

# Juvenile Atypical Ductal Hyperplasia: A Case Report

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## Abstract

Breast masses, benign and malignant, are extremely common in women. Benign breast masses have a variety of risk factors and are usually most common in women aged 30 - 40 years. Although much research has been conducted on benign breast disease and breast cancer in adult women, there remains a paucity of data on breast masses in adolescent women. More specifically, there is very little evidence regarding atypical ductal hyperplasia (ADH) in young women and the future risks it may carry as it is known to have a 4 to 5-fold increase for breast cancer in the adult population. We present a case of an 18-year-old female with ADH involving a fibroadenoma, with the hopes of highlighting the unique concerns and questions this diagnosis may bring to the young female population. Our patient’s questions centered around the risk this diagnosis carries of future cancer and what steps should be taken to minimize that risk. Although many models and guidelines exist to answer these questions in the older female, there is no consensus about treatment and monitoring ADH in a juvenile. By reviewing this case, we emphasize the need for future studies to quantify the risk of cancer progression from ADH in young females. We also demonstrate the need for guidelines to monitor and treat these findings in our younger populations.

**Keywords:** Atypical ductal hyperplasia; Juvenile; Adolescent

## Introduction

Atypical ductal hyperplasia (ADH), a term given to a histological finding of breast tissue resembling low nuclear grade ductal carcinoma *in situ* (DCIS), is a rare finding in adolescent and young adult women [1]. ADH is considered to be a non-obligate precursor to ductal carcinoma with a 4 to 5-fold increased risk of developing breast cancer in bilateral breasts within 5 years [2] and up to 30% in 25 years [3, 4]. Whether this applies to adolescent and young women, however, has not

been investigated. Since 1992, when juvenile ADH was first described, few additional case reports have been published, and even less research has been conducted to demonstrate what this means for this patient population [5-7]. Here, we present the case of an adolescent with ADH involving a fibroadenoma and review the current literature regarding ADH.

## Case Report

### Investigations

An 18-year-old healthy Asian female was referred by her primary care physician for evaluation of a right breast mass. She had no family history of breast cancer, denied any skin changes over the mass and had no nipple drainage or breast pain associated with the mass. On exam, she had an approximately 3-cm nodule at the retroareolar area mostly palpable between 6 - 8 o’clock. No skin dimpling, nipple inversion or nipple discharge was present, and no lymphadenopathy was appreciated.

### Diagnosis

Our patient underwent a targeted ultrasound showing a 3.2 × 1.9 × 3.1 cm benign-appearing palpable mass in the right retroareolar region. Although this had a typical fibroadenoma appearance, given the size, the decision was made with the patient to remove the mass. Our differential included benign fibroadenoma, phyllodes tumor, or malignant mass.

### Treatment

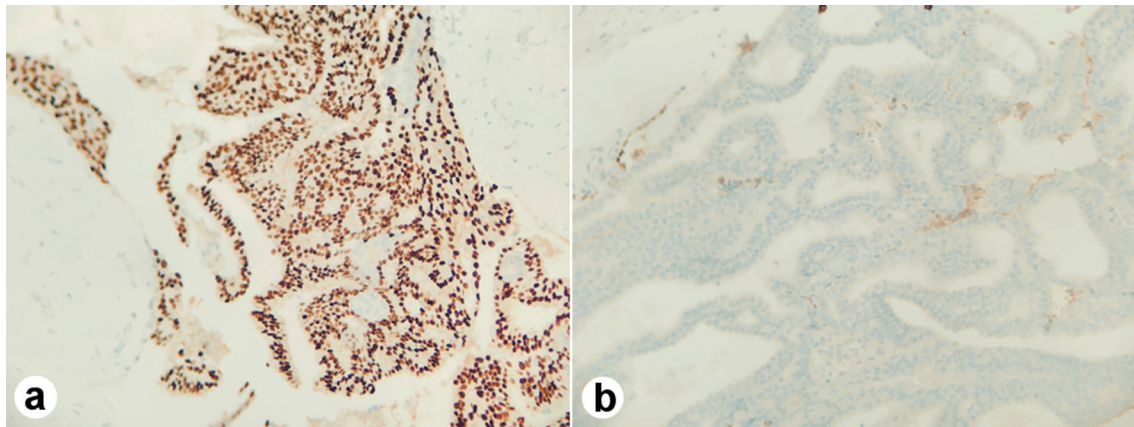
Shortly after, she underwent a right breast excisional biopsy. Two masses within the same specimen measuring at 1.2 × 1.0 × 1.0 cm and 3.8 × 3.0 × 2.8 cm, respectively, were identified on pathology. Grossly, they were noted to be tan-pink to white with typical whorled appearance consistent with fibroadenoma. The two masses, however, differed in their histology. The larger specimen was noted to display micropapillary and cribriform architectural with estrogen receptor (ER)-positive and cytokeratin 5/6 (CK 5/6)-negative staining, consistent with ADH involving a fibroadenoma (Fig. 1a, b). There was focal ADH present beyond the boundaries of the fibroadenoma bordering on carcinoma *in situ* (Fig. 2). The second smaller mass was noted to be a juvenile fibroadenoma.

