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Pseudoangiomatous Stromal Hyperplasia of the Breast: Multimodality Imaging Findings

Memenin Psödoanjiomatöz Hiperplazisi: Görüntüleme Bulguları

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Abstract

Aim: Pseudoangiomatous stromal hyperplasia (PASH) is a rare benign mesenchymal proliferative breast lesion. The literature contains limited information on the radiological results of this uncommon tumor. In this study, we aim to define the radiologic findings of PASH through our institutional experience.

Material and Method: Patients with PASH of the breast reported in the surgical database of our institution from 2020 to 2023 were retrospectively reviewed. PASH was detected in 11 female patients among the patients who underwent a total of 2172 breast tru-cut biopsies. Nine patients whose imaging studies could be recalled from the picture archiving systems (PACS) were included in the study. BI-RADS 5th edition was used to analyze and classify radiologic findings.

Results: The median age of cases was 41 (range 22–53). Our single-center incidence was found to be 0.5%. Considering the sonographic findings, all of the lesions had an oval shape. On mammography, they were defined as focal asymmetry or circumscribed masses. MRI was available in 3 cases. All 3 cases were hypointense on T1-weighted sequences and hyperintense on T2-weighted sequences. They displayed type 1 or type 2 enhancement curves in the dynamic contrast-enhanced images. No diffusion restriction was detected.

Conclusion: In this study, tumor-forming PASH were generally circumscribed, oval hypoechoic solid masses with minimal vascularity and no posterior acoustic features on ultrasound. On mammography calcification, architectural distortion or spiculation were not present in any of the cases. MRI findings were t2 hyperintensity, type 1–2 enhancement kinetics, and no diffusion restriction. In all imaging modalities, the imaging characteristics point to a benign lesion.

Keywords: Breast tumor, pseudoangiomatous stromal hyperplasia, ultrasound, mammography, MRI

Öz

Amaç: Psödoanjiyomatöz stromal hiperplazi (PASH) memenin nadir görülen benign mezenkimal proliferatif lezyonudur. Literatür, bu nadir tümörün radyolojik sonuçları hakkında çok az bilgi içermektedir. Bu çalışmada PASH'ın radyolojik bulgularını kurumsal deneyimlerimizden hareketle tanımlamayı amaçladık.

Gereç ve Yöntem: Kurumumuzun cerrahi veri tabanında 2020-2023 yılları arasında bildirilen meme PASH'li hastalar retrospektif olarak incelendi. Toplam 2172 meme tru-cut biyopsisi yapılan hastalardan 11'inde kadın hastada PASH saptandı. Görüntüleme çalışmaları resim arşivleme sistemlerinden (PACS) geri çağrılabilen dokuz hasta çalışmaya dahil edildi. BI-RADS 5. baskı, radyolojik bulguları analiz etmek ve sınıflandırmak için kullanıldı.

Bulgular: Olguların ortanca yaşı 41'di (22-53 arası). Tek merkezli insidansımız %0,5 olarak bulundu. Sonografik bulgulara bakıldığında lezyonların tamamı oval bir şekle sahipti. Mamografide fokal asimetri veya sınırlı kitleler olarak tanımlandı. 3 olguda MRG mevcuttu. 3 vakanın tümü, T1 ağırlıklı sekanslarda hipointens ve T2 ağırlıklı sekanslarda hiperintens idi. Dinamik kontrastlı görüntülerde tip 1 veya tip 2 geliştirme eğrileri gösterdiler. Difüzyon kısıtlaması saptanmadı.

Sonuç: Bu çalışmada, tümör oluşturan PASH'lar genel olarak sınırlı, minimal vaskülariteye sahip, ultrasonda posterior akustik özelliği olmayan, oval hipoekoik solid kitlelerdi. Mamografide kalsifikasyon, distorsiyon veya spikülasyon olguların hiçbirinde yoktu. MRG bulguları t2 hiperintensite, tip 1-2 kontrastlanma kinetiği ve difüzyon kısıtlaması olmamasıydı. Tüm görüntüleme modalitelerinde, görüntüleme özellikleri iyi huylu bir lezyona işaret etmekteydi.

