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Extremely rare borderline phyllodes tumor in the male breast: a case report☆

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ABSTRACT

Phyllodes tumor of the male breast is an extremely rare disease, and far fewer cases of borderline phyllodes tumors than benign or malignant tumors in the male breast have been reported. We report a case of borderline phyllodes tumor in the male breast with imaging findings of the tumor and pathologic correlation.

1. Introduction

Phyllodes tumor of the breast is a neoplasm, accounting for less than 1% of all breast tumors and only 2–3% of fibroepithelial neoplasms [1]. Since Johannes Mueller first described it in 1838, it has been given diverse names, one of which is "cystosarcoma phyllodes" [2]. Phyllodes tumors have been classified into benign, borderline, and malignant categories, by the current World Health Organization (WHO) classification of breast tumors. The classification is based on the combination of various histological features such as mitotic count, margin type, stromal overgrowth, and degree of cellular pleomorphism [3]. It appears mainly in women, with only a few reports describing this breast neoplasm in men with preexisting gynecomastia. We now present a rare case of phyllodes tumor with borderline features in the male breast.

2. Case report

A 39-year-old man visited our hospital for a palpable left breast mass first noticed 20 days ago. Physical examination revealed a fixed hard mass on the subareolar area of the left breast without tenderness. Mammography revealed a 1-cm-sized circumscribed oval mass with equal density, located on the same site as the mass found during physical examination. The overlying skin was thickened, and nipple retraction was seen. There were no associated calcifications (Fig. 1). The lesion was diagnosed as Breast Imaging Reporting and Data System (BI-RADS) category 0; thus, ultrasonography (USG) was performed. USG found a 1.5-cm-sized indistinct oval heterogeneously echoic mass with mild edema, but there was no vascularity (Fig. 2). As per BI-RADS category 4B, a USG-guided core needle biopsy was performed.

Microscopic examination of the core biopsy specimen revealed chronic and active inflammation with infiltration of lymphocytes, histiocytes, neutrophils, and a few multinucleated giant cells (Fig. 3).

Due to the radiologic/pathologic discordance, a mass excision was performed. The excised left breast consisted of a 3.0×2.5 cm sized gray-whitish, multinodular mass with tense and elastic consistency. Histologic sections revealed a phyllodes tumor with a generally well-circumscribed border, moderate stromal cellularity, minimal stromal cell atypia, and absent stromal overgrowth. However, foci of permeative margins and increased mitotic activity [up to 9/10 high-power fields (hpf)] were found. Taken together, a pathologic diagnosis of borderline phyllodes tumor was made. Additionally, gynecomastia was noted with abortive lobule formations in the surrounding breast parenchyma (Fig. 4).

3. Discussion

Phyllodes tumor of the breast is rare and mainly observed in middle-aged women, with a widely varying mean age of from 30 to 52 years in the literature [2]. It is arise from stromal component and characterized by double-layered epithelial component arranged in clefts surrounded

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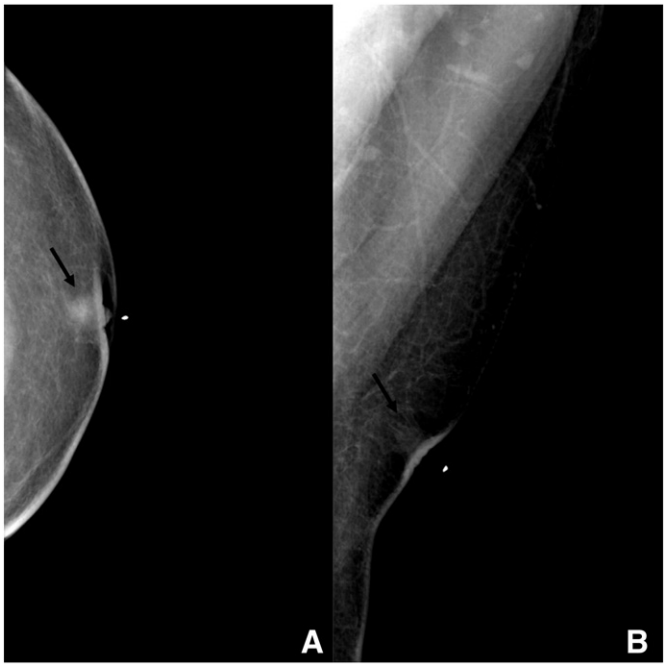

Fig. 1. (A) Craniocaudal and (B) mediolateral oblique mammogram view shows a 1-cm-sized circumscribed oval mass with equal density on the subareolar area of the left breast (arrow) with skin thickening and nipple retraction. There were no associated calcifications.

by a hypercellular mesenchymal component that is typically organized in a leaf-like pattern [4,5]. Although it is regarded as a hormonal disturbance and/or gynecomastia like male breast cancer, its pathobiology is considered different from that of breast cancer [6]. Gynecomastia is the most common clinical and pathologic abnormality of the male breast. Disproportion between estrogen and androgen is the main etiological element for gynecomastia and the imbalance may have a variety of causes such as hormone secreting tumors, hormonal therapy for prostate cancer, obesity, liver, thyroid, and renal diseases, or drug abuse [5]. Our patient was not obese and was not taking any medications. Routine laboratory findings were within normal limits; however, he was not evaluated for endocrine dysfunction.

