





# Juvenile papillomatosis: A case report

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## ABSTRACT

Juvenile papillomatosis (JP) is a very rare benign proliferative breast disease, especially in young women under 30 years of age. Its etiology is not clear yet. Although some patients have breast cancer in their family history, up to 10% breast cancer can develop in the follow-up of JP patients. In this pathology, which is diagnosed with biopsy, history, clinical and radiological findings help in diagnosis. In this case report, a 37-year-old patient diagnosed with JP will be discussed in the light of the literature.

**Keywords:** Juvenil papillomatosis, proliferative lesion, malignancy

## INTRODUCTION

Juvenile papillomatosis (JP) is a rare benign mass-like lesion which is pre-dominantly seen in young women under 30 years old. It can confuse with fibroadenoma which is multinodular mass on breast examination, but JP lesions are generally are larger than fibroadenomas. Radiological and histopatological examinations play key role in certain diagnosis.

## CASE REPORT

A 37-year-old female patient with no known cancer history and additional disease in her family was referred to the radiology clinic after she applied to the general surgery outpatient clinic with a spontaneous discharge from the left. As a result of this mammography examination, asymmetric fibroglandular tissue density in the right midline posterior of the right breast was detected and ductal dilatations with secretion content were detected in bilateral retroareolar areas in both breasts that did not exceed one cm in both breasts (Figure 1A-1B, 2A-C). Since the nipple discharge of the patient could not be explained with defined mammographic and sonographic findings, a dynamic contrast-enhanced MR examination was performed in order to solving problem. In breast MRI examination; In the right breast at 12 o'clock, a lesion with a posterior location of 18 x 15 mm, which is hardly differentiated from the breast parenchyma, which contains millimetric cysts, was observed in the post-contrast series, with peripheral-weighted heterogeneous contrast, and in the late series, contrast loss was observed. In addition, in the subtraction images, non-mass heterogeneous segmental enhancements were recorded in an area of approximately 5 x 4.5 x 2.5 cm, including the neighborhood of the defined lesion, in the upper right outer quadrant of the right breast. The findings were evaluated in the category suspected for malignancy (Figure 3A-3B, 4A-4B). Corre biopsy was performed by correlating the lesion with the second aspect US, and the result of pathology was reported as ductal epithelial hyperplasia, sclerosing adenosis. The pathology result of the excisional biopsy performed for the lesion due to radiological-pathological incompatibility; It has been evaluated as "juvenile papillomatosis"

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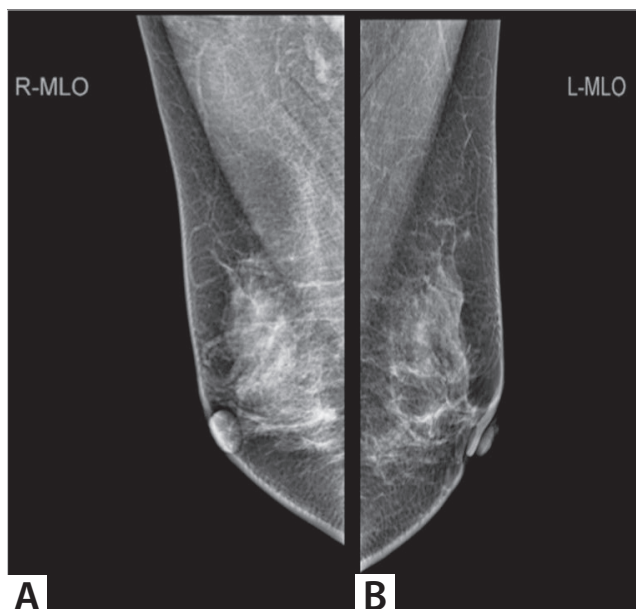
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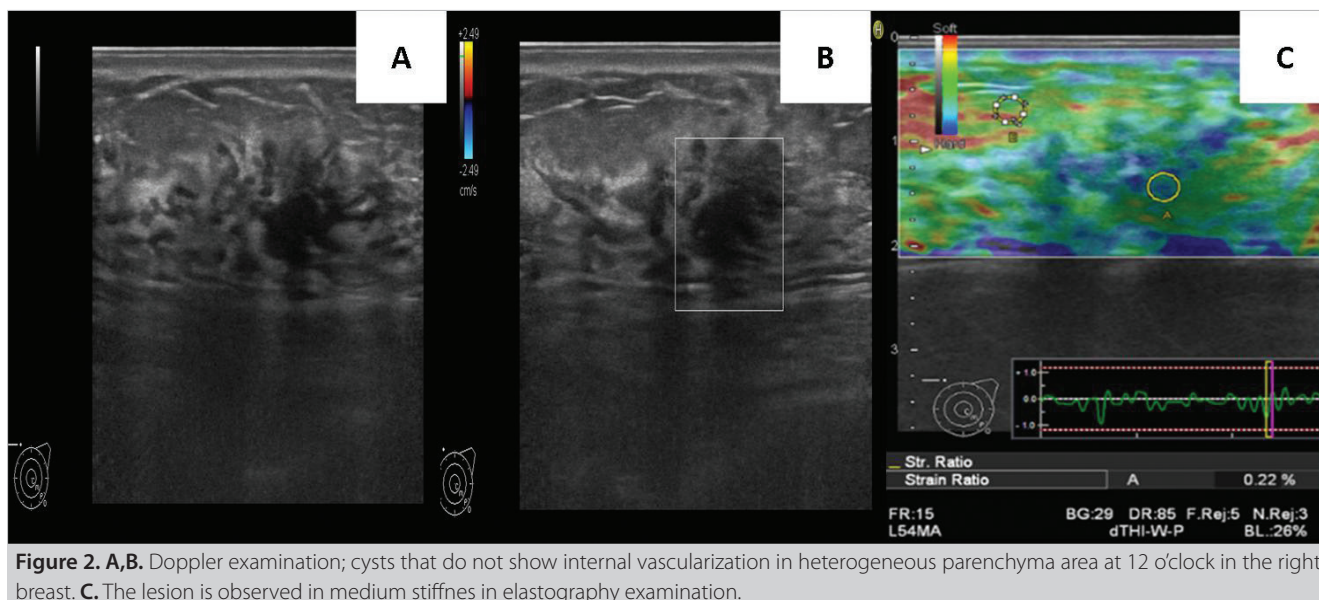
**Figure 1. A,B.** Asymmetric fibroglandular tissue density observed in the midline posterior on the right breast MLO view.

