

Recurrent juvenile fibroadenoma of the breast in an adolescent

A case report

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Abstract

Rationale: Most breast masses are benign and are rare in adolescent girls. The most frequent tumor is fibroadenoma. Juvenile fibroadenoma is a rare variant of fibroadenoma and is characterized by rapidly enlarging, painless, and unilateral masses occurring at ages between 10 and 18 years.

Patient concerns: A 17-year-old girl who first presented to a hospital due to a left breast tumor.

Diagnoses: Juvenile fibroadenoma of the left breast.

Interventions: She underwent fibroadenoma excision, but she complained of a mass that presented in her left breast 3 months after surgery. After 2 years of observation, the mass became tender and enlarged. We conducted a wide excision of the tumor. Recurrent juvenile fibroadenoma with hypercellular fibromyxoid stroma of the breast was noted.

Outcomes: The patient is in good general condition without tumor relapses during the 4 months of follow-up.

Lessons: Recurrent fibroadenomas, particularly those of a large size with a rapid growth rate, in the same breast suggest a high possibility of transformation. We recommend wide tumor resection.

Abbreviations: BI-RADS = Breast Imaging Reporting and Data System, HPF = high power field.

Keywords: adolescent, fibroadenoma, juvenile, recurrence

1. Introduction

Breast masses in adolescence are uncommon and are mostly benign or self-limited. The most common benign tumor in adolescent girls and young women is fibroadenoma, followed by fibrocystic disease.^[1] Fibroadenomas can be of juvenile, cellular, or giant types. Juvenile fibroadenoma is a rare variant of fibroadenoma and is characterized by rapidly enlarging, painless, and unilateral masses occurring at ages between 10 and 18 years.^[2] The potential maximum size is >10 cm, which can result in an esthetic problem due to breast asymmetry or hypertrophy.^[3]

Juvenile fibroadenomas account for 0.5% and 4% of all fibroadenomas.^[4] On physical examination, they are often well-defined and movable and exhibit a rubbery texture.^[5] Treatments

for juvenile fibroadenomas include surgical resection or observation.^[6]

We report the case of a 17-year-old girl who underwent resection for juvenile fibroadenoma and who showed recurrent unilateral breast juvenile fibroadenoma. We successfully resected the recurrent tumor. The patient has provided informed consent for publication of the case (CR107–02).

2. Case report

A 17-year-old girl presented to our hospital complaining of a left breast mass that had been growing progressively for several months. Medical and family histories did not appear to contribute to the present illness. On physical examination, a well-defined 2-cm tumor was palpable at the 9 o'clock position and was located 1 cm from the areola on the left breast. The nodule was movable and soft. Breast sonography revealed a round hypoechoic nodule measuring 2.3 × 1.7 × 1.2 cm in the left breast. The Final Breast Imaging Reporting and Data System category was 2, revealing a benign mass.

The breast tumor was widely excised due to the patient's symptoms. A skin incision approximately 2 cm in length was made above the tumor. The incision was made deep down to the Cooper fascia. The tumor with the surrounding breast tissue was excised with a safety margin of approximately 1 cm. Surgical specimens were sent for histologic examination at a pathology laboratory; based on the results, a diagnosis of juvenile fibroadenoma with a glandular structure and fibrous stroma was made.

The patient noticed a palpable painless lump in her left breast approximately 3 months following surgery. Physical examination revealed a 2 × 1.5 cm tumor located in the left inferior quadrant beneath the scar near the areola. No axillary nodes

