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Juvenile Cellular Adenofibroma of Breast

— A case report —

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Juvenile cellular adenofibroma of the breast is a unique neoplasm of the breast that should be differentiated from other important benign and malignant lesions of the juvenile breasts.

We report a case with it's characteristic clinical, gross and histological features. The tumor was in the right breast with the size of 20 cm in maximum extent. This patient was also associated with hemihypertrophy of the right side. Microscopically the masses were characterized by prominent stromal cellularity associated with pericanalicular duct proliferation.

Key Words: Breast, Fibroadenoma, Childhood tumor, Adenofibroma

INTRODUCTION

Fibroadenoma is the most common cause of breast masses in adolescent girl, followed by virginal hypertrophy[1,2]. Although most of these lesions are of little diagnostic difficulty histologically, a small number of these fibroepithelial lesions present with marked stromal hypercellularity and coincidental intraductal epithelial proliferation.

The lesions with these stromal hypercellularity and florid intraductal hyperplasia have been designated as "fetal" or "juvenile" adenofibroma or benign cystosarcoma phyllodes[3,4]. Recently Pike and Oberman[5] proposed to use "juvenile (cellular) adenofibroma" to connote a distinctive tumor that most often occurs in the adolescent girl, attaining large size and exhibiting prominent cellularity of both epithelium and stroma.

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CASE REPORT

This 12 years old girl was admitted to Seoul National University Children's Hospital because of a huge mass in the right breast that developed 2 years ago. She had a history of repeated operations for hemihypertrophy of the right arm and leg. The pathologic examinations of arm lesions were reported as mature fatty tissue (lst operation), lymphangioma (2nd operation) and mature fat (3rd operation).

Pathologic Finding: Received specimen was a large ovoid mass covered with an ellipse of intact skin. The mass was well delineated from the surrounding breast tissue, and measured 20×11×7 cm. Cut sections showed pinkish white solid masses, in which many cystic structures of variable size, ranging from 0.3 to 1.5 cm were seen (Fig. 1). The tumor masses were irregularly mixed with fatty tissue. The cystic lesions were filled with the grayish white nodules of papillary nature and contained jelly-like material easily shelled out from the smooth surfaced cystic wall. Remaining solid portion showed

