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Case Report

An Uncommon Cause of a Breast Mass in a Child: Periductal Stromal Hyperplasia

Çocukta Nadir Meme Kitlesi Nedeni: Periduktal Stromal Hiperplazi

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ABSTRACT

Periductal stromal hyperplasia (PDSH) is a biphasic breast tumor characterized by spindle stromal cell proliferation. Fewer than 15 adult cases have been reported in the literature. A 14-year-old girl presented with swelling in axillary tail of the right breast persisting for 3 months. Ultrasonography was compatible with accessory breast tissue in both axillary regions. Macroscopic examination revealed 3x2x1 cm, non-encapsulated nodular lipomatous mass. Cut surface showed thin fibrotic bands through the adipose tissue. The major histopathological feature was spindle cell stromal cell proliferation concentrated around the ducts. There is no local recurrence during postoperative fourteen months. PDSH is a rare lesion frequently observed in older women. It has no specific clinical and radiological features. Definitive diagnosis can be made by histopathological examination of excision materials. The PDSHs do not show phyllodes tumor features. The spindle cells concentrating around ductal areas don't show marked atypia, demonstrate fewer mitosis and low proliferation index. Present case report is important in terms of unexpected age group and localization. PDSH should be kept in mind as a differential diagnosis of accesory breast tissue in children with axillary lump.

Keywords: Breast, periductal stromal hyperplasia, periductal stromal tumor, childhood

ÖZ

Periduktal stromal hiperplazi (PDSH) iğsi stromal hücre proliferasyonu ile karakterize bifazik meme tümörleridir. Literatürde 15'ten az erişkin olgu bildirilmiştir. On dört yaşında kız olgu, 3 aydır sağ aksiller bölgede ele gelen şişlik nedeniyle başvurmuştur. Ultrasonografide her iki aksiller bölgede aksesuar meme dokusu ile uyumlu görünüm izlenmiştir. Sağ aksiller eksizyon materyalinin makroskopik incelemesinde 3x2x1 cm boyutlarda, kapsülsüz nodüler lipomatö görünümde kitle izlenmiştir. Kesit yüzünün yağ dokudan zengin ve ince fibrotik bantlar içerdiği dikkati çekmiştir. Histopatolojik değerlendirmede duktusların çevresinde yoğunlaşan iğsi hücreli stromal hücre proliferasyonu izlenmiştir. Olgumuzun postoperatif 14. ayında lokal nüks izlenmemiştir. PDSH sıklıkla ileri yaş kadınlarda gözlenen nadir lezyonlardır. Spesifik klinik ve radyolojik özellikleri yoktur. Genellikle eksizyon materyallerinde kesin tanı histopatolojik olarak verilebilmektedir. PDSH'ler filloid özellikler göstermezler. Periduktal alanlarda yoğunlaşan, düşük mitoz ve proliferasyon indeksi gösteren, belirgin atipi bulunmayan iğsi hücreli stromal proliferasyon izlenmektedir. Olgumuz beklenmeyen yaş grubu ve lokalizasyon açısından oldukça önemlidir. Aksiller kitle ile başvuran çocuklarda aksesuar meme dokusunun ayırıcı tanısı olarak PDSH akılda tutulmalıdır.

Anahtar kelimeler: Meme, periduktal stromal hiperplazi, periduktal stromal tümör, çocukluk çağı

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INTRODUCTION

Periductal stromal hyperplasia (PDSH) is a biphasic tumor of the breast showing spinal cell stromal cell proliferation without phyllodes tumor features as leaf-like patern [1]. PDSH is an extremely rare fibroepithelial lesion of the breast with recurrence potential [1-3]. Less than 15 cases have been reported in the literature without any pediatric case. Here we present a case of PDSH detected in a 14-year-old girl by considering literature data.

CASE REPORT

A 14-year-old girl presented with swelling in the axillary tail of the right breast persisting for 3 months. Slight swelling was palpated in the left axillary region. Ultrasonographic examination revealed 25x10 mm sized mass consistent with accessory breast tissue. Excised

nodular mass weighed 5 grams with dimensions of 3x2x1 cm. It was non-encapsulated. Its cross-sectional surface showed adipose tissue with thin fibrotic bands. Histopathological examination demonstrated breast ducts with open lumens and spindle stromal cell proliferation especially concentrated around the ducts (Figure 1). There was no significant cytological atypia in stromal cells, 2 mitoses were observed in 10 high power fields. Stromal proliferation extended into the surrounding adipose tissue and surgical excision margins were positive (Figure 2). In focal areas gynecomastoid type ductal epithelial proliferation was observed (Figure 3). Stromal cells were diffusely positive with CD34 (Figure 4), and negative with estrogen (ER) receptor, and progesteron (PR) receptor by immunohistochemistry. Its Ki67 proliferation index was 3%. Breast lobule, acini, leaf-like formation did not exist through the lesion. According to morphological findings, our diagnosis was PDSH. Reexcision and medical therapy was not performed. There is no local recurrence during postoperative fourteen months. Informed consent was obtained from patient who participated in this case.