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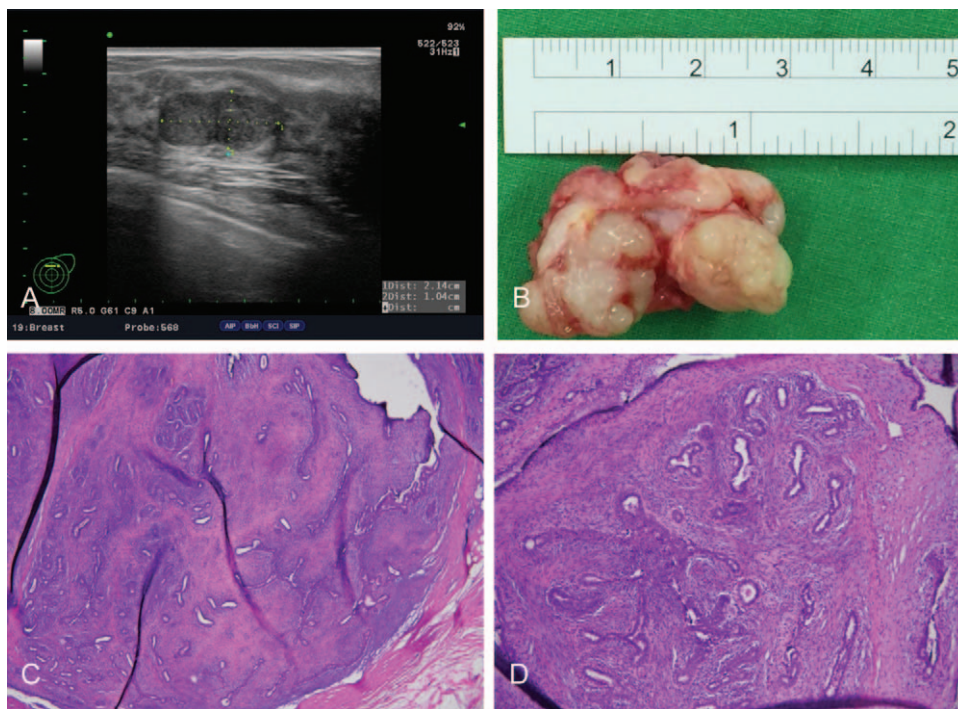


Figure 1. Ultrasonographic, gross, and pathological image of juvenile fibroadenoma. A, Ultrasonographic image of juvenile fibroadenoma. B, Gross image of juvenile fibroadenoma. C, Recurrent juvenile fibroadenoma with a well-defined border and fibromyxoid stroma. D, Higher magnification shows stromal hypercellularity.

were present. Tumor recurrence was noted, and conservative therapy with observation was recommended.

Two years later, an enlarged breast tumor with tenderness was noted again. Breast ultrasound approximately 2 years after surgery revealed a 2.14×1.04 cm tumor at the 12 o'clock position that was 2 cm from the areola and showed posterior enhancement (Fig. 1A). The patient subsequently underwent left breast partial mastectomy for the removal of the 2 cm-wide mass (Fig. 1B). Pathology indicated a mass measuring $3.0 \times 2.2 \times 1.3$ cm that showed histologic features consistent with those of juvenile fibroadenoma. The recurrent tumor showed hypercellular fibromyxoid stroma (Fig. 1C, D). The patient has been regularly followed up and is currently in good general condition without evidence of recurrence at her 4-month follow-up visit.

3. Discussion

We report a case with recurrent juvenile fibroadenoma received tumor resection in a 17-year-old girl. Fibroadenoma is the most common type of benign breast mass in premenopausal women^[11]; however, it is rare in the pediatric and adolescent populations, accounting for only 1% of breast masses. The pathogenesis of fibroadenoma is unknown, but estrogen may contribute to its development.^[11] Juvenile fibroadenomas typically develop at the onset of puberty.^[4] During puberty, the lobules and stroma in the breast may respond to the increased levels of estradiol and progesterone, leading to an increase in breast size of up to 15% and the development of single or multiple palpable fibroadenomas. Fluctuations in breast size may occur with the menstrual cycle.^[7,8] Thus, fibroadenomas can range from asymptomatic masses to painful and fast-growing tumors. In our case, she presented with a painful and fast-growing breast tumor.

The diagnosis of juvenile fibroadenomas relies on careful history and physical examination; these fibroadenomas clinically present as painless, mobile, solitary, unilateral, and rapidly growing breast masses with a distinct border.^[9] A comprehensive gynecology history should also be obtained, including menarche, menstrual cycle, pregnancy history, and the first occurrence of the breast mass. Once the breast mass is observed, gynecologists should perform detailed breast examination through inspection and palpation to document tumor location, size, texture, skin change, nipple discharges, and lymph nodes. Imaging studies such as ultrasound, mammography, or magnetic resonance imaging studies should also be performed. In our case, detailed history taking and physical examination were performed and found a left breast tumor located at 12 o'clock direction.