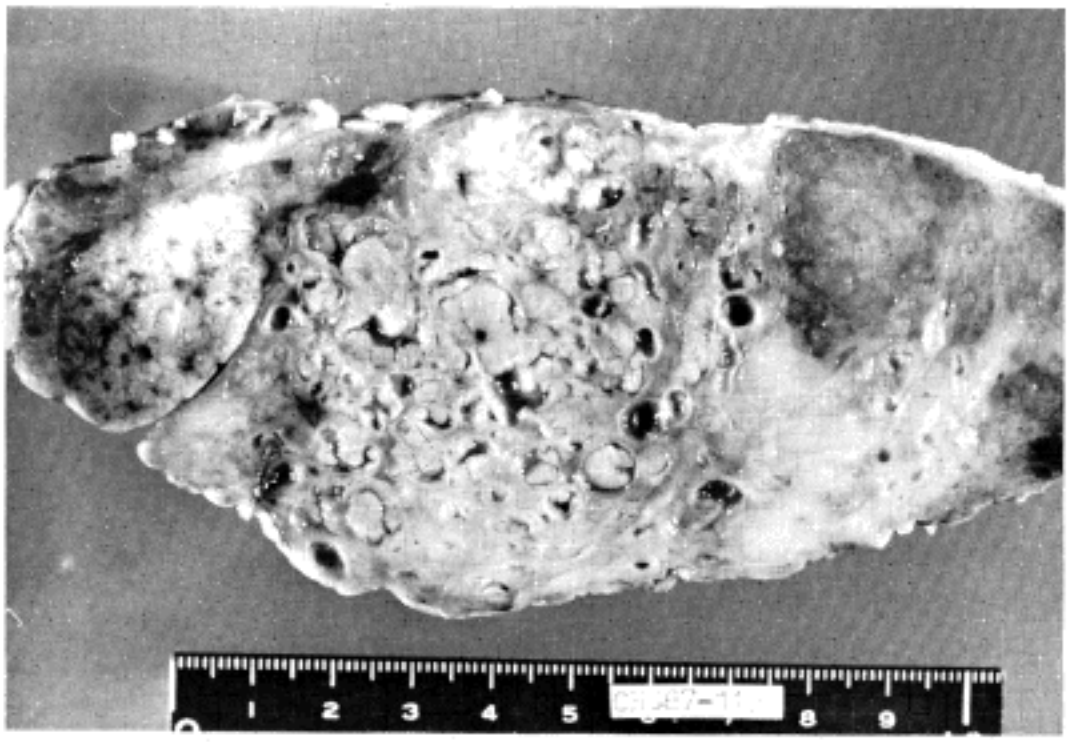

Fig. 1. The tumor is well demarcated and showing poorly defined multinodular masses and many cystic structures containing nodules of papillary nature.

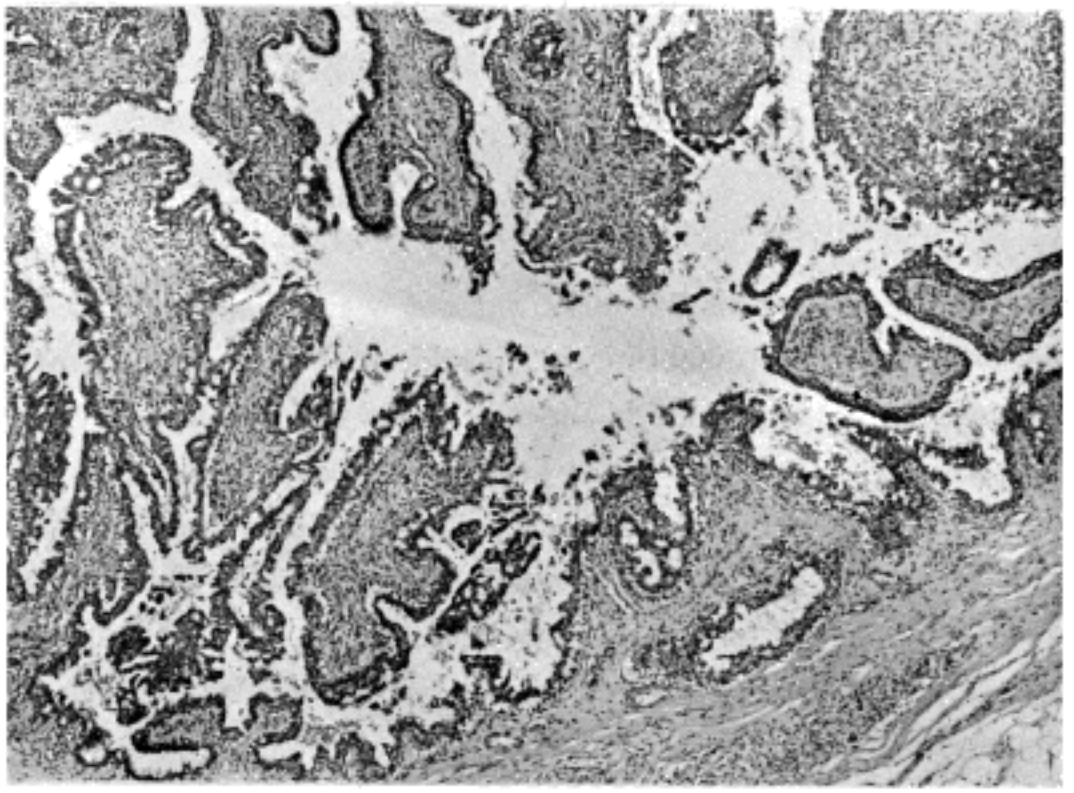

Fig. 2. Photomicrograph of the tumor showing the branching, leaf-like pattern that is also a feature of cystosarcoma phyllodes. But no atypism of stromal cells is present (H&E, x40).