Anahtar Kelimeler: Meme tümörü, psödoangiomatöz hiperplazi, ultrason, mamografi, MRG

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INTRODUCTION

Pseudoangiomatous stromal hyperplasia (PASH) of the breast is a benign mesenchymal proliferative disease. Vuitch et al described PASH in 1986.[1] It is hypothesized that hormonal influences contribute to its development.[2] It may present clinically as a mass or incidental microscopical finding. Rarely does PASH cause tumor. However, PASH might be discovered incidentally in up to 23% of breast biopsies. [3] Histologically, it must be distinguished from low-grade angiosarcoma and phyllodes tumors when there is a mass. It is identified by stromal cells with slit-like channels lined by myofibroblasts that resemble vascular channels on pathology specimens. As a result, PASH can be misdiagnosed as a low-grade angiosarcoma histologically. Angiosarcoma can be differentiated based on malignant cytology and positive immunohistochemical staining to endothelial markers. However, no association of PASH with malignancy has been proven.

Although PASH typically manifests as a localized lesion, diffuse and multifocal involvement have also been reported. [4,5] Clinically, it is a firm, palpable, painless breast mass that may have a diameter of up to 15 cm. It may be misdiagnosed as a fibroadenoma or phyllodes tumor based on clinical, mammographic, and ultrasonographic features.[5-7] The recommended course of treatment for tumor-forming PASH is local surgical excision with sufficient margins when it is growing, or exhibits suspicious imaging findings. The likelihood of recurrence is low, and the prognosis is favorable.

In this study, we aim to define the radiologic findings of PASH through our institutional experience.

MATERIAL AND METHOD

In this retrospective descriptive study, review of the pathological database of our institutions from 2020 to 2023 revealed 11 cases of PASH of the breast among a total of 2172 tru-cut breast biopsies. All of the patients were female. Nine of these cases had radiological studies available in the picture archiving systems (PACS) systems. These 9 cases make up the study population of this study.

Age, gender, the patient's current symptoms, and the results of the tru-cut biopsy and postoperative pathology reports were noted. All of the available radiological studies were retrieved from PACS. Two breast radiologists (5 and 10 years of experience) reevaluated the images in agreement. The Breast Imaging Reporting and Data System (BI-RADS) 5th edition lexicon was used to categorize imaging findings.[8] The morphological characteristics listed below were examined: shape, margin, density, and associated calcifications on mammography; shape, margin, orientation, echo pattern, posterior acoustic features, vascularity on ultrasonography; and shape, margin, internal enhancement patterns, T2 signal, diffusion characteristics, and kinetic features on magnetic resonance imaging (MRI).

Descriptive statistics (mean, standard deviation, minimum, median, maximum) were used to define continuous variables.

The study was carried out with the permission of Ümraniye Training and Research Hospital Clinical Research Ethics Committee (Date: 21.03.2023, Decision No: B.10.1.TKH.4.34.H.JP.0.01/85), and patient consent was waived.

RESULTS

Among a total of 2172 trucut breast biopsies 11 cases of PASH result in an incidence of 0.5%. All patients were women with a median age of 41 (range 22-53), and all were premenopausal except one. The presenting symptom was and palpable mass in 8 patients, one of which was painful. One lesion was detected on screening. None of the patients had a breast cancer history. Two patients had second-degree family history of breast cancer.

The maximum diameter of the lesions ranged between 14-60 mm (mean 36.3 mm) on ultrasound imaging. Seven of these patients had previous medical records which demonstrated 6-34% enlargement in the largest diameter in 6 of the masses. None of the patients had multifocal lesions. Five patients were treated with simple excision and one patient had a mastectomy. On postoperative pathology reports, five patients had an accompanying fibroadenoma, while one patient had isolated PASH. None of the patients had accompanying ductal carcinoma in situ (DCIS) or invasive cancer on imaging findings or pathology.

Imaging Findings

US images of all 9 lesions were available. Five patients had mammography, and 3 patients underwent dynamic contrast-enhanced MRI. There was no multifocality. A summary of the imaging findings is demonstrated in **Table 1**.

