



Case Report

A Juvenile Female with Ductal Carcinoma *In situ* Arising from a Fibroadenoma

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Abstract

Ductal carcinoma *in situ* (DCIS) and atypical ductal hyperplasia are rare in teenagers. We report an incidental finding of DCIS in a 17-year-old girl who presented with a mass in the right breast. The pathology confirmed lower-grade DCIS in a fibroadenoma. She subsequently underwent right-breast conservative surgery (BCS) without radiotherapy or adjuvant hormone therapy. Six months of clinical surveillance was recommended, and she remained disease-free 25 months after BCS. Hormone therapy and radiotherapy are still controversial in juvenile patients with DCIS, and long-term surveillance and evaluation are still indispensable.

Keywords: Atypical ductal hyperplasia, ductal carcinoma *in situ*, fibroadenoma, teenager

INTRODUCTION

Ductal carcinoma *in situ* (DCIS) is rare in women younger than 30 years of age, and the incidence increases with age.^[1] Atypical ductal hyperplasia (ADH) is considered to be a potential malignancy of DCIS or invasive ductal carcinoma (IDC). A previous study of 1834 cases reported that ADH within a fibroadenoma was not a significant risk factor for breast cancer.^[2] There are only a few reports of teenagers with DCIS or ADH within a fibroadenoma, and no previous study has reported juvenile DCIS with an ADH component. Herein, we present a very rare case of an adolescent with DCIS with ADH, arising from a fibroadenoma.

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CASE REPORT

A 17-year-old girl visited our comprehensive breast cancer center with a palpable mass in the right breast; the painless mass was first noted 2 months previously. She had no family history of breast cancer, denied oral contraceptive pill use, and was a nonsmoker. Ultrasonography demonstrated a 2.12-cm, irregular hypoechoic, heterogeneous echogenic mass lesion over her right breast, at 12 o'clock and 1 cm from the nipple [Figure 1]. Nevertheless, no significant vascularity was present within the tumor. Under shared decision-making, a

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vacuum-assisted breast biopsy was performed. Unexpectedly, microscopic pathology confirmed DCIS Grade 1, noncomedo type, within a fibroadenoma. Histology results were strongly positive for estrogen and progesterone receptors.

Breast magnetic resonance imaging was indicated for tumor region management and excluding other lesions. After discussion with the patient and her parents, breast conservative surgery (BCS) with preservation of the nipple–areola complex was performed. The surgical specimen revealed DCIS tumor cells within a fibroadenoma of Grade 1, and diffuse positivity for hormone receptors was confirmed by immunohistochemistry staining. Additionally, ADH was observed in the resected specimen [Figure 2]. Otherwise, there was no microcalcification in the tumor, and clear resection margins of more than 1 cm were achieved. Considering the influence on the health and

development of teenagers, hormone replacement therapy was not offered. The patient refused genetic testing to detect BRCA1/BRCA2 mutations. Six-monthly clinical breast examinations were recommended, and she remained disease-free after surgery after 25 months of follow-up.

DISCUSSION

Fibroadenoma is one of the most common benign breast tumors in young women. Although benign, there is a 0.002%–0.125% risk of the development of malignant tumors in patients with complex fibroadenomas and those with a family history of breast cancer.^[2,3] In contrast, DCIS is less common in the younger population, and the risk factors include a family history of breast cancer, increased breast density, obesity, early menarche, or late birth.^[3] DCIS is the most common type of noninvasive breast cancer with a lower mortality rate, and the 20-year mortality rate for patients with DCIS after surgical intervention is 3.3%.^[4] DCIS is considered to be a nonessential precursor of invasive breast cancer, and the incidence of breast cancer gradually increases with age.^[5] Additionally, compared to women without ADH, those with ADH have a five times greater risk of developing DCIS or invasive breast cancer.^[6] ADH is considered to be a potential malignancy of DCIS or IDC, and it is histologically characterized as a lesion with structures of monotonous epithelium forming cribriform-like cells. DCIS, ADH, and fibroadenoma are rare in adolescent girls, and only two studies have reported DCIS in female teenagers, and three have reported juvenile ADH [Table 1].^[7–11] Our 17-year-old patient is an extremely rare case of DCIS with an ADH component in a fibroadenoma. Her incidental diagnosis was based on vacuum-assisted breast biopsy, followed by BCS without radiotherapy and hormone therapy, considering the expected side effects on fertility and quality of life during adolescence, as well as further lactation requirements.

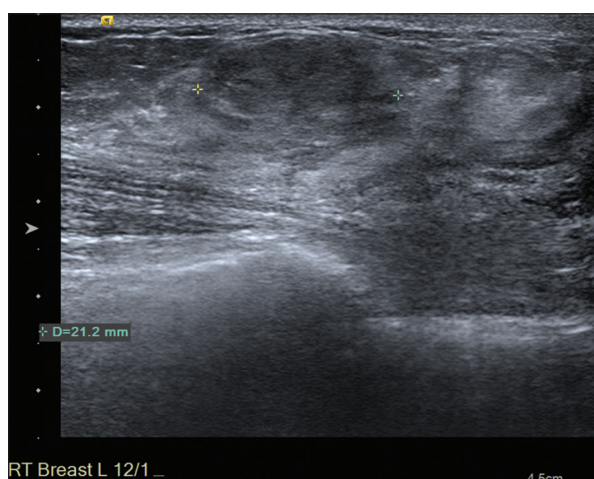


Figure 1: Ultrasonography of the right breast demonstrated a 2.12-cm irregular hypoechoic, heterogeneous echogenic mass lesion over her left breast at 12 o'clock and 1 cm from the nipple