containing atypical ductal hyperplasia areas (Figure 5A-C, 6A-C). The lesion causing the left nipple discharge of the patient was also detected by breast MRI and it was understood that it was a benign intraductal papilloma located at retroareolar location.

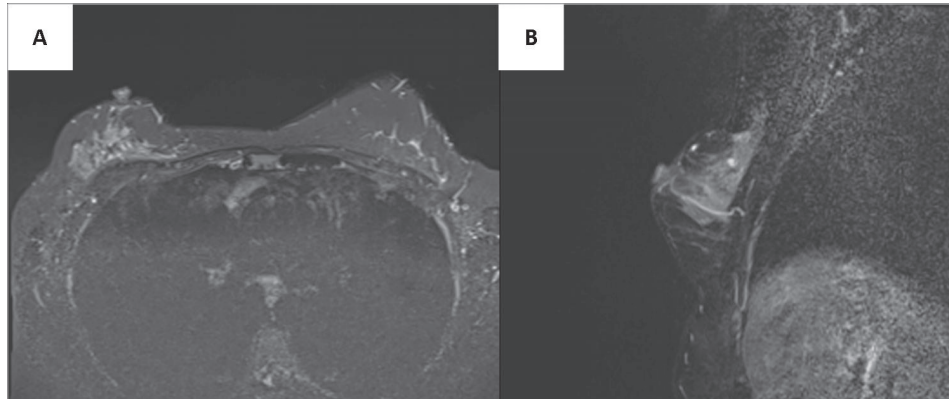
## DISCUSSION

JP is a pathology that was defined by Rosen et al. between 1947 and 1980 as a series of 37 cases. In the case series that was first described, the average age of the patients was 19 and mostly consist of Caucasian patients (1,2). Considering the general cases in the literature, it has been reported that 70% of the pre-