All of the masses could be visualized in the US (**Figures 1,2,3**). The orientation of all the lesions was parallel. The shape was oval in all of them. The margins were circumscribed in 8 and microlobulated in one. There were no posterior acoustic features in any of the lesions. The echogenicity was hypoechoic in all lesions. In one lesion, microcystic changes were present within the mass. On color Doppler imaging, one lesion was avascular, while 8 lesions displayed minimal vascularity. Based on US features, 2 of the masses were categorized as BI-RADS 4B, 6 as BI-RADS 4A, and 1 as BI-RADS 3.

Mammography was available in 5 patients (**Figures 1,2**). Three patients had type C, one had type B and one patient had type D breast parenchymal density. One lesion was not seen due to dense breast parenchyma (type D). The shape was oval in the other 4 lesions. The density of one mass was hyperdense while others were isodense. Margins were circumscribed in 1 and indistinct in 3. None of the cases had spiculated margins. Calcification was not present in any of the masses.

Dynamic contrast-enhanced breast MRI was available in 3 cases (**Figures 1,3**). The shape was oval in all cases. The

margins were circumscribed in one, and indistinct in 2. All masses displayed heterogeneous internal enhancement. None displayed rim enhancement. On T2 weighted images, all masses were isohyperintense or hyperintense. On kinetic analysis, enhancement pattern was persistent or plateau-type, and none demonstrated washout kinetics. Diffusion-weighted imaging (DWI) demonstrated no restricted diffusion.

Table 1. Summary of findings

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8	Case 9
MAMMOGRAPHY									
Breast density type	C	C	C					B	D
Shape	oval	oval	oval					oval	-
Margin	circumscribed	indistinct	indistinct					indistinct	-
density	dense	dense	isodense					isodense	isodense
calcification	none	none	none					none	none
ULTRASOUND									
orientation	parallel	parallel	parallel	parallel	parallel	parallel	parallel	parallel	parallel
shape	oval	oval	oval	oval	oval	oval	oval	oval	oval
margin	circumscribed	circumscribed	circumscribed	circumscribed	circumscribed	circumscribed	microlobulated	circumscribed	circumscribed
Echo pattern	Heterogenous Cystic changes	hypoechoic	hypoechoic	hypoechoic	hypoechoic	hypoechoic	hypoechoic	hypoechoic	hypoechoic
Posterior acoustic features	none	none	none	none	none	none	none	none	none
vascularity	minimal	minimal	minimal	minimal	minimal	avascular	minimal	minimal	minimal
BIRADS category	4B	4A	4A	4A	4A	4A	4B	3	4A
MRI									
shape		oval			oval				oval
Internal enhancement		heterogenous			heterogenous				heterogenous
Kinetics		persistent			persistent				plateau
T2 signal		isohyperintense			hyperintense				hyperintense
DWI		No restriction			No restriction				No restriction
ADC value		2077 mm2/s			1777 mm2/s				1500 mm2/s

