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JUVENILE GIANT BREAST ADENOFIBROMA: A CASE REPORT AND LITERATURE REVIEW

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ABSRACT

Juvenile giant adenofibroma is a rare benign mastopathy. It is a particular form of adenofibroma, occurring in adolescence, and requires special diagnostic and therapeutic management. The main etiopathogenic factor is relative hyperoestrogenism related to early puberty. The differential diagnosis is made with phyllodes tumor. Its appearance can be worrying due to the large size of the tumor, the rapid growth, the inflammatory aspect of the skin in front of it, and the collateral venous circulation. We report a case of giant juvenile adenofibroma in a 12 years old girl at the Mohammed VI Center for Cancer Treatment at the IBN ROCHD University Hospital in Casablanca. The aim is to review the clinical, radiological and histological diagnosis and therapeutic management.

INTRODUCTION

Adenofibroma is a benign tumor consisting of stroma and glandular epithelial cells. It constitutes 10% of benign tumors in the female population, and 75% in adolescents.[1,2] Giant fibroadenoma is a rare benign mastopathy and constitutes 2-4% of fibroadenomas[2] more frequent in adolescent girls. It differs from adenofibromas by its rapid growth, evolution, and its size of more than 5 cm and up to 20 cm. Its pathophysiology remains poorly understood and is thought to be the consequence of an inadequate local response to estrogenic stimulation.[3,4] In its juvenile form, fibroadenoma generally occurs after the onset of the first menstrual period. Usually between 10 and 18 years of age.[2,5] But there is also a second peak of occurrence around the peri-menopause. Its diagnosis is clinico-radiological. Its main diagnostic difference is with phyllodes tumor, and only histology allows to make a diagnosis of certainty. The treatment is surgical, whose interest is essentially aesthetic in order to lose the elasticity of the skin.[3]

CASE REPORT

Mrs. G.K, 12 years old, without any particular pathological history, with the onset of menarche at the age of 10.5 years, irregular cycles, and primary dysmenorrhea. No medication or oral contraceptives were mentioned. The young girl presented to the clinic with a nodule of the left breast, which had increased rapidly in size over a period of four months, with redness opposite and associated with mastodynia, without any notion of prior trauma. On clinical examination, the left breast was enlarged, red, with collateral circulation, with a deformed the nipple and areola complex, a mass of 8 x 8 cm taking almost all the upper quadrants, irregular, firm, without associated nipple discharge. Examination of the right breast was without patricularity, the axillary and supraclavicular lymph node areas were free. Other secondary sexual characteristics were present.

On breast ultrasound the mass was homogeneous hypoechoic polylobate, measuring 40x60x40mm, with fibrillated trabeculae, and posterior enhancement, vascularized on color Doppler, seizing in the superior-internal quadrants, with no notable abnormalities in the contralateral breast or axillary lymph nodes. Evoking at first a giant adenofibroma or a phyllodes tumor.

A trucut biopsy was performed and found a fibroepithelial proliferation that could be related to an adenofibroma or a phyllodes tumor. The surgical indication was a left oriented lumpectomy with an extemporaneous examination which revealed an adenofibroma. The final pathological examination revealed a 68 gram tumor measuring 8x6.5x2.5 cm, encapsulated, white-beige fasciculated, flush with all the borders. Microscopic examination showed a well-limited fibroepithelial proliferation. The epithelial component is made up of galactophoric ducts of variable size, sometimes dilated and ectatic. They are lined by a hyperplastic lining with intra luminal papillary

projections, associated with florid epithelial hyperplasia without atypia. The stromal component is fibrous with moderate cellularity without atypia, interspersed with lymphoplasmacytic elements.

The patient stayed in the department for 3 days. The postoperative course was simple.

The follow-up maintained for 6 months did not reveal any recurrence with a total regression of the breast edema.

DISCUSSION

Giant breast fibroadenoma represents 2 to 4% of fibroadenomas. It can present in two forms, juvenile and adult giant adenofibroma.