glistening and poorly defined multinodular appearance. Microscopically, these masses consisted of proliferation of both ductal epithelial and stromal connective tissue components. Active proliferation of fibroblasts associated with collagenization was seen prominently around the ducts, and it also protruded into the ductal lumens to take the branching or leaf-like pattern. The cellular stroma lacked the mesenchymal mucopolysaccharide or cellular atypia characteristic of the usual adenofibroma. Grossly visible cystic lesions consisted of dilated ducts containing irregular papillary masses formed by hyperplastic ductal epithelial lining and dense stromal cores (Fig. 2). Also seen were eosinophilic secretory material and floating macrophages in the dilated ducts. In the peripheral solid area of tumor, lobule formation was present, with separation of lobular ducts by the stromal components. The ducts maintained regular round configuration, but showed circumferential or focal papillary nature of

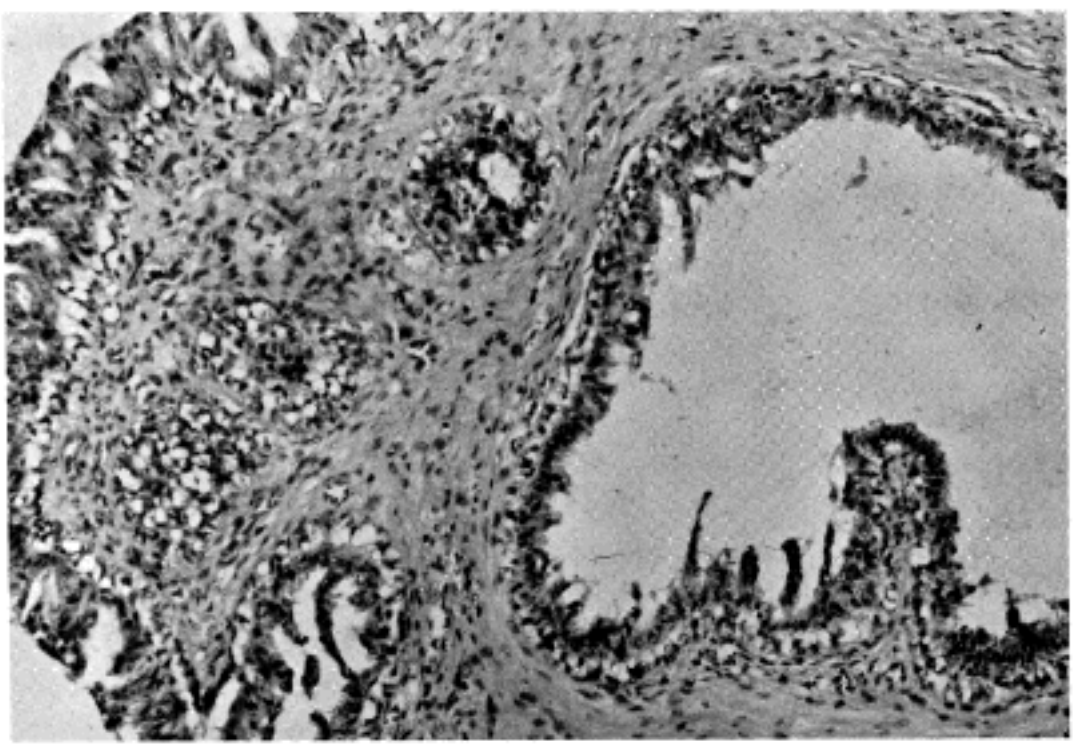

Fig. 3. Intraductal epithelial hyperplasia associated with stromal collagenization. (H&E, x200).

epithelial cell hyperplasia (Fig. 3). Despite cellular atypia or mitotic figures were seldom found, and if present it was typical mitosis.