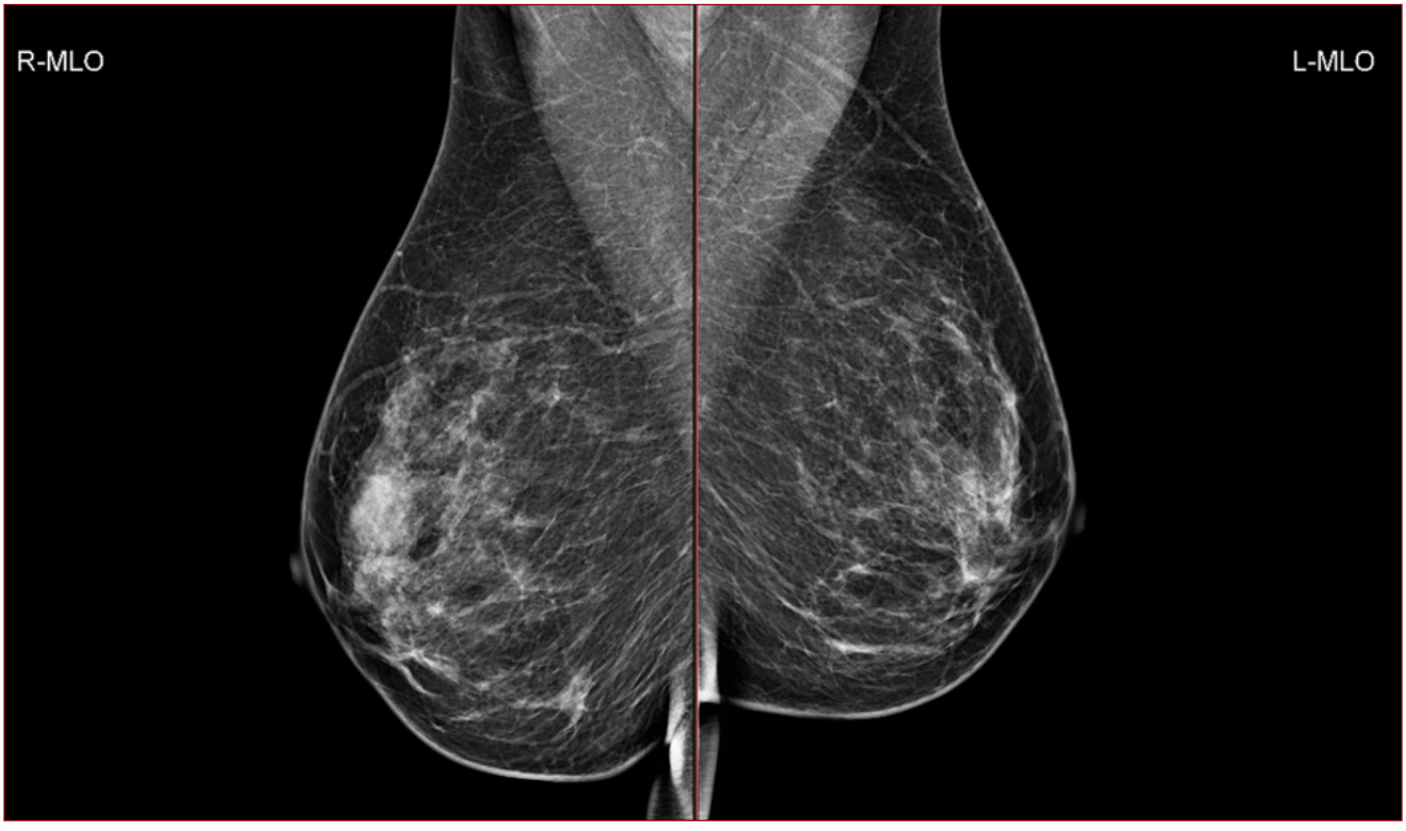

Figure 1a: 42 y/o female patient: Screening mammograms depict an oval mass with indistict margins in the upper outer quadrant of the right breast.

Figure 1b: US image shows a hypoechoic solid mass with circumscribed margins.

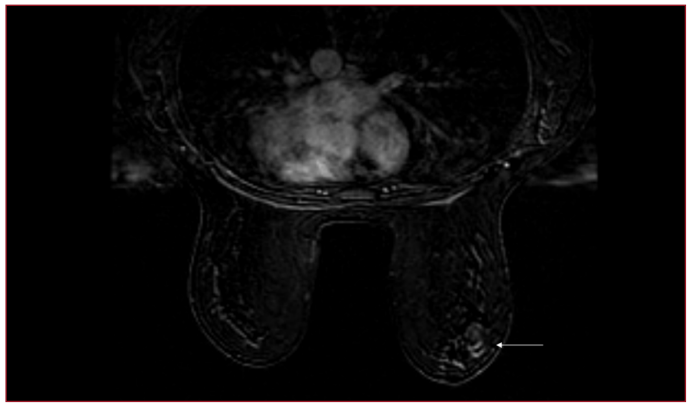

Figure 1g: On contrast-enhanced MR images, the lesion (arrow) shows slight and persistent enhancement.