In adolescents, breast ultrasound is considered the most effective diagnostic tool due to breast tissue density. The most common sonographic feature of juvenile fibroadenoma is a well-defined hypoechoic or isoechoic mass with posterior acoustic enhancement and hypervascularity in color Doppler sonography. The diameter of the mass shows wide variation.^[10,11] In a retrospective study, Kim et al^[10] reported the size of 34 juvenile fibroadenomas in Korea ranged from 8 to 110 mm, with a mean size of 30 mm. Giant fibroadenomas are defined as those more than 50 mm in diameter and more than 500 g in weight. They comprise more than 80% of the breast's mass.^[12] These tumors represent approximately 1% of breast masses in adolescents. In our case, breast ultrasound found a 20 mm breast tumor with posterior acoustic enhancement. However, we did not find hypervascularity in the tumor.

Histologic features of juvenile fibroadenomas are well-circumscribed lesions with predominantly pericanalicular growth patterns and hypercellular stroma, accompanied by intraductal epithelial hyperplasia. Leaf-like fronds are less common.^[13,14] In

the presented case, the tumor showed a glandular structure and hypercellular fibromyxoid stroma, which was compatible with juvenile fibroadenoma.

Management of juvenile fibroadenoma ranges from tumor excision to simply observation.^[4] Spontaneous regression may occur for small fibroadenomas less than 5 cm. Surgical intervention is indicated for fibroadenomas that are more than 5 cm in diameter, rapidly growing, cause severe pain, distort the breast architecture, or lead to overlying skin changes. Multiple and bilateral breast masses, a persistent mass without apparent regression, those with stromal hypercellularity, and those with cystic change on ultrasound are also indicated for surgery. Suspicions of malignancy, presence of a high-risk genetic mutation or syndrome, or a histologically complex fibroadenoma are the indications for surgical resection.^[15] In our case, we resected the tumor due to painful and fast-growing tumor.

Preoperative assessment should be performed, and informed consent should be obtained from the affected patient; it is vital to clarify the risks versus the benefits of intervention, including scarring and contour deformities of the breast after the surgery.

Malignancy associated with preexisting fibroadenoma in adolescents is rare.^[4] The incidence of malignant transformation ranges from 0.002% to 0.125%.^[16] However, several studies have indicated a higher risk of subsequent breast cancer in patients with fibroadenoma than in the healthy population.^[17] Therefore, close follow-up of these patients is warranted.

Open excision of fibroadenomas under local or general anesthesia is still the most frequently used technique for enucleation of fibroadenomas.^[18] Wang et al^[19] examined ultrasound images obtained 6 months after ultrasonographically guided vacuum-assisted excision and demonstrated a recurrence rate of 3.4%. In our case, she received open excision of the fibroadenoma.

Cowan et al^[20] retrospectively reviewed 90 cases of fibroadenoma that showed phyllodal features and positive surgical margins and revealed a low recurrence rate of 3%. Recurrence is more likely to occur for masses larger than 2 cm at initial diagnosis, with an overall recurrence of 15% over 22 months, and for masses with a considerable stromal mitotic count of >2 per 10 high power fields. Recurrence may not be associated with the surgical margin, age of the patient, or position of the mass.^[14,21] Follow-up is suggested at 4 and 8 weeks postoperatively, every 3 months in the next year, biannually in the second year, and then annually.^[3] This case also had one time of recurrence within 2 years with the initial tumor diameter larger than 2 cm. In our case, we also arranged follow-up schedule as the above suggestion.