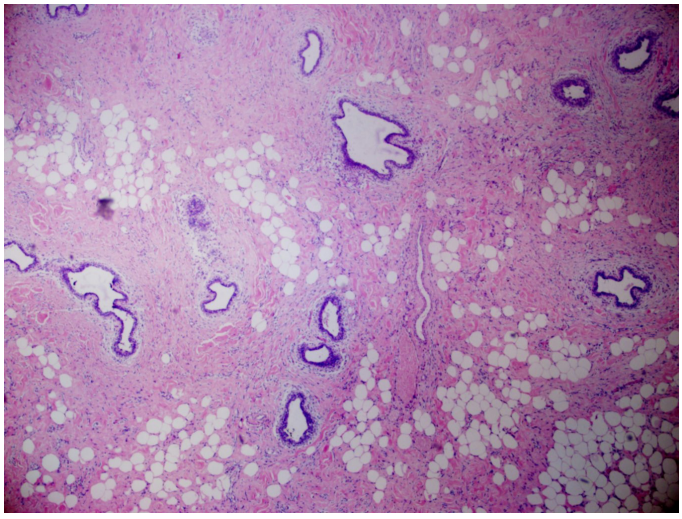

Figure 1. Spindle stromal cell proliferation especially concentrated periductal areas (H&E, x100)

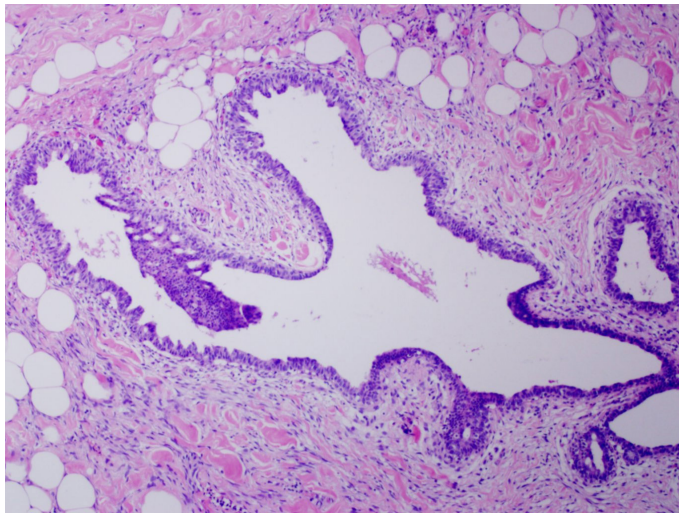

Figure 3. Gynecomastoid type ductal epithelial hyperplasia accompanying PDSH (H&E, x200)

PDSH: Periductal stromal hyperplasia

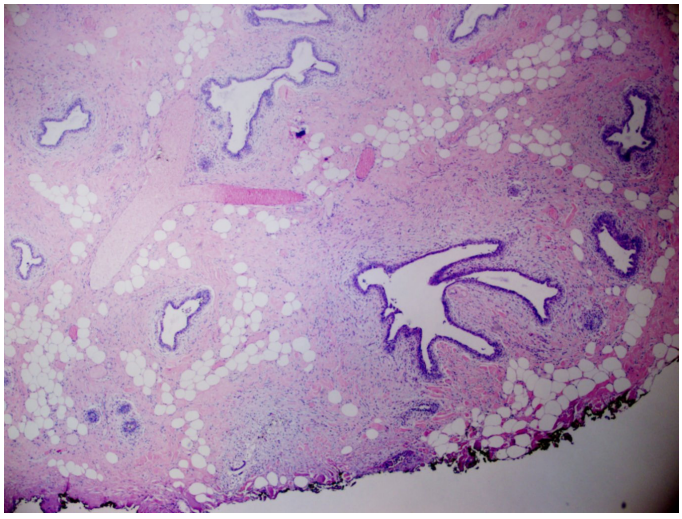

Figure 2. Surgical excision margins were positive (H&E, x100)

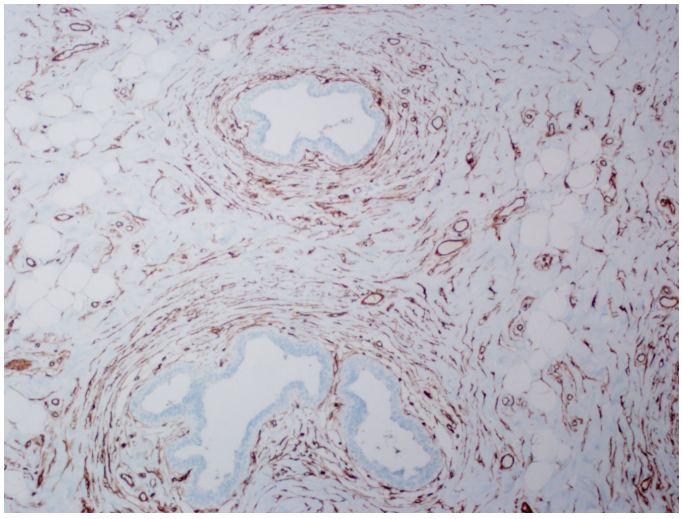

Figure 4. CD34 positivity in stromal cells (DAB, x200)