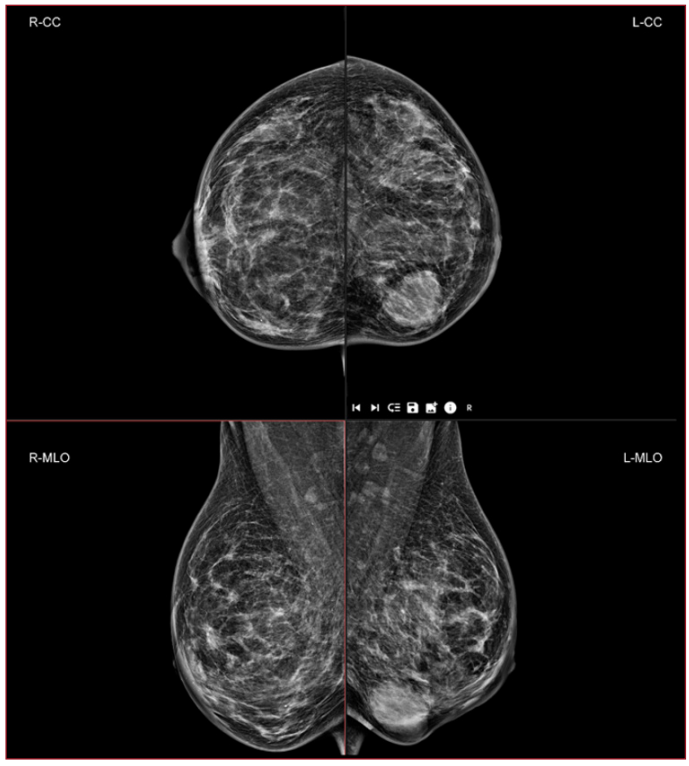

Figure 2a: 41 y/o patient who presented with a palpable mass and tenderness in her left breast. Mammograms of the left breast shows a dense oval mass with circumscribed margins.

