The Korean Journal of Pathology 2014; 48: 164-166
http://dx.doi.org/10.4132/KoreanJPathol.2014.48.2.164

■ BRIEF CASE REPORT ■

Simultaneous Occurrence of Ductal Carcinoma *In Situ* within Juvenile Fibroadenoma in Both Breasts: A Brief Case Report

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Juvenile fibroadenoma is a rare variant of fibroadenoma accounting for 4% of all fibroadenomas and characterized by rapid enlargement of single or multiple, painless, discrete masses, usually occurring in subjects aged < 20 years.[1] The incidence of carcinoma within fibroadenoma is between 0.0125% and 0.3% in the general population,[2,3] and only 0.9% of such carcinomas occur as synchronous bilateral carcinomas arising in bilateral fibroadenomas.[3] Carcinoma arising in fibroadenoma was first described by Cheatle and Cutler.[4] Since then, more than 200 cases have been reported, with the largest series comprising 105 patients reported by Diaz *et al*.[3] in 1991. In Korea, the incidence of bilateral fibroadenomas has been reported to be 9.3%,[5] and one case study reported three cases of ductal carcinoma *in situ* (DCIS) arising in fibroadenoma.[6] However, all DCIS of the three cases occurred from unilateral fibroadenoma.[6] To the best of our knowledge, simultaneous occurrence of bilateral DCIS lesions arising in bilateral juvenile fibroadenomas has never been reported in the Korean literature. Herein, we present an exceptional case of synchronous bilateral DCIS lesions arising within bilateral juvenile fibroadenomas.

CASE REPORT

A 20-year-old Korean woman presented with bilateral, rapidly enlarging breast lumps for one month. Her past medical history was unremarkable, and she denied family history of breast malignancies. On physical examination, the bilateral lumps were found to be about 3 cm, painless, firm and movable. Axillary lymph nodes were not palpable. Ultrasonography (US) revealed a 2.4×1-cm, lobulated, hypoechoic mass in the right breast (Fig. 1A) and a 2.8×1.5-cm, oval hypoechoic mass in the left breast (Fig. 1B), favoring benign-looking nodules. US-guided core needle biopsy was performed on both breasts for histological examination. The core needle biopsy specimens showed pericanalicular and intracanalicular patterns of fibroadenoma and epithelial hyperplasia with mild cytologic atypia, suggesting juvenile fibroadenoma. She underwent a lumpectomy of both breasts for the impression of bilateral juvenile fibroadenomas. Grossly, the right breast mass was a well-circumscribed, lobulated, whitish, solid but softer than typical fibroadenoma, measuring 2.4×2×1 cm, and the left breast mass was relatively circumscribed with a slightly irregular border, measuring 2.8×2.5×1.5 cm. On low-power view microscopic examination, atypical ductal cells with compacted or cribriform, papillary patterns were extensively proliferated within most dilated ducts of fibroadenoma, which revealed half juvenile fibroadenoma and half DCIS in the right breast specimen (Fig. 1C) and two-third fibroadenoma/one-third DCIS in the left breast (Fig. 1D). On high-power view, most proliferative cells were monotonous, round to oval, mildly enlarged cells with relatively abundant eosinophilic or clear cytoplasm and inconspicuous nucleoli forming cribriform and microapillary architectures. Several foci of atypical mitosis, single epithelial necrosis and microcalcifications were identified. The immunohistochemical (IHC) staining for calponin and p63 revealed