DISCUSSION

Fibroepithelial tumor of the breast in pediatric age usually cause little problem in making diagnosis for the pathologists. However, certain tumors cause a considerable diagnostic difficulty because of its large size, rapid growth and more importantly hypercellularity and intraductal epithelial hyperplasia. This case presented exactly the same problems as it attained a size of 20 cm, had a history of recent rapid growth and a marked intraductal papillary growth as well as stromal hypercellularity.

The differential diagnosis of this tumor lies among papillary duct hyperplasia, papillomatosis and cystosarcoma phyllodes. Papillary duct hyperplasia (papilloma and/or papillomatosis) are relatively commom breast lesions in adult women. However, they are rarely seen in women younger than 30 years of age or in children. There are three types, i.e., papilloma, papillomatosis and sclerosing papillomatosis. Juvenile papillomatosis (JP) is different from papillary duct hyperplasia as it lacks gross cystic change, apocrine metaplasia and stasis. Rosen[6] reported initially 37 children and young adult women aged 10 to 26 years (average 17 years). Since its description in 1978 by Rosen et al.[3] juvenile papillomatosis became recognized as a specific clinicopathologic disease entity. The clinical preoperative diagnosis is most often fibroadenoma. Cysts are a dominant microscopic feature in conjunction with duct hyperplasia. At low power field the "swiss cheese" characterization is evident by the presence of the dilated ducts and cysts. Although size of tumor is not a criterion for the differential diagnosis, the majority of hypercellular adenofibroma is in excess of 10 cm in diameter, while juvenile papillomatosis is 3 cm in greatest dimension. Papillary apocrine metaplasia is common. It was noted that surprisingly high frequency of breast carcinoma has been found among relatives of patients with juvenile papillomatosis and that these patients may develop mammary carcinoma coincidental with JP or subsequently[7].

For cystosarcoma phyllodes juvenile cellular fibroadenoma lacks the exaggerated intracanalicular growth pattern seen in most cystosarcomas. According to Pike and Oberman[5] juvenile cellular adenofibroma presents most often as a solitary mass, ocu-

rring shortly after menarche. However, this tumor is not exclusively found in juveniles. Examples of "Juvenile" adenofibroma of this type can be enocountered in adults. It should be reminded that the adult-type adenofibroma is still far more common among fibroepithelial tumors during adolescence[1].

These patients with juvenile cellular fibroadenoma should be treated by local excision. Even when the tumor occupies most of the breast, preservation of minimal amounts of breast tissue can result in normal breast development. These patients who present with multiple and bilateral tumors will suffer repeated recurrences[5].

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＝국문요약＝

유방의 약년성 세포충실성 선섬유종(1증례)

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지 제 근·서 연 림

유방의 약년성 세포충실성 선섬유종은 1985년 Pike와 Oberman에 의하여 기술된 양성종양으로서 어린이에 호발하며 육안 및 현미경적으로 악성종양을 의심케하는 소견을 나타내기 때문에 잘 알아 두어야 할 병변이다.

이 증례는 12세 여아의 오른쪽 유방에서 발생한 20 cm 크기의 종괴로서 육안적으로 낭성구조와 유두성 성장이 관찰되었으며 현미경적으로 심한 관내의 상피증식과 더불어 관주위의 섬유모세포 증식이 현저하였다. 관내 상피증식은 흔히 유두성 성장을 보였으나 괴사나 간질의 침윤은 관찰되지 않았다.

이 종양은 반드시 어린이에만 발생하는 것은 아니며 드물지만 어른에게도 발생하지만 어디까지나 양성종양이고 악성화의 가능성이 거의 없다는 것은 이 연령층에서 생길 수 있는 유방의 기타 병변과의 감별이 필요하다.