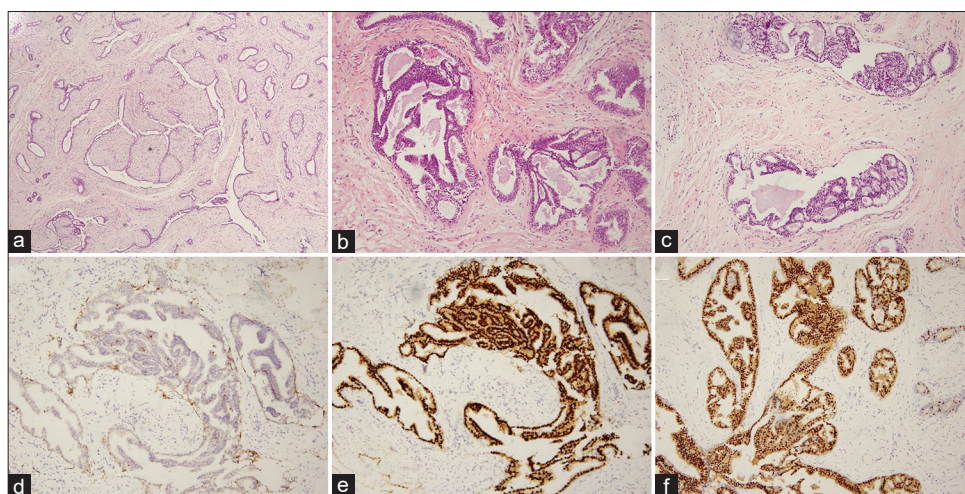


Figure 2: Histopathology images of DCIS with an ADH component in a fibroadenoma. (a) The background showed fibroadenoma, and the benign ducts were surrounded by fibrous stroma. (b and c) Low-grade DCIS was found focally in the fibroepithelial lesion. ADH components, including cribriform-like cells and some myoepithelial cells, were still present. (d) CK5/6 immunostaining was negative in the DCIS. (e) ER was diffusely strongly positive in the DCIS. (f) PR was diffusely strongly positive in the DCIS. ADH: Atypical ductal hyperplasia, DCIS: Ductal carcinoma *in situ*, ER: Estrogen receptor, PR: Progesterone receptor

Table 1: Ductal carcinoma *in situ* and atypical ductal hyperplasia in adolescence population

Published year	Age (years)	Tumor type	With/without fibroadenoma	Treatment	Outcome
T Sato, 2013	17	DCIS	No	Mastectomy; LHRH; Hormone therapy	DF: 1 year
O Frisch, 2018	18	DCIS	Yes	Tumor excision	Not mentioned
D Johnson, 2003	16	ADH	No	Mastectomy	DF: 4 years
S Gandhi, 2018	15	ADH	Yes	Tumor excision	Not mentioned
S Diko, 2022	18	ADH	Yes	Tumor excision	Not mentioned
WY Hung, 2022	17	DCIS, ADH	Yes	BCS	DF: 25 months

LHRH: Luteinizing hormone-releasing hormone, DF: Disease-free, ADH: Atypical ductal hyperplasia, DCIS: Ductal carcinoma *in situ*

There is a lack of data on the definite disadvantages of omitting radiotherapy and hormone therapy in juvenile patients with DCIS, and treating very young patients presents unique challenges. An analysis of patients aged 15–39 years with hormone-positive DCIS who underwent breast conservation therapy suggested that larger tumor areas or narrowing margins were indications for radiation, and approximately 60% of patients were not recommended for hormone therapy. However, a multivariate analysis of predictors for physician recommendations for endocrine therapy did not show an odds ratio.^[12] Another retrospective study reported that radiotherapy could reduce the locoregional recurrence rate in patients younger than 40 years with DCIS >1.5 cm in size. Nevertheless, the study did not analyze the teenage group separately because of the small number of cases.^[13] Sasha reported that patients aged younger than 30 years were significantly less likely to receive endocrine treatment, and the overall survival rate was lower.^[14]

Although DCIS has previously been thought to have a low mortality rate, the risk of invasive recurrence of breast cancer has been reported to be 5.9% in the same breast as DCIS at 20 years and 6.2% for contralateral invasive recurrence. Radiotherapy has been reported to halve the risk of ipsilateral invasive recurrence from DCIS at 10 years in women undergoing curative surgery, but not to reduce breast cancer mortality.^[15] A better understanding of the biological characteristics of those at highest risk of DCIS is imperative for developing targeted approaches to reduce breast cancer deaths.

Although fibroadenoma is a common benign lesion in younger females, DCIS and ADH can develop in fibroadenoma in teenagers. Curative surgery is the main treatment, and hormone therapy and radiotherapy remain controversial in juvenile patients with DCIS. Our patient remained disease-free 25 months after BCS without adjuvant hormone therapy or radiotherapy, and long-term surveillance and evaluation are still fundamental.

Acknowledgment

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal.

The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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