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Received: February 13, 2013 **Revised:** June 12, 2013
Accepted: June 13, 2013

pISSN 1738-1843
eISSN 2092-8920

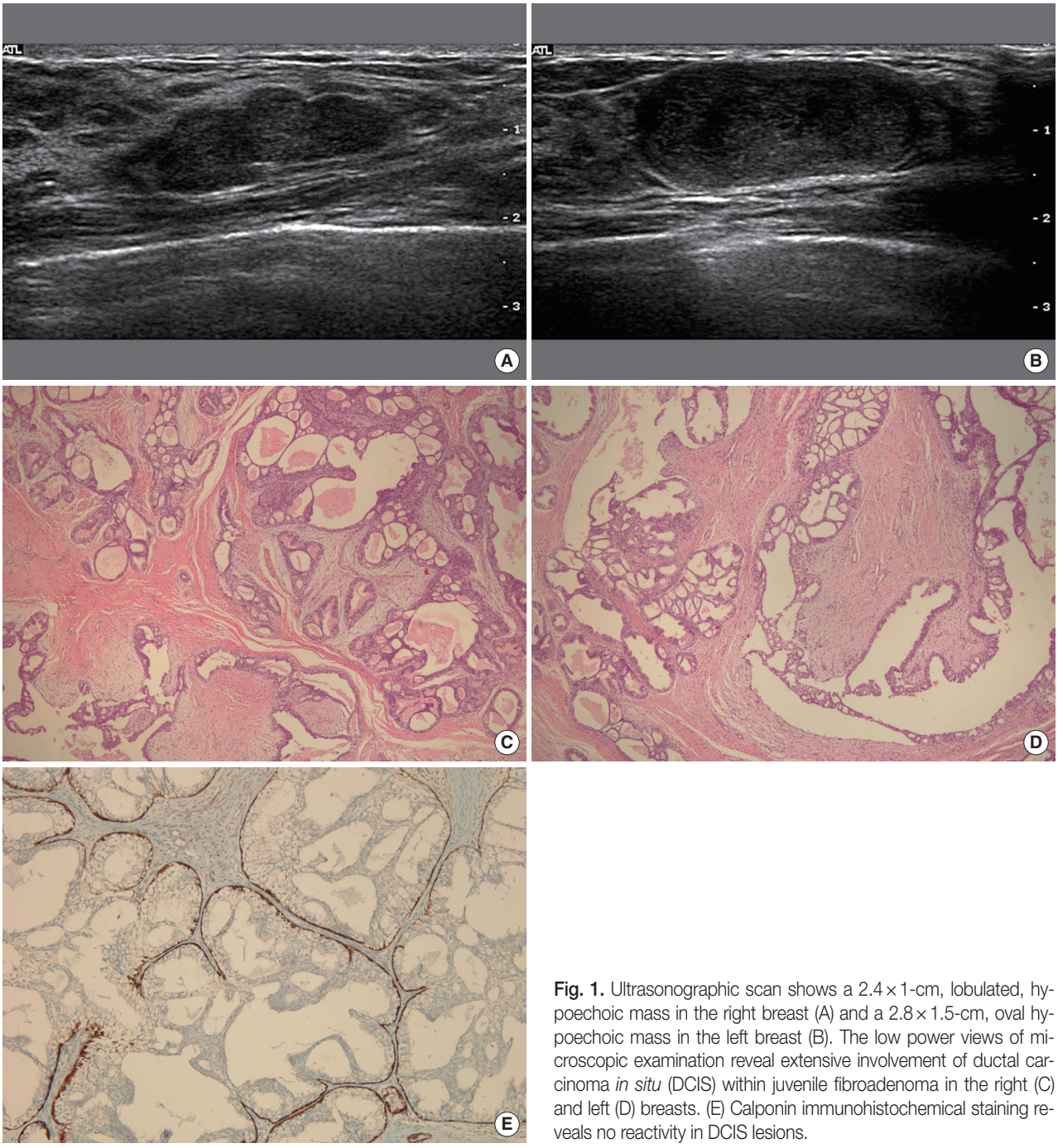

Fig. 1. Ultrasonographic scan shows a 2.4×1-cm, lobulated, hypoechoic mass in the right breast (A) and a 2.8×1.5-cm, oval hypoechoic mass in the left breast (B). The low power views of microscopic examination reveal extensive involvement of ductal carcinoma *in situ* (DCIS) within juvenile fibroadenoma in the right (C) and left (D) breasts. (E) Calponin immunohistochemical staining reveals no reactivity in DCIS lesions.