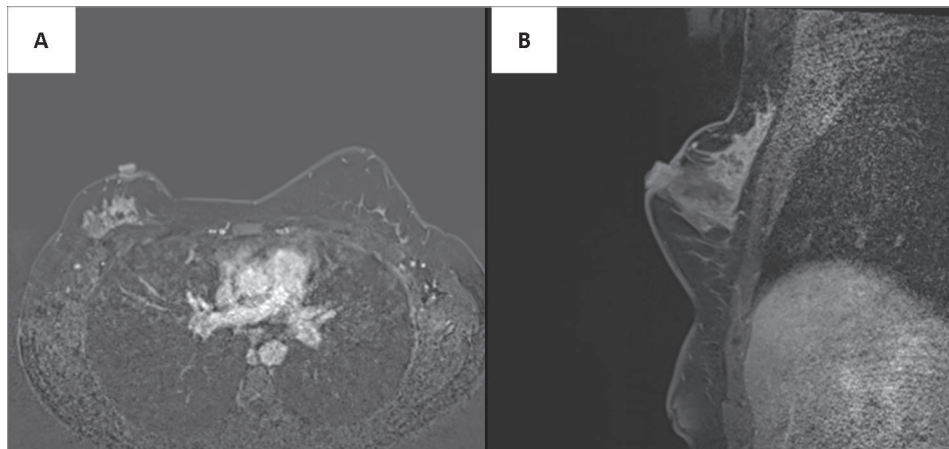
sented cases are under the age of 26, but this spectrum has expanded until the age of 40 (3,4). Although histopathological examinations may have different findings, the most common findings are accumulation with papillomatosis of ductus with or without JP epithelial atypia, papillary apocrine hyperplasia, sclerosing adenosis and stasis in ducts (5). In JP, which is also named in the literature as the Swiss cheese disease of the breast, the reason for this naming is that there are many cysts and ducts in the dense stromal areas in the microscopic images (6). JP may rarely be associated with severe atypia and focal epithelial necrosis. Patients with such histopathological characteristics generally display premalignant features and have been observed in elderly female patients (7). In the clinic, patients are usually presented with a well-circumscribed, rigid and mobile mass in the upper outer quadrant of the breast but not causing discharge from the nipple. Although the exact diagnosis of JP is made by biopsy, imaging modalities are used in the pre-operative diagnosis and follow-up of patients. In the ultrasound image, it is observed as hypoechoic mass, which is not generally well-circumscribed, and multiple small-sized weighted peripheral localized cysts (8). While the findings of the lesion are generally not observed in mammograms, rarely, asymmetric densities can be observed with pleomorphic and amorphous microcalcifications (8,9). Contrast-enhanced T1-weighted fat saturated images are useful in the evaluation of lesion contours and internal matrix. In T1-weighted images, a lobule contoured mass image is observed in the hypointense view; T2-weighted images, on the other hand, are the most helpful sequence in diagnosis since they show many small cysts (10). Since JP has active proliferative hyperplastic epithelial tissue, it shows involvement in contrast-enhanced MRI examinations. Contrast media involvement patterns are in the form of type 1 and type 2 curves, such



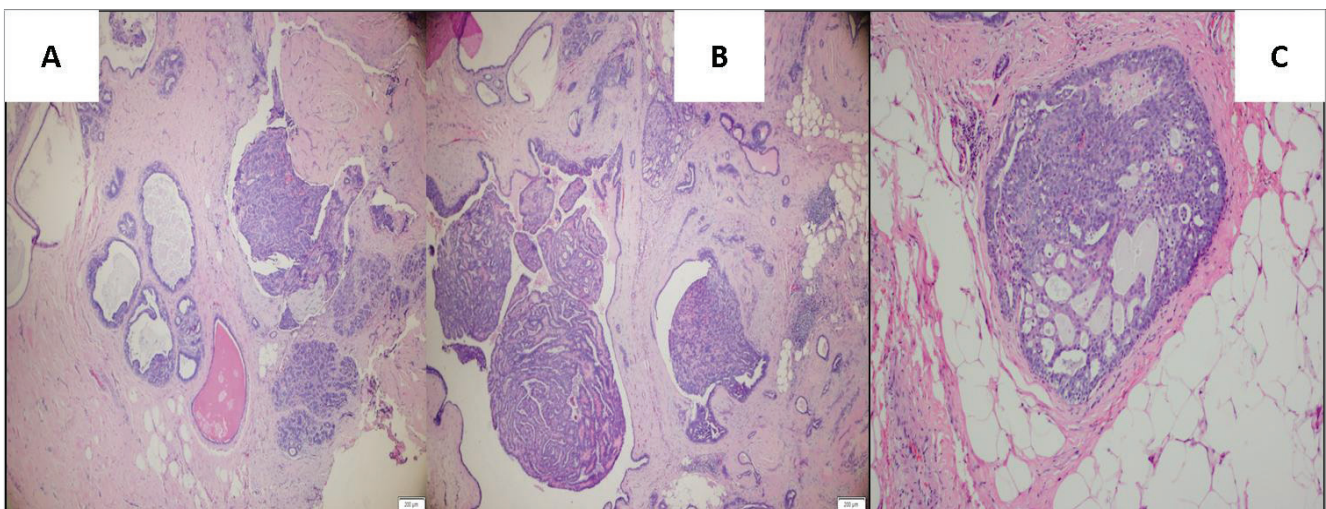
**Figure 2. A,B.** Doppler examination; cysts that do not show internal vascularization in heterogeneous parenchyma area at 12 o'clock in the right breast. **C.** The lesion is observed in medium stiffness in elastography examination.