In conclusion, benign breast diseases in the pediatric and adolescent populations are commonly encountered by practicing gynecologists. These diseases may evoke anxiety and concerns in both the patient and family. Therefore, a communicative approach is crucial when caring for young women with a breast mass. The physician should reassure the patient by fully explaining the natural and benign courses of fibroadenomas. Most fibroadenomas can be managed conservatively without surgery. However, juvenile fibroadenomas of larger sizes are associated with an increased risk of recurrence. Surgical excision may be performed if the mass is symptomatic. Due to the higher risk of subsequent breast cancer in patients with fibroadenoma, these patients should undergo regular follow-up to assess the outcome, complication, and recurrence postoperatively. It is vital to rule out malignant transformation promptly and accurately.

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All authors contributed equally in concept, literature review, and drafting of the manuscript and approved the final version of this manuscript.

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References

- Valdes EK, Boolbol SK, Cohen JM, et al. Malignant transformation of a breast fibroadenoma to cystosarcoma phylloides: case report and review of the literature. *Am Surg* 2005;71:348–53.
- Wechselberger G, Schoeller T, Piza-Katzer H. Juvenile fibroadenoma of the breast. *Surgery* 2002;132:106–7.
- Cerrato F, Labow BI. Diagnosis and management of fibroadenomas in the adolescent breast. *Semin Plast Surg* 2013;27:23–5.
- Ng WK, Mrad MA, Brown MH. Juvenile fibroadenoma of the breast: treatment and literature review. *Can J Plast Surg* 2011;19:105–7.
- Greydanus DE, Matytsina L, Gains M. Breast disorders in children and adolescents. *Prim Care* 2006;33:455–502.
- Sosin M, Pulcrano M, Feldman ED, et al. Giant juvenile fibroadenoma: a systematic review with diagnostic and treatment recommendations. *Gland Surg* 2015;4:312–21.
- Houssami N, Cheung MN, Dixon JM. Fibroadenoma of the breast. *Med J Aust* 2001;174:185–8.
- Santen RJ, Mansel R. Benign breast disorders. *N Engl J Med* 2005;353:275–85.
- Chung EM, Cube R, Hall GJ, et al. Breast masses in children and adolescents: radiologic-pathologic correlation. *Radiographics* 2009;29:907–31.
- Kim SJ, Park YM, Jung SJ, et al. Sonographic appearances of juvenile fibroadenoma of the breast. *J Ultrasound Med* 2014;33:1879–84.
- García CJ, Espinoza A, Dinamarca V, et al. Breast US in children and adolescents. *Radiographics* 2000;20:1605–12.
- Gobbi D, Dall'Igna P, Alaggio R, et al. Giant fibroadenoma of the breast in adolescents: report of 2 cases. *J Pediatr Surg* 2009;44:e39–41.
- Pike AM, Oberman HA. Juvenile (cellular) adenofibromas. A clinicopathologic study. *Am J Surg Pathol* 1985;9:730–6.
- Tay TKY, Chang KTE, Thike AA, et al. Paediatric fibroepithelial lesions revisited: pathological insights. *J Clin Pathol* 2015;68:633–41.
- Jayasinghe Y, Simmons PS. Fibroadenomas in adolescence. *Curr Opin Obstet Gynecol* 2009;21:402–6.
- Wu YT, Chen ST, Chen CJ, et al. Breast cancer arising within fibroadenoma: collective analysis of case reports in the literature and hints on treatment policy. *World J Surg Oncol* 2014;12:335.
- El-Wakeel H, Umpleby HC. Systematic review of fibroadenoma as a risk factor for breast cancer. *Breast* 2003;12:302–7.
- Lee M, Soltanian HT. Breast fibroadenomas in adolescents: current perspectives. *Adolesc Health Med Ther* 2015;6:159–63.
- Wang WJ, Wang Q, Cai QP, et al. Ultrasonographically guided vacuum-assisted excision for multiple breast masses: non-randomized comparison with conventional open excision. *J Surg Oncol* 2009;100:675–80.
- Cowan ML, Argani P, Cimino-Mathews A. Benign and low-grade fibroepithelial neoplasms of the breast have low recurrence rate after positive surgical margins. *Mod Pathol* 2016;29:259–65.
- Grady I, Gorsuch H, Wilburn-Bailey S. Long-term outcome of benign fibroadenomas treated by ultrasound-guided percutaneous excision. *Breast J* 2008;14:275–8.