only myoepithelial cells at the peripheral portion of the proliferative ducts (Fig. 1E). IHC stains for estrogen and progesterone receptors revealed a diffuse strong reaction in the foci of DCIS but discontinuous reaction in the normal and hyperplastic ducts. Foci of DCIS were also detected in adjacent breast tissue to fibroadenoma and resection margins in both breasts. No invasiveness was identified in either breast mass. The stroma of each fibroadenoma was moderately cellular, and pericanalicular and intracanalicular patterns of fibroadenoma were seen in both breast specimens.

After pathologic examination, the patient underwent re-excision of both breasts due to resection margins involving carcinoma within fibroadenoma. The re-excised breast margins were clear of DCIS. The patient received 5,040 cGy of radiotherapy in 28 fractions during over six weeks to both breasts after operation.

DISCUSSION

Carcinoma arising in fibroadenoma usually occurs in women in their fifth decade of life, with the peak age between 42 and 44 years.[3] Most of the bilateral and multicentric carcinomas within fibroadenomas have been shown to be lobular carcinoma.[3,7] Carcinoma of the ductal type within juvenile fibroadenoma is extremely rare. The present case suggests the possibility that bilateral DCIS lesions can occur in bilateral juvenile fibroadenomas in a relatively young patient.

Carcinoma in juvenile fibroadenoma is a diagnostic challenge for surgical pathologists. A hyperplastic myoepithelial cell layer is typically present in juvenile fibroadenoma, which helps discriminate from carcinoma.[1] The diagnostic criteria of DCIS within fibroadenoma requires showing at least one of the following findings: 1) intraductal carcinoma focus is also seen in the adjacent breast tissue and 2) intraductal proliferative lesions within fibroadenoma show cancer-characteristic findings, e.g., epithelial necrosis.[1] Azzopardi[8] also emphasized the involvement of carcinoma in adjacent breast tissue to diagnose carcinomas involving fibroadenomas as follows: those arising within the adjacent breast tissue with engulfing and infiltrating fibroadenoma; those arising within the crevices of fibroadenoma as well as within the adjacent breast tissue; and those restricted entirely or at least dominantly to fibroadenoma. Diaz *et al.*[3] followed general criteria established for the recognition of carcinoma of the breast. Some investigators have defined carcinoma arising within fibroadenoma as having carcinoma cells which are limited to a well-defined fibroadenoma or which only focally extend into the adjacent stroma or ducts.[9] In the present case, foci of intraductal carcinoma were detected in adjacent breast tissue of resection margins and DCIS was predominantly involved in fibroadenoma. Therefore, we confirmed the diagnosis of the present case as DCIS in juvenile fibroadenoma.

The overall biologic behavior of carcinoma which occurs in fibroadenoma is similar to that of breast carcinoma, which does not.[3] Patients with carcinoma in situ arising in fibroadenoma can be treated by tumor excision, with favorable prognoses.[3] However, local recurrences at the original operation site or distant metastases have rarely been reported in the literature.[10] Pick and Iossifides[2] demonstrated that four (10.3%) of the 39 patients with carcinoma *in situ* in fibroadenoma had recurrences or metastases. Diaz *et al*.[3] noted that multicentric carcinomas in situ arise far from the initial biopsy site. Therefore, patients with carcinoma *in situ* in juvenile fibroadenoma should be treated with breast-conserving surgery with a wide margin of clearance.[3]

In summary, although DCIS arising in bilateral juvenile fibroadenomas is a very rare lesion, clinicians, radiologists and pathologists should be aware of the potential for malignancy.

Conflicts of Interest

No potential conflict of interest relevant to this article was reported.

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 http://dx.doi.org/10.4132/KoreanJPathol.2014.48.2.164