It is more frequent in young African and Asian girls with an average age between 11 and 16 years at the time of diagnosis.[6] Our patient was 12 years old.

The juvenile form may be of abrupt onset with a large fibroadenoma size in non-pubescent patients or those close to menarche. In these patients, no contraception is taken. However, the proximity of puberty and the major hormonal changes that result from it probably play a fundamental role in the physiopathology.[7]

The nodule increases rapidly in volume to reach an average size of 10 to 20 cm, occupying the whole breast, with regular contours, round, lobulated or even poly-lobulated, mobile in relation to the superficial and deep planes, revealing collateral venous circulation, an orange peel appearance, or rarely ulcerations of the skin opposite. The nodule may sometimes be accompanied by a mass effect, explaining the associated pain.[8] This clinical description is similar to that found in our case.

The examination of choice is breast ultrasound, which allows evaluation of the adolescent's breast. It does not reveal any pathognomonic sign. The diagnosis can be evoked in front of a well limited, oval, round, lobulated or polylobulated, hypoechoic lesion. Small intracystic echoes without visible microcalcifications and posterior acoustic enhancement can also be observed.[9,10] The presence of areas of heterogeneous echo-structures with cystic anechoic zones suggests the existence of hemorrhagic or necrotic zones.[11] The ultrasound performed in our patient did indeed find the above-mentioned characteristics.

On anathomopathological study, it is a fibroepithelial proliferation with many histological similarities with the low grade phyllodes tumor.[9] Both are fibroepithelial tumors with significant stromal cellularity. The cellularity remains higher in the phyllodes tumor, which allows to differentiate between these two histological types, according to Scolyer et al.[12,13,14] The stroma is of uniform cellular density, which varies from little acellular, to a high cellular density also called 'cellular fibroadenoma'. Whether intraductal or peri-ductal, the clinical description is unchanged. It should be noted that the intraductal form is the most likely to pose diagnostic difficulties.[15]

However, unlike the phyllodes tumor, this form of cellular adenofibroma does not recur and is not malignant in adolescents. According to the study of Bouhafa's team carried out in 2009 in Morocco, comparing the phyllodes tumor and the giant adenofibroma, the latter differs from the phyllodes tumor by the presence of a true capsule and a more harmonious distribution of the stroma and the epithelium.[16,17]

Medical treatment with progesterone and danazol has been tested, attempting to counteract the antiestrogenic effect involved in the pathophysiology of adenofibroma, without satisfactory results and this therapeutic strategy has never been evaluated in this context.[16]

Surgical treatment is still the preferred option and is based on lumpectomy with immediate breast reconstruction, which may be difficult given the large size of the tumor, but it does restore symmetry and comfort to the patient.[18]

In the literature, there is a difference of opinion regarding the surgical treatment of giant fibroadenomas of the breast. McDonald and Harrington in 1950 stressed that these breast tumors are potentially malignant and should be considered until proven otherwise. Therefore the recommended surgical treatment was a simple mastectomy.

Hines and Guerkink noted that these rare adolescent breast tumors can create a management problem, and the rapid growth, distended veins, and varied histologic findings often led to diagnostic difficulties, and in many cases, surgical interventions were unnecessarily radical.

Ashikari et al distinguished between the more common adenofibroma and the variant of this type, namely juvenile giant fibroadenoma, suggested a curved incision at the lateral or inferior border of the breast, and this should avoid mastectomy even if the tumor is large.[19]

According to Nambiar R and Yamamoto Y, The most common approach is excision and mastopexy. Reported approaches include peri-areolar, infra mammary (Gaillard-Thomas) and reverse "T" incisions. The peri-areolar incision is simple and leaves the least visible scar, but it is associated with loss of nipple sensation.

All the classic techniques of breast reduction surgery are applicable in the adolescent, each with its disadvantages and advantages.[20,21,22]

Fibroadenoma may recur after surgical removal, hence the need for prolonged surveillance. Tumor recurrence has been described in the literature.[23,24,25] However, no

recurrence has been noted during the period of surveillance.