**Figure 3.** A. T2-weighted fat saturated axial MR image B. T2-weighted fat saturated sagittal MR image - Lesion with millimeter cysts approximately 2 cm from the nipple at 12 o'clock.

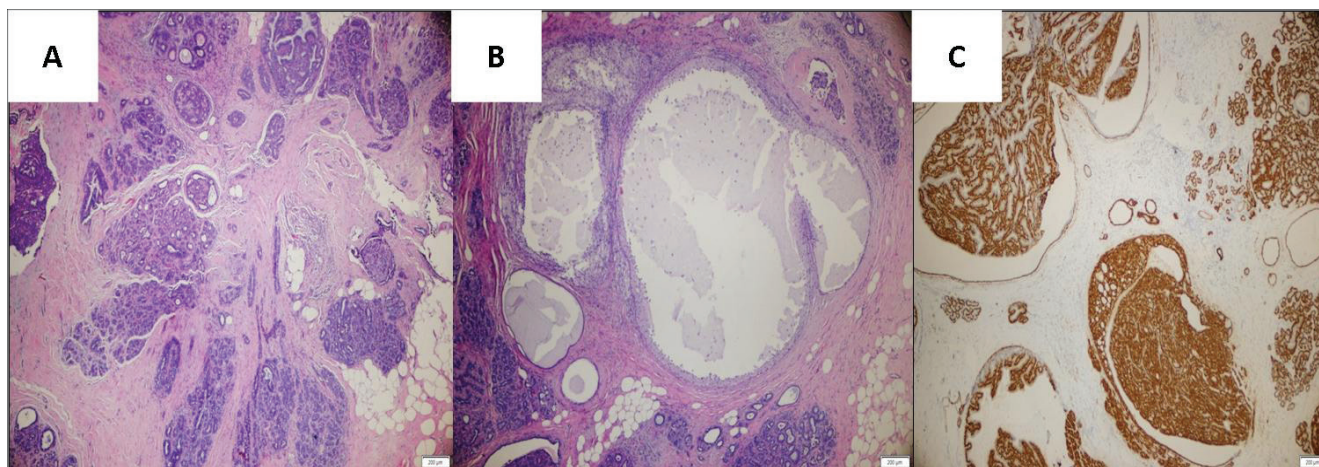


**Figure 4.** A. T1-weighted fat saturated axial MR image. B. T1-weighted post-contrast sagittal MR image - Peripheral enhancement at the lesion and adjacent non-mass segmental enhancements are observed.



**Figure 5.** A,B. Papilloma areas (HE x 40) characterized by papilla-like structures in the ductus lumens within the breast parenchyma (HE x 40). C. Atypical epithelial hyperplasia areas (HE x 40).





**Figure 6.** A. Sclerosing adenosis areas (HE x 40). B. Cystic enlarged ducts (HE x 40). C. Immunohistochemical calponin positivity around the ducts (HE x 40).

as benign diseases (11,12). Despite its benign nature, there is an increased risk of breast cancer in JP cases. In 4-15% patients, JP and breast cancer co-exist at the time of diagnosis. Although JPs are generally associated with intraductal papillomas, they can also co-exist with invasive ductal, invasive lobular and secretory cancers of the breast (13). In addition, approximately 10% of patients with JP have a risk of developing breast cancer in their follow-up (13). Since incomplete surgical excision causes recurrences, extensive surgical excision and clinical follow-up are the most common treatment options in JP cases. In patients with bilateral, multifocal and recurrent family history, the frequency of follow-up should be increased since the risk of breast cancer increases (7-14).

## CONCLUSION

JP is a benign proliferative lesion of the breast, which is frequently observed at a young age. Clinically, radiologically it can be confused with malignancy, requiring histopathological examination for a definitive. In terms of breast cancer that can accompany and occur during follow-up, extensive surgical excision, multiple sampling and clinical follow-up are recommended.

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**OLGU SUNUMU-ÖZET**

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**Juvenil papillomatozis: Olgu sunumu**

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**ÖZET**

Juvenil papillomatozis (JP) özellikle 30 yaş altında genç kadınlarda izlenen oldukça nadir görülen benign proliferatif meme hastalığıdır. Etiyolojisi henüz net olarak bilinmemektedir. Hastaların bir kısmında aile öyküsünde meme kanseri bulunmakla birlikte, JP hastaların takiplerinde %10 kadar meme kanseri gelişebilmektedir. Kesin tanısı biyopsi ile konan bu patolojide öykü, klinik ve radyolojik bulgular tanı konulmasında yardımcı olmaktadır. Bu olgu sunumunda 37 yaşında JP tanısı alan hasta literatürdeki veriler ışığında tartışılacaktır.

**Anahtar Kelimeler:** Juvenil papillomatozis, proliferatif lezyon, malignite

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