands and named as "calcifying epithelioma".¹ In 1961, Forbis and Helwig established that the origin of the tumor was the hair follicle, thus the name

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of "pilomatrixoma" was proposed.² Pilomatrixoma occurs at any age but mostly in the first two decades of life, and shows a female predominance. Pilomatrixoma arises most commonly in the head and neck region, and upper extremities. To the best of our knowledge, there have been only a few case reports of pilomatrixoma of the breast in the literature in both women and men.^{1,3-8} In addition, the mammographic and sonographic features of breast pilomatrixomas mimic malignant tumors due to the calcifications that they contain.¹ Histopathologically, pilomatrixoma is a relatively well-circumscribed, deep dermal or dermal-subcutaneous neoplasm that is characterised by basophilic cells derived from hair matrix cells and anucleated shadow cells, equivalent to the hair cortex.9 Also, intratumoral calcification may accompany more than two-thirds of the tumor, especially in the shadow cells.¹⁰ The size of tumor usually ranges from 0.5 cm to 3 cm, however rapidly growing giant tumors exceeding to 15 cm in diameter may be seen rarely.⁹

CASE REPORT

A 71-year-old female presented with a painful palpable mass in the upper outer quadrant of the left breast. On physical examination, red coloration and edema were observed on the overlying skin, and a superficial firm lump in the subcutaneous location of 2 cm in diameter was palpated. Mammography showed a nodular lesion with ill-defined margins that contained coarse calcifications just under the skin. Ultrasound examination revealed a hypoechoic lesion of approximately 2x1 cm with irregular borders that showed posterior acoustic shadowing and internal echoes. Also, the overlying skin was thickened and the subcutaneous adipose tissue adjacent to the lesion showed an increase in echogenicity, indicating an edema pattern. There was no pathological lymphadenopathy in the axillary region. The ultrasonographic findings were reported as BI-RADS 4, and it was noted that the findings were suspicious for malignancy and inflammation as well. Ultimately, an excisional biopsy was performed. Grossly, there were two specimens with dimensions of 1x0.5x0.5 cm and 0.7x0.5x0.3 cm. Microscopically, the biopsies were composed of skin and subcutaneous tissue, and free tumor fragments. Histopathologically, there were a few tumor nests consisting of anucleated eosinophilic shadow cells that caused marked giant cell reaction, infiltration of dense inflammatory cells, and granulation tissue in the both dermis and subcutaneous tissue (Figure 1). Also there were some calcifications and basophilic cells accompanying shadow cells both in the dermis and the free tumor fragments (Figure 2). No cellular pleomorphism, mitoses or necrosis was present in the lesion. Then, the histopathological findings were diagnosed as pilomatrixoma.

DISCUSSION

Any palpable breast mass can cause anxiety and suspicion for malignancy since the breast carcinoma is one of the most common cancer causing death. Herein we reported a satisfactory benign lesion, pilomatrixoma that showed calcifications mimicking breast carcinoma on mammographic and/or ultrasonographic examination. The characteristic ultrasonographic findings of pilomatrixoma, regardless of localization, are known as a well-circumscribed mass in deep dermis or subcutaneous tissue with targetoid appearence due to the central echogenic nidus and the hypoechoic

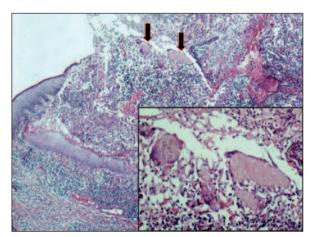


FIGURE 1: The ulcerated skin including infiltration of dense inflammatory cells, granulation tissue, and a few tumor nests composed of eosinophilic shadow cells (arrows) that caused marked giant cell reaction in the dermis (Hematoxylin and Eosin stain, x40). Inset: The higher power magnification of shadow cells and giant cell reaction (Hematoxylin and Eosin stain, x200). (See color figure at http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/)

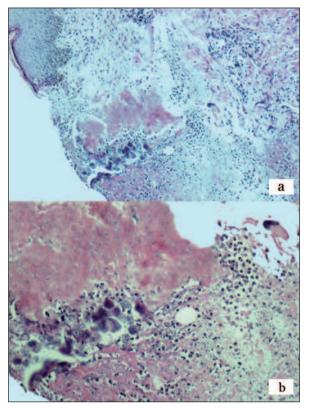


FIGURE 2: a-b. The basophilic cells accompanying shadow cells in the dermis with a giant cell and dense inflammatory cells (a. Hematoxylin and Eosin stain, x100, b. Hematoxylin and Eosin stain, x200). (See color figure at http://www.turkiyeklinikleri.com/journal/dermatoloji-dergisi/1300-0330/)

peripheral rim.¹¹ Central echogenic nidus indicates the epithelial component existing in the central part of the lesion and the hypoechoic peripheral rim indicates the pseudo-capsule composed of reactive fibrous tissue. Posterior acoustic shadowing due to calcification located in the central nidus is observed.¹¹