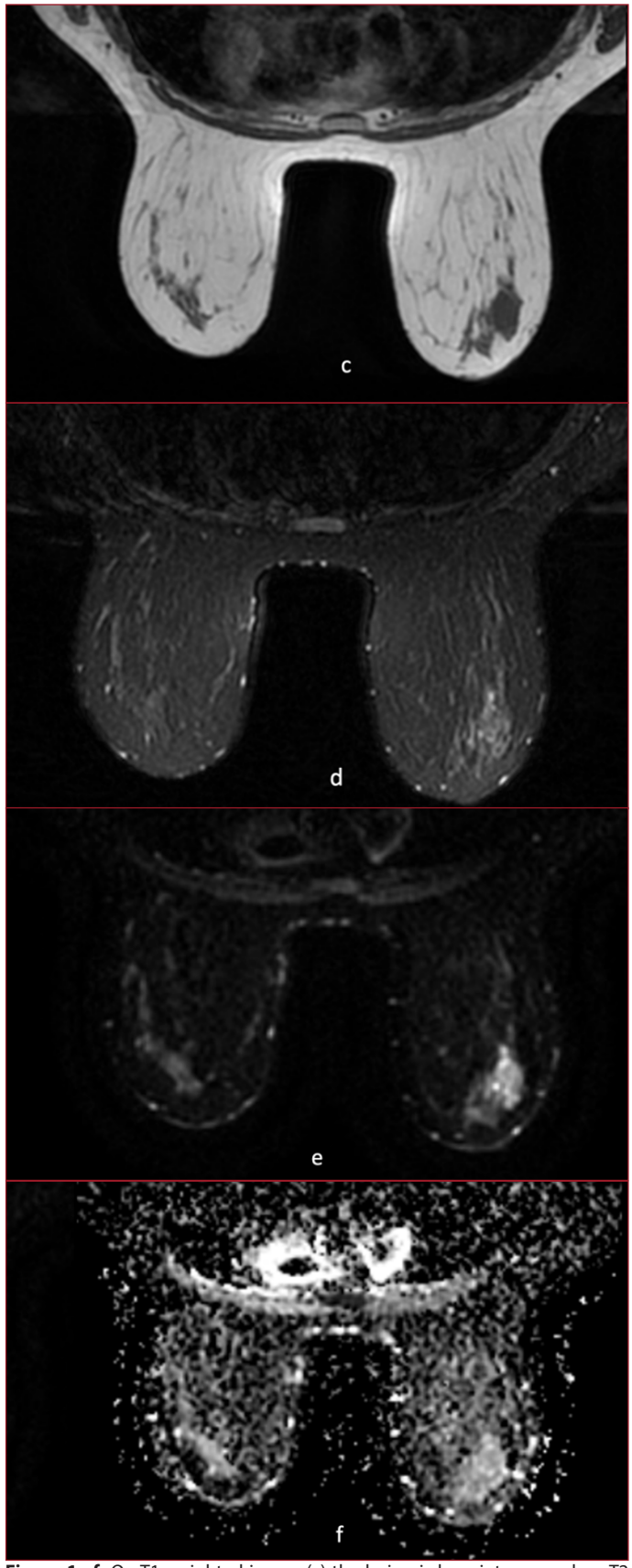

Figure 1c-f: On T1 weighted image (c) the lesion is hypointense and on T2 weighted image (d), the lesion is isohyperintense. On diffusion weighted image (e) slight hyperintensity is due to t2 effect and ADC (f) map indicate that there is no diffusion restriction.

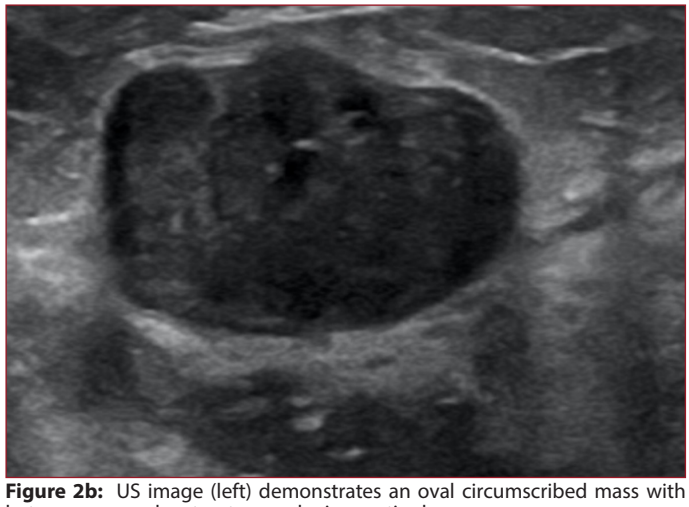

Figure 2b: US image (left) demonstrates an oval circumscribed mass with heterogenous echo structure and microcystic changes

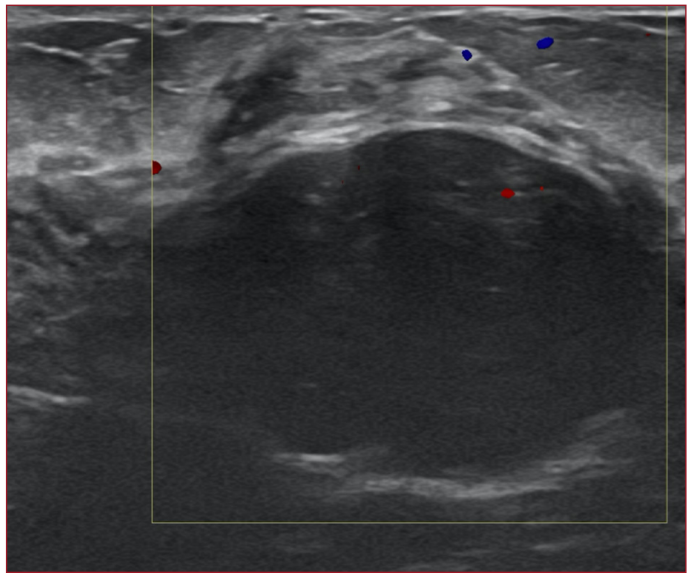

Figure 3a: 38y/o patient who presented with a palpable circumscribed oval solid mass with minimal vascularity on color doppler US image.