The other form of giant fibroadenoma is similar to the adult form. It occurs in older adolescent girls or young women. The already pre-existing nodule progressively enlarges.

This form is more frequent in the Caucasian population and often associates several fibroadenomas. A family history of benign breast disease is more common. Histologically, this fibroepithelial tumor has a low stromal cellularity.

CONCLUSION

Juvenile giant adenofibroma is a particular form of fibroadenoma, defined by a size greater than 5 cm. Its pathophysiological mechanism is poorly understood, and it is thought to be the consequence of an inadequate local response to estrogenic stimulation. It represents 2 to 4% of fibroadenomas. It increases in size rapidly and can reach a diameter of 10 to 15 cm. It is mostly unilateral, unique and its appearance can sometimes be worrying with inflammatory skin and dilatation of the venous network, due to its large size and rapid growth. The diagnosis is clinical and ultrasound, the treatment is surgical based on a lumpectomy with generally good results. The main differential diagnosis of juvenile giant adenofibroma is phyllodes tumor, the clinical presentation is not different from that of giant adenofibroma, only the histology allows the diagnosis.

FIGURES

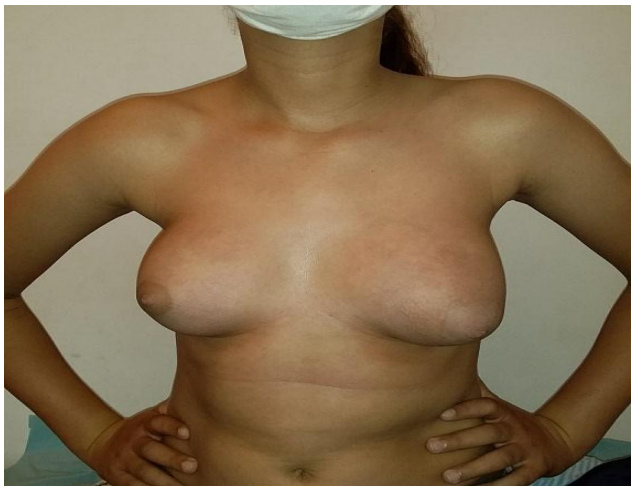

FIG 1

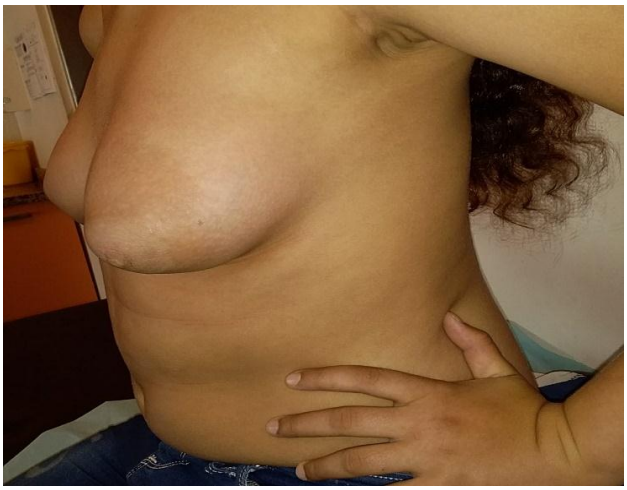

FIG 2

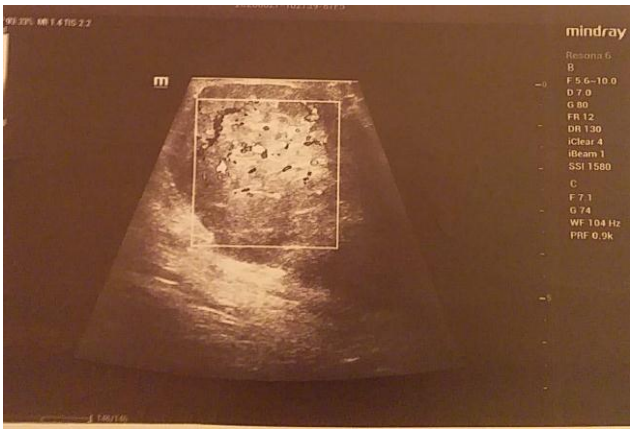

FIG 3

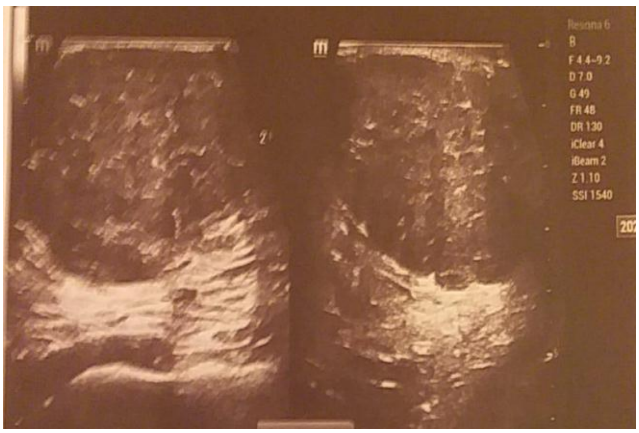

FIG 4

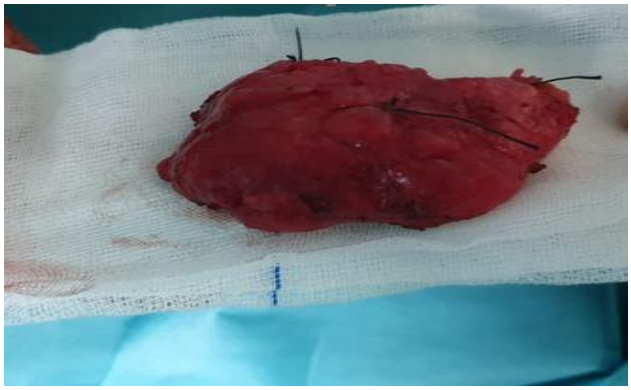

FIG 5

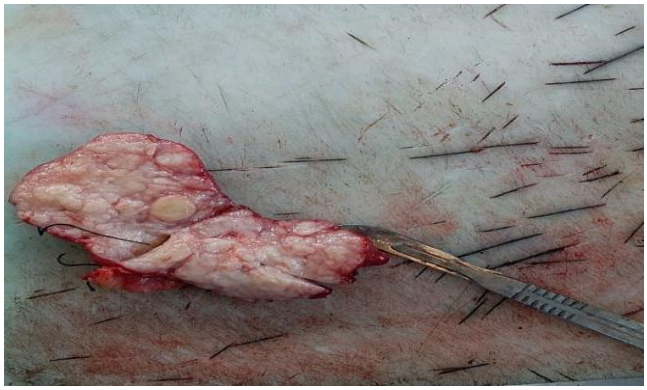

FIG 6

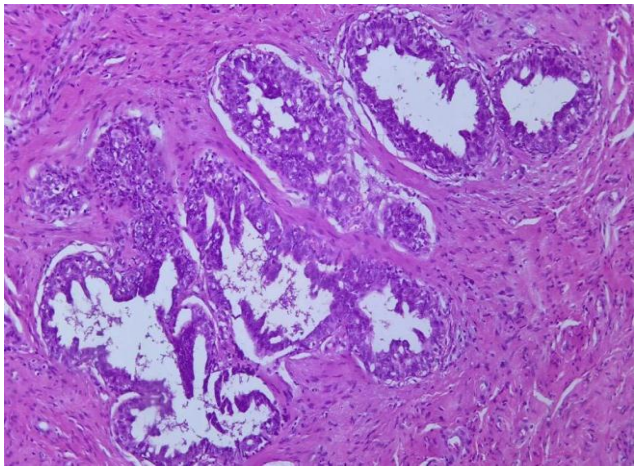

FIG 7