There are a few case reports about the radiological features of breast pilomatrixoma in the literature.^{1, 6-7} Gilles et al. and Reynaud et al. described the mammographic findings as a well-defined subcutaneous large nodule containing gross calcification.^{6,7} Imperiale et al. reported initially the ultrasonographic features of the breast pilomatrixoma as a hypoechoic nodule with posterior acoustic shadowing that were nonspesific and mimic breast cancer.¹ Hyper- to isoechoic acoustic shadowing and hyperechoic calcifications were detected in the literature as well. The radiological features of pilomatrixoma are different from case to case, attributed to the different stages of the lesion. In addition, variable histological findings may be observed due to the different stages of the development of it as well.9 Characteristically, welldeveloped pilomatrixomas have a transition from the peripheral basaloid cells to the central shadow cells. The amount of basaloid cells in the earlier lesion is more than the older lesion. In contrary, the amount of shadow cells in the older lesion is more than the earlier lesion.9 Inflammatory reaction with histiocytic giant cells, siderophages, melanophages, granulation tissue, ossification and transepidermal elimination may be present in the old regressing lesion as well.⁹ The histopathological findings of our case were consistent with an old regressing pilomatrixoma. The nonspesific radiological features of our case as irregular borders of the lesion can be attributed to the age of the lesion and to the destructive dense inflammatory reaction. The exact histopathologic diagnosis was not predicted before the operation because of the nonspesific radiological features of our case. Although it is rare in the literature, pilomatrixoma should be taken into consideration in the radiological differential diagnosis of breast carcinoma when calcifications are present especially just beneath the skin.

Surgical excision of pilomatrixoma is usually curative, however recurrences have been reported in some cases.9 Spontaneous regression may occur as well. Pilomatrixoma is a benign lesion but pilomatrix carcinoma, the malignant counterpart, has been reported recently.9,10 Active proliferating pleomorphic basaloid cells with conspicious nucleoli and numerous mitoses, invasion of blood vessels, necrosis and infiltration into surrounding tissues refer pilomatrix carcinoma.10 It should be kept in mind that pilomatrix carcinoma may occur de novo or secondary to pilomatrixoma, either by malignant transformation in a present pilomatrixoma or at the site of a previously excised lesion.¹⁰ Thus, pilomatrix carcinoma should be ruled out before diagnosing the lesion as pilomatrixoma histopathologically. Also, metastasis of some visceral tumors that can show shadow cell differentiation such as the tumors of the colon or the uterus should be ruled out as well.¹⁰

REFERENCES

- Imperiale A, Calabrese M, Monetti F, Zandrino F. Calcified pilomatrixoma of the breast: mammographic and sonographic findings. Eur Radiol 2001;11(12):2465-7.
- Yamauchi M, Yotsuyanagi T, Saito T, Ikeda K, Urushidate S, Higuma Y. Three cases of giant pilomatrixoma--considerations for diagnosis and treatment of giant skin tumours with abundant inner calcification present on the upper body. J Plast Reconstr Aesthet Surg 2010;63 (6):e519-24.
- Hubeny CM, Sykes JB, O'Connell A, Dogra VS. Pilomatrixoma of the adult male breast: a rare tumor with typical ultrasound features. J Clin Imaging Sci 2011;1:12. doi: 10.4103/ 2156-7514.76690.

- Hamilton A, Young GI, Davis RI. Pilomatrixoma mimicking breast carcinoma. Br J Dermatol 1987;116(4):585-6.
- Bhalotra R, Jayaram G. Fine-needle aspiration cytology of pilomatrixoma: a case report. Diagn Cytopathol 1990;6(4):280-3.
- Gilles R, Guinebretière JM, Gallay X, Vanel D. Pilomatrixoma mimicking male breast carcinoma on mammography. AJR Am J Roentgenol 1993;160(4):895.
- Reynaud P, Orliaguet T, Robin YM, Buono JP, Darcha C, Suzanne F, et al. [Mammary pilomatrixoma clinically mimicking carcinoma]. Ann Pathol 1997;17(3):213-4.
- 8. Viero RM, Tani E, Skoog L. Fine needle aspiration (FNA) cytology of pilomatrixoma: report

of 14 cases and review of the literature. Cy-topathology. 1999;10(4):263-9.

- Hurt MA, Kaddu S, Kutzner H. Benign tumors with follicular differentiation. In: Leboit PE, Burg G, Weedon D, Sarasin A, eds. World Health Organisation Classification of Tumours: Pathology and Genetics of Skin Tumours. Lyon: IARCH Press; 2006. p.152-8.
- Weedon D. Tumors with matrical differentiation. Weedon's Skin Pathology. 3rd ed. New York: Churchil-Livingstone Elsevier; 2010. p.768-70.
- Hughes J, Lam A, Rogers M. Use of ultrasonography in the diagnosis of childhood pilomatrixoma. Pediatr Dermatol 1999;16(5): 341-4.