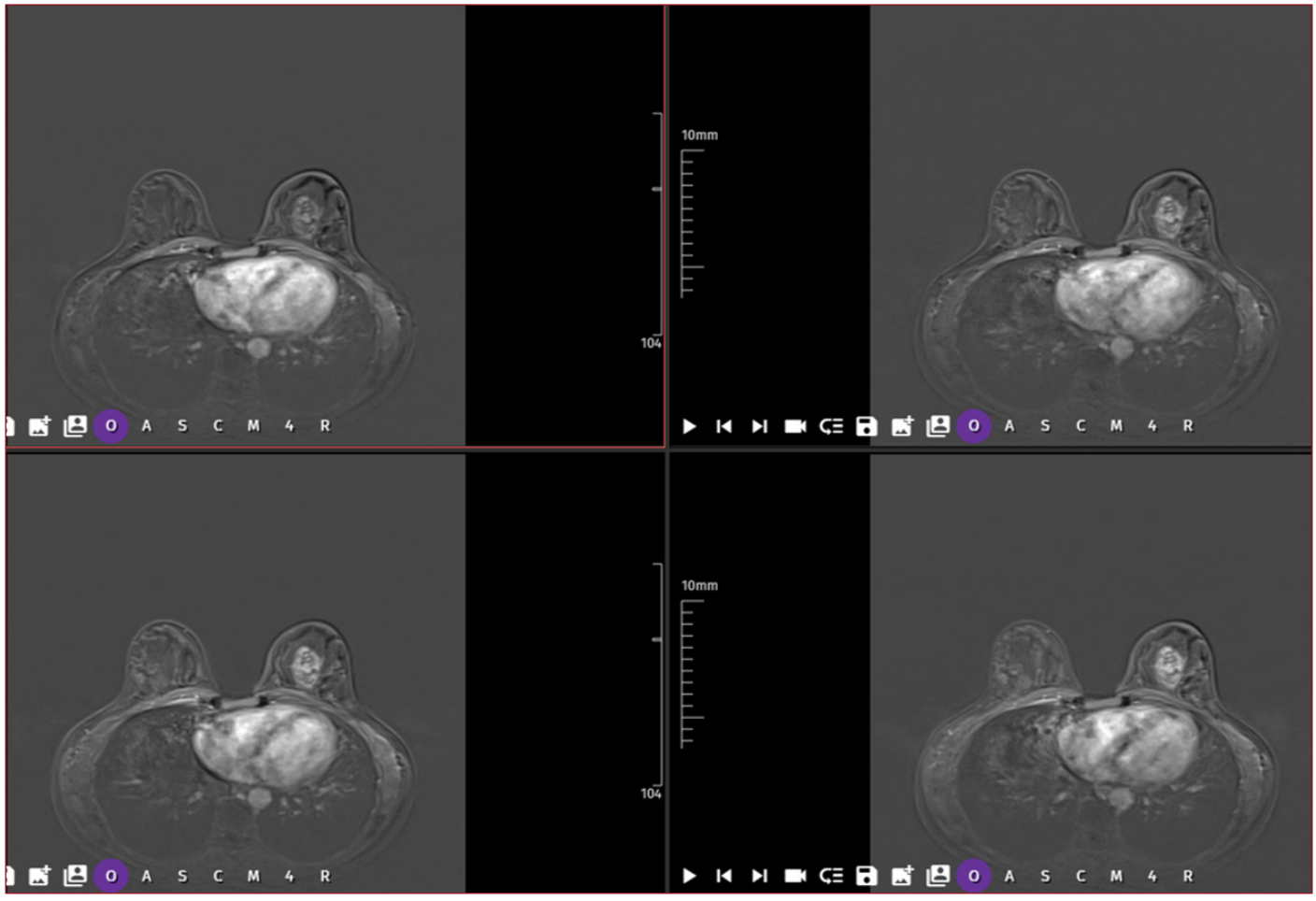

Figure 3b: On contrast-enhanced MR images, the lesion shows heterogenous and persistent enhancement

DISCUSSION

In this retrospective study, we presented the radiological findings of 9 patients with PASH lesions, collected from the databases of our institution between 2020-2023. All patients underwent ultrasound, 5 of them underwent mammography, and 3 of them underwent MRI. Color Doppler imaging was available in all patients. We have reviewed all radiological examinations in detail to determine the most common imaging features.

The radiological characteristics of PASH are not adequately described in the literature. The radiological findings are only briefly discussed in a few studies, the majority of which are case reports. According to the literature, the most common signs of PASH on mammography are non-calcified, round or oval, circumscribed or partially circumscribed masses or an uneven density.[9,10] As their research comprised cases in which PASH was incidentally discovered on histology, Hargaden et al. observed that 69% of patients with PASH did not display any mammographic abnormalities.[5] Only 10 of the 169 cases reported by Hargaden et al. had architectural distortions or calcifications on mammography. In this study, the most common mammographic features were an oval shape, and circumscribed or indistinct margins. Spiculations, microcalcification, or architectural distortion were not present in any of the cases. These findings are consistent with the literature.

