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Case Report

Bilateral diffuse tumorous pseudoangiomatous stromal hyperplasia treated with bilateral mastectomy in a 40-year-old woman

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ABSTRACT

Pseudoangiomatous stromal hyperplasia (PASH) is a rare benign breast disease characterized by breast stromal proliferation mimicking low grade angiosarcoma. PASH is frequently detected as an incidental component coexisting with other breast lesions. However, it can also present as a single localized mass and is typically unilateral. Mammographic and sonographic findings are nonspecific and indistinguishable from those of benign lesions. We report an unusual case of PASH presenting with diffuse enlargement of the bilateral breasts in a 40-year-old woman treated with bilateral mastectomy.

Introduction

Pseudoangiomatous stromal hyperplasia (PASH) is a benign mesenchymal proliferative lesion of the breast that was first described by Vuitch et al., in 1986. PASH can affect patients in any age group, although it is more common in premenopausal women. The clinical presentation of PASH ranges from the frequently encountered microscopic focus to a discrete mass. A microscopic focus is a relatively common incidental finding in breast specimens from various benign or malignant lesions [1]. PASH can manifest as a well demarcated mass (referred to as tumorous PASH), and most of these cases present as a single unilateral mass [2]. Tumorous PASH can also present as diffuse massive enlargement of the breast; however, this presentation is extremely rare [2,3]. We herein report a case of PASH presenting as diffuse bilateral breast enlargement requiring bilateral mastectomy.

Case report

A 40-year-old woman presented with progressive bilateral breast enlargement for approximately 1 year. She denied any family history of breast diseases, and did not take oral contraceptives or hormonal medications. Physical examination revealed marked enlargement of both breasts with massive bilateral palpable masses accompanied by breast tenderness. Mammography revealed marked enlargement of both breasts

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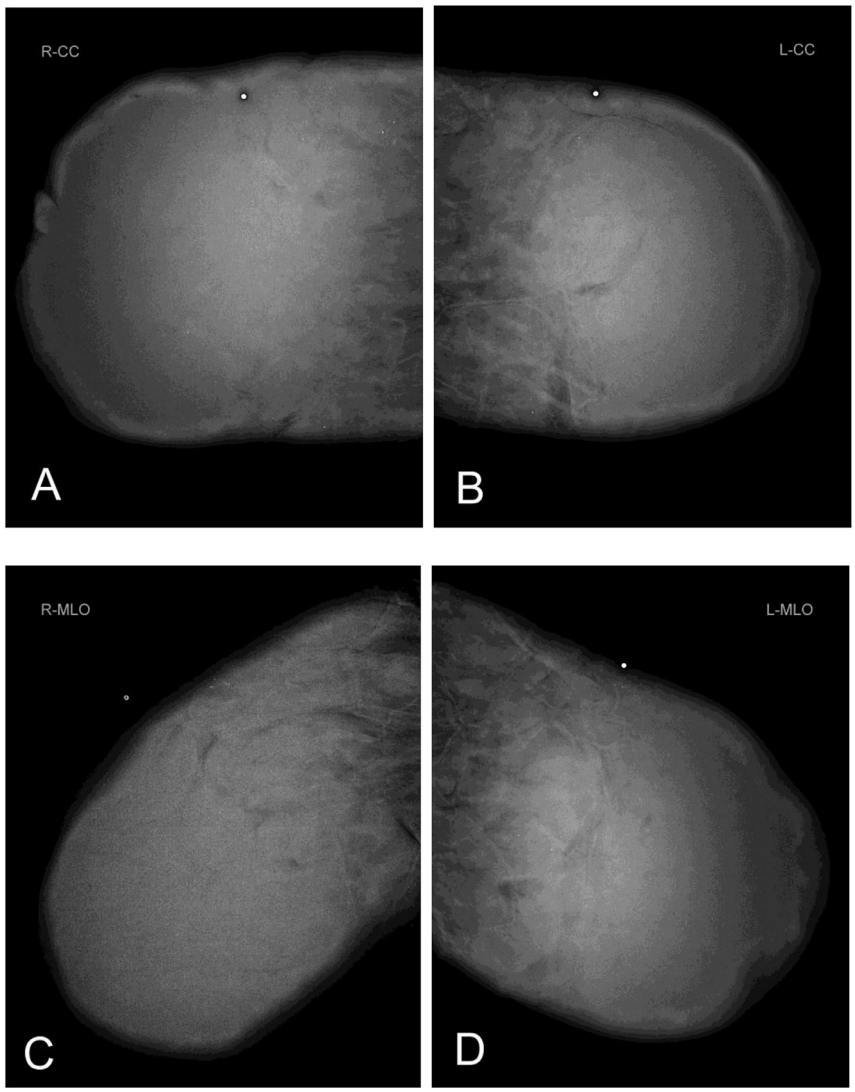

Fig. 1 – Bilateral craniocaudal view (CC) (A and B) and mediolateral oblique view (MLO) (C and D) mammographic images. Mammography shows significantly enlarged bilateral breasts with increased breast density and without a discrete mass. Global asymmetry of the bilateral breasts as well as skin thickening are shown.

with a background of extremely dense glandular parenchyma (Fig. 1). Because the abnormality was extremely large and affected most of the breast, the border of the lesion was poorly defined and covered by the adjacent parenchyma. Breast ultrasonography (US) performed using a linear transducer with a frequency of 8-15 MHz detected a heterogenous, hypoechoic, and circumscribed mass measuring approximately 16 cm at the largest diameter in each breast. Color Doppler US detected blood flow within the mass (Fig. 2). The mass was categorized as Breast Imaging Reporting And Data System (BI-RADS) 4a, and 14-gauge core needle biopsies (5×) of the right breast were performed, leading to a diagnosis of fibrocystic disease (FCD). Differential diagnoses based on imaging studies and clinical presentation included PASH, low-grade angiosarcoma, and phyllodes tumor. A bilateral mastectomy was performed because of the extent of the lesions and pain symptoms. Tumor weight was 3700 g on the right and 3300 g on the left. Macroscopic examination of mastectomy specimens showed diffusely enlarged breasts with fibrotic changes and multifocal myxoid and/or mucinous lesions bilaterally (Fig. 3). Microscopic examination of surgical specimens led to the diagnosis of PASH. Immunohistochemical studies revealed diffuse positivity for CD34, and focal positivity for smooth muscle actin and desmin, supporting the diagnosis of PASH. The postoperative period was uneventful, and the patient has been followed-up for 1 year postsurgically.

Discussion

PASH is a benign breast lesion characterized histologically by a complex network of slit-like spaces lined by endothelial-like spindle cells against a background of stromal hyperplasia simulating blood vessels [4]. The term “pseudoangiomatous” in PASH refers to the histologic appearance of the slit-like spaces,