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Juvenile fibroadenoma with features of phyllodes tumor showing intraductal growth and prominent epithelial hyperplasia in an 11-year-old girl☆

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Abstract Breast tumors in children are uncommon, with the majority of them being adult-type fibroadenoma (FA). We report a case of juvenile FA (JFA) with features of a benign phyllodes tumor (PT) in an 11-year-old girl, showing very unusual intraductal/intracystic growth. The tumor was located at the outer peripheral portion of the right breast apart from the nipple. Histologically, the tumor showed extensive leaf-like papillary structures with a broad fibrous stroma, protruding into multiple contiguous cystic spaces lined by flat ductal epithelium, and closely resembled PT but the stroma of the tumor was only slightly cellular, showing no nuclear atypia and very few mitotic figures. In contrast, epithelial cells covering the fronds exhibited marked hyperplasia, forming a thick multilayered epithelium. The histology of the tumor with intracystic papillary structures and epithelial hyperplasia showed some similarities with intraductal papilloma (IDP). The mechanism of such unusual intraductal growth of fibroepithelial tumors, including FA/JFA and PT, and their possible common histogenesis with IDP are discussed.

1. Introduction

Breast tumors are uncommon in children and adolescents. The tumor most often seen in these ages is fibroadenoma (FA), which comprises 44–70% of all breast lesions [1,2]. Although adult-type FA accounts for the large majority (93% [3]) of FA cases in childhood, a small proportion of fibroepithelial tumors clinically show rapid growth, often giving rise to large-sized mass lesions, and pathologically exhibit somewhat different histology, including cellular stroma and epithelial hyperplasia, which is often florid. These tumors are called juvenile fibroadenoma (JFA) [3].

Phyllodes tumor (PT) is another fibroepithelial tumor, rarely seen (about 1% [1,2,4]) in children. PT is distinguished

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from FA by higher degree of stromal cell growth and peculiar intracanalicular growth pattern, producing characteristic leaf-like structures with very broad cellular fibrous stroma. PT is classified into three categories, namely, benign, borderline and malignant, depending on the degree of stromal hypercellularity, cytological atypia and mitoses, stromal overgrowth, and nature of tumor borders/margins [5].

Although JFA typically lacks an intracanalicular growth pattern and leaf-like structures, which characterize PT, JFA may exhibit such a growth pattern and show considerable overlapping features with benign PT, both clinically as well as pathologically [2,4]. Some authors have included benign PT in childhood and adolescents into the entity and concept of JFA [4]. Here, we report a case of JFA with an extensive phyllodes pattern in an 11-year-old girl, which showed extremely unusual intraductal growth in multiple contiguous cystic spaces along with coincident, prominent epithelial hyperplasia, showing some similarities with intraductal papilloma (IDP). Only a limited number of cases of fibroepithelial tumors, including FA and PT, showing intraductal/intracystic growth have been reported in the literature, with some of them described as having foci of IDP. These cases may indicate a possible causative link and common histogenesis between intraductal fibroepithelial tumors and IDP.

2. Case report

An 11-year-old girl visited the hospital since she had noticed a palpable lump in her right breast for 5 months, during which it gradually increased the size. She had no particular past history and family history for breast cancer and her general health condition was good. Physical examination confirmed a well-defined movable mass at the boundary portion between upper and lower outer quadrants of her right breast, apart from the nipple. Ultrasonography revealed a slightly lobulated oval tumor with sharply demarcated smooth margins, measuring 19 × 15 × 10 mm in diameter (Fig. 1). The tumor showed slightly hypoechoic homogeneous internal structure with slight posterior echo enhancement. No signs of echogenic spots indicative of calcification were present. FA was clinically suspected and the tumor was surgically enucleated. The resected tumor was pathologically diagnosed as JFA with features of benign PT as described below and the patient underwent postoperative follow-up with physical examination and ultrasonography at 3- to 6-month intervals. She has been well without any signs of recurrence for 3 years since the surgery.

3. Results

On gross examination, the surgically resected specimen was a fairly circumscribed tumor, the cut surface of which was solid, yellowish white in color, and 17 × 14 mm in diameter. Histologically, the tumor consisted of fibroepithelial cells forming large leaf-like structures with a broad fibrous stroma covered by thick columnar epithelial layers, protruding into multiple contiguous cystic cavities (Fig. 2A and B). This phyllodes pattern was extensively present throughout the tumor, with only a small part showing an intracanalicular FA pattern. The inner surface of the cyst wall was covered with a thin monolayer of flat ductal epithelium (Fig. 2C and D). The tumor contained a mildly cellular stroma, with some

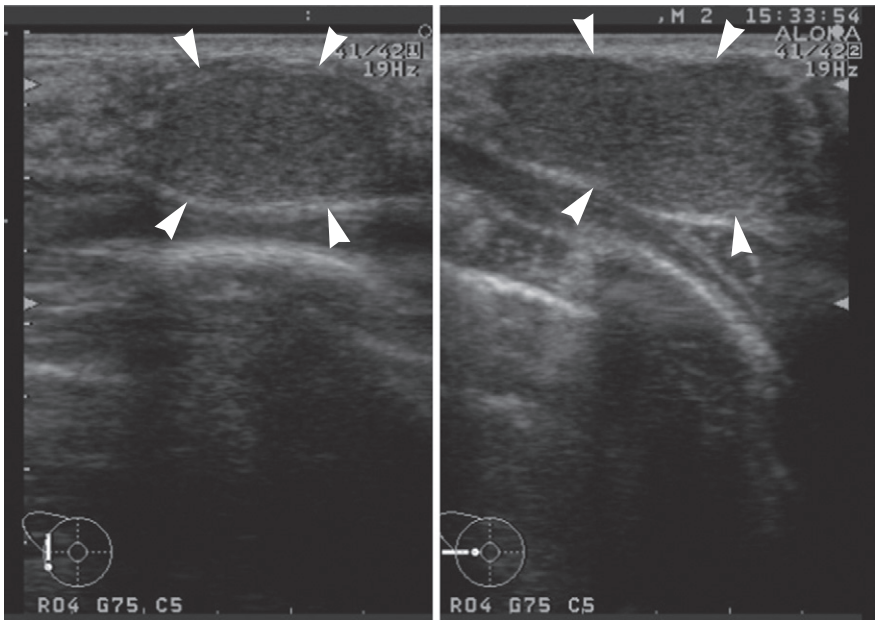

Fig. 1 Ultrasonography. The tumor exhibits a slightly lobulated oval mass lesion with smooth margins (arrow heads), slightly hypoechoic homogeneous internal structure and slight enhancement of posterior echoes.