On ultrasound, all lesions presented as solid masses in this study. Jones et al. noted that the most typical ultrasonography appearance of PASH was an oval, hypoechoic mass with circumscribed margins.[7] However, there are also suspicious characteristics defined such heterogenous echotexture, high echogenicity, and ill-defined borders.[2] In our study, all the lesions were circumscribed hypoechoic oval masses with circumscribed margins, except one, which was microbulated. Posterior acoustic features were not present in any of the cases in our study. Doppler imaging revealed minimal vascularity in almost all cases. We have categorized the findings as BI-RADS 3-4A in 77% (7/9) of the ultrasound cases. Two lesions, one with microlobulation and the other with heterogeneous echo structure were categorized as BI-RADS 4B. When all imaging findings were taken into account, none of the lesions were considered BI-RADS 4C or 5.

Few studies have defined the appearance of PASH on MRI.[11-13] In this study, T2 hyperintensity and the lack of diffusion restriction are remarkable features on MRI. All three lesions demonstrated type 1 or type 2 contrast enhancement kinetics and washout kinetics were not seen in any of them. Findings on MRI point out the benign nature of the lesion. Alicassi et al. reported a case with multiple masses showing low signal in T1 sequences and high signal in T2 sequences, early homogenous and intense contrast enhancement with all three types of enhancing curves that are more common for persistent kinetics.[13] According to Nia et al.'s analysis of 69 cases, PASH shows in various appereaances on MRI but most frequently as clumped non-mass enhancement with persistent kinetics.[11] Their study group included MRI-guided biopsies, indicating that the lesions were only visible on MRI. Our study differs in way that the lesions are masses which all are also visible on ultrasound.

PASH is primarily present in pre- or perimenopausal women and is thought to be hormone-related.[14,15] In this study, age distribution is in line with the literature data that almost all of them were premenopausal. None of the cases in this study had a breast cancer history or coincidental breast cancer. None of them had first-degree breast cancer family history. To date, PASH associated with malignancy has been rarely reported.[16,17]

Gradual enlargement of PASH masses has been mentioned in mammography.[1,3] We also detected 6-34% enlargement in one year in 6 of the patients who had previous medical records. Although the lesions were benign-appearing, gradual enlargement conveyed a histologic verification.

PASH occurs as a major histological finding in ~6% of surgical breast biopsies[18] and microscopic non-tumor forming PASH is an incidental finding in up to 23% breast biopsies.[3] Cases in our study are the tumor-forming type of PASH. Nodular PASH is a rare entity with an incidence of 0.4% in breast biopsies.[9] Our single-center incidence is 0.5% which is compatible with this literature data. The exact incidence is difficult to estimate as it is related to the awareness of this rare lesion by pathologists.

Limitations

The small number of cases and retrospective design are the main limitations of this study. Although there were more cases with a diagnosis of PASH, not all of them had access to their radiological images. Some cases did not undergo mammography or MRI. Retrospective analysis of radiological data, particularly US findings, can be deceptive.

CONCLUSION

PASH is a benign breast tumor, diagnosed more commonly in premenopausal women. The imaging features suggest a benign lesion in all imaging modalities. In this study tumor forming PASH were generally circumscribed, oval hypoechoic solid masses with minimal vascularity and with no posterior acoustic features on ultrasound. On mammography calcification, architectural distortion or spiculation were not present in any of the cases. MRI findings were t2 hyperintensity, type 1-2 enhancement kinetics, and no diffusion restriction. Although nonspecific, these imaging features of PASH suggest a benign process. Despite the fact that imaging results properly identified the benign nature of the lesions, biopsy verification may be necessary due to the lesions' size and gradual enlargement over time.

ETHICAL DECLARATIONS

Ethics Committee Approval: The study was carried out with the permission of Ümraniye Training and Research Hospital Clinical Research Ethics Committee (Date: 21.03.2023, Decision No: B.10.1.TKH.4.34.H.JP.0.01/85).

Informed Consent: Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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