Phyllodes tumors have been classified into benign, borderline, and malignant categories, by the current WHO classification of tumors of the breast. This classification is based on the combination of various histological features such as mitotic count, type of margin, stromal overgrowth, and degree of cellular pleomorphism [3].

Benign phyllodes tumor is defined as having 0–4 mitoses/10 hpf, pushing margins, and minimal or moderate stromal overgrowth with minimal stromal cellularity and atypia [7]. On mammography, it mostly appears as a spherical or phylloid nonspiculated marginated mass without calcification. On USG, it appears as a well-defined oval hypoechoic mass with or without posterior enhancement [8]. Since intratumoral cystic spaces presenting as a horizontal linear cleft, slit-like cleft, or anechoic variable-sized cystic lesions can be seen, it is helpful to differentiate phyllodes tumor from other benign breast tumor such as fibroadenoma that shows similar spaces [8].

Borderline or low-grade malignant phyllodes tumor is defined as having 5–9 mitoses/10 hpf, pushing or infiltrating margins, moderate stromal cellularity, and atypia, with overgrowth. Malignant or high-grade phyllodes tumor is defined as having >10 mitoses/10 hpf, infiltrating margins, moderate to marked stromal cellularity, and atypia, with overgrowth [7]. Borderline and malignant phyllodes tumors are larger than benign tumors and more frequently found with cleft or cystic change, but the difference in frequency is not statistically significant [9,10]. On mammography, phyllodes tumors tend to have well defined margins than ill-defined margins. On USG, borderline and malignant phyllodes tumors are observed as well-defined oval lobulated masses with posterior enhancement, like in benign phyllodes tumors [8].

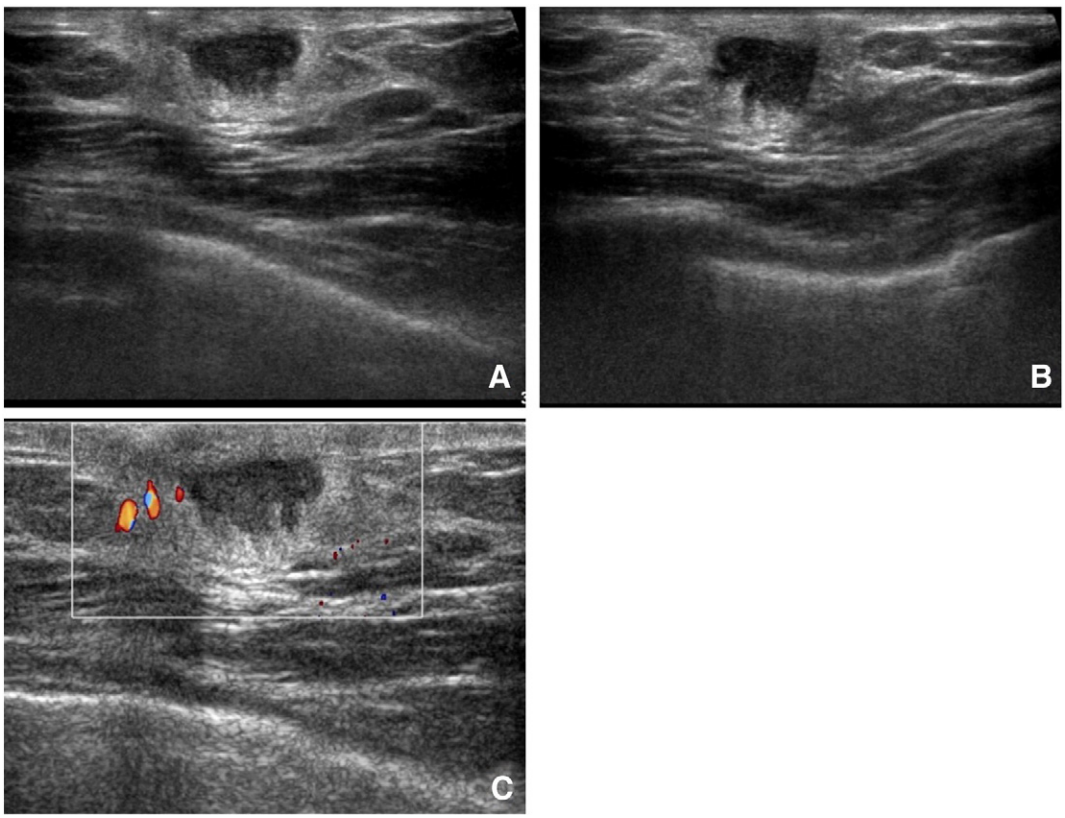

Fig. 2. Transverse (A), longitudinal (B), and color doppler (C) USG reveals a 1.5-cm-sized indistinct oval heterogeneously echoic mass with mild edema, but with no vascularity.