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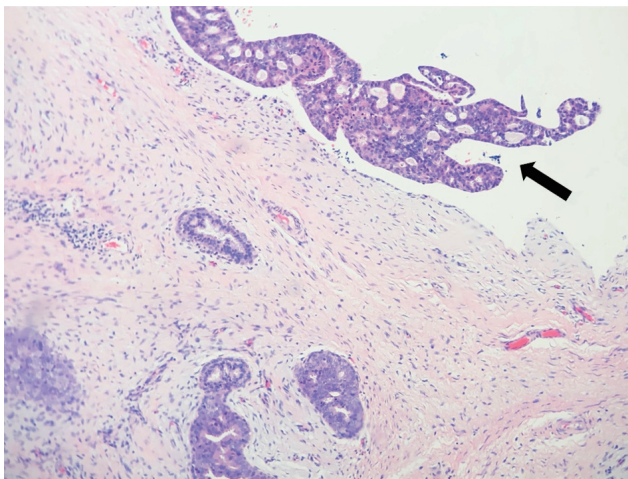
**Figure 1.** The micropapillary and cribriform architectural with estrogen receptor (ER)-positive (a) and cytokeratin 5/6 (CK 5/6)-negative staining (b).

### Follow-up and outcomes

Post operatively, she did well and healed appropriately. We recommended yearly annual exams and breast ultrasounds given that she did have atypia.

### Discussion

Breast masses in adolescent and young adult females are overwhelmingly benign with the incidence of breast cancer in women younger than 25 years old being 3.2 per million [8]. The most common mass found in young women, especially women younger than 20, is a fibroadenoma. A juvenile or cellular fibroadenoma is a subset of adenomas characterized by hypercellular stromal proliferation and more rapid growth than a fibroadenoma with rare malignant transformations [9]. There is no increased cancer risk associated with fibroadenomas and management includes surgical excision only when the masses are symptomatic or rapidly growing [10].



**Figure 2.** Atypical ductal hyperplasia (arrow) beyond the boundaries of the fibroadenoma.

Following biopsy based on atypical mammographic findings or a palpable breast mass, if ADH is found, there is a known cumulative risk of breast cancer of approximately 1% per year [11]. If diagnosed on core needle biopsy, excisional biopsy is recommended as it has been found to be upstaged to DCIS or carcinoma in 15-30% of cases [12]. Risk assessment models have been used to estimate future risk of breast cancer, including the Breast Cancer Risk Assessment Tool (BCRAT), a modified version of the well-known Gail model, and the International Breast Cancer Intervention Study (IBIS), also known as the Tyler-Cuzick model. Both have an adjustment for atypical hyperplasia, however the BCRAT has not been validated for women younger than 35 and shown to underestimate the risk of ADH, while the IBIS has been shown to overestimate the risk [13, 14].

Screening guidelines for women with high-risk lesions are offered by the National Comprehensive Cancer Network (NCCN), the American Cancer Society (ACS), and the American College of Radiology. However, all three indicate there is insufficient evidence to make recommendation for or against magnetic resonance imaging (MRI) screening in women with atypical hyperplasia [11]. Two studies to date have evaluated the role of MRI for ADH and have both concluded that it is not indicated [15, 16].

In regard to chemoprevention, the American Society of Clinical Oncology recommends chemoprevention with tamoxifen for premenopausal women or raloxifene in postmenopausal women with a 5-year absolute risk of breast cancer of 1.7% or greater [17]. Patients with ADH meet this criterion; however, it is unclear how often chemoprevention is used, and we are unable to find discussions regarding its use for juvenile ADH [11].

The most current guidelines regarding ADH are controversial and do not discuss best practices as it relates to juvenile ADH. Our patient highlights the need for further research into ADH, specifically within the adolescent and young adult age group in order to better guide her management.

### Learning points

The incidence of ADH in young females remains widely uninvestigated and unknown. The current risk assessment models do

not incorporate young age into their risk stratification which hinders accurate recommendations and treatment plans for young women with ADH. Considering these current circumstances, honest and open discussions with patients remain paramount. It is extremely important to present all the available information to make treatment and follow-up decisions as a care team.

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## Financial Disclosure

None to declare.

## Conflict of Interest

None to declare.

## Informed Consent

Informed consent was obtained.

## Author Contributions

SD and LOK contributed to literature search, writing and editing. NM contributed to editing of the manuscript.

## Data Availability

The authors declare that data supporting the findings of this study are available within the article.

## Abbreviations

ADH: atypical ductal hyperplasia; DCIS: ductal carcinoma *in situ*; ER: estrogen receptor; CK 5/6: cytokeratin 5/6; BCRAT: Breast Cancer Risk Assessment Tool; IBIS: International Breast Cancer Intervention Study; NCCN: National Comprehensive Cancer Network; ACS: American Cancer Society

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