DISCUSSION

Biphasic tumors are frequent lesions of the breast. Tumors consisting epithelial and stromal proliferation can be either benign or malignant [1]. Among these tumors, fibroadenoma and phyllodes tumors are well-defined entities. PDSHs are rare breast lesions without specific clinical and radiological findings [1]. They show intermediate behaviour. Whether this tumor belongs to the phyllodes tumor spectrum or a separate entity is controversial [1]. Periductal stromal tumors (PDST), initially thought to be a variant of phyllodes tumors, are now recognized as a separate entity typically found in perimenopausal and postmenopausal women[1,4]. According to the criteria of the Armed Forces Institute of Pathology, PDST are divided into two groups as PDSH and periductal stromal sarcoma (PDSS) [1]. PDST have been described as rarely seen lesions behavioring like a low- grade sarcoma. There is no specific macroscopic specific finding of PDST [5,6]. Histologically, diagnostic criteria of PDSH are as follows: a) nodular mild stromal hyperplasia that enlarges by forming cuffs around normal or altered ducts b) lack of atypia or minimal atypia c) maximum 0-2 stromal mitoses per 10 HPF [1,6];

PDSS are defined as follows [6]: a) open tubules without phyllodes morphology and stromal spindle cell proliferation and atypia with variable cellularity around the ducts, b) one or more than one multiple nodules separated by adipose tissue, c) stromal mitotic activity (3/10 HPF) 4) stromal infiltration in surrounding breast tissue [1,6].

Immunohistochemically stromal cells stain diffusely positive with CD34, vimentin and negative with ER, PR [4].

In the literature 11 cases that met PDSH criteria have been shown [1]. The largest PDST case serial was reported by Burga and Tavassoli [6]. In this report, 20 of the cases were classified as PDSS and 7 of them as PDSH [6]. PDSH cases aged between 24-58 years. Histopathologically nodules of bland spindle cells surrounding the duct and tubules were examined. In some of them stromal edema and rare mitoses were observed (1-2/10 HPF). Atypia was not detected in spindle cell proliferation. They observed atypical features in two of the seven cases.

Burga and Tavassoli [6] defined PDSS and PDSH as PDST in peri- and postmenopausal women.

Askan et al. [3] presented a 50-year-old woman diagnosed as fibroadenoma based on tru-cut biopsy results, and excisional biopsy was performed during one year follow-up due to disease progression. Histopathological examination showed PDSH consisting of spindle cells with minimal atypia.

In the study of Coyne [2], a 52-year-old woman presented with a breast mass. This multinodular lesion had indistinct contours. In microscopic examination, they observed multinodular, mildly hypercellular, spindle cell proliferation surrounding individual or clustered ducts with one mitosis. They also observed a localized florid pseudoangiomatous stromal hyperplasia (PASH) directly adjacent to the periductal stromal tumor [2]. They emphasized that if PASH is a prominent feature in adjacent breast tissue, sampling with tru-cut biopsy may lead to erroneous diagnosis [2]. Fard and Zhang [1] presented a 57-year-old female patient with a history of stage IIB triple- negative right breast cancer treated with segmental mastectomy, adjuvant chemotherapy and radiotherapy. Ultrosonographic examination revealed a newly identified breast lesion [1]. Microscopic examination of the lesion revealed nodular periductal spindle cell proliferation that did not show phyllodes tumor features [1]. PDSTs are of mesenchymal origin [4]. Non-phyllodes sarcomas of the breast are included in the differential diagnosis of PDSH. These non-phyllodes sarcomatous breast tumors are very rare [7]. PDSSs generally don't form a mass lesion with regular smooth contours. It does not push or disrupt epithelium, like fibroadenoma and phyllodes tumor [8].

PDSTs, including PDSH, should be removed with wide excision, providing intact surgical margins [1,4]. Axillary lymph node dissection, chemotherapy or radiotherapy has no effect on the course of the disease. Their prognosis is better than the other stromal tumors of the breast [4]. Surgical border negativity is very important in recurrence and progression in PDSS [5,7].

PDSH is a rare lesion frequently observed in older women. It has no specific clinical and radiological features. Definitive diagnosis can be made by histopathological examination of excision materials. The PDSHs do not show phyllodes tumor features. The spindle cells concentrating around ductal areas don't show marked atypia, demonstrate fewer mitosis and low proliferation index. Present case report is important in terms of unexpected age group and localization. PDSH should be kept in mind as a differential diagnosis of accesory breast tissue in children with axillary lump.

Informed Consent: Informed consent was obtained from patient who participated in this case.

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Author Contributions

Surgical and Medical Practices: S.E., E.E.P., Concept: S.E., E.E.P., Design: S.E., E.E.P., Data Collection and/or Processing: S.E., E.E.P., Analysis and/or Interpretation: S.E., E.E.P., Literature Search: S.E., E.E.P., Writing: S.E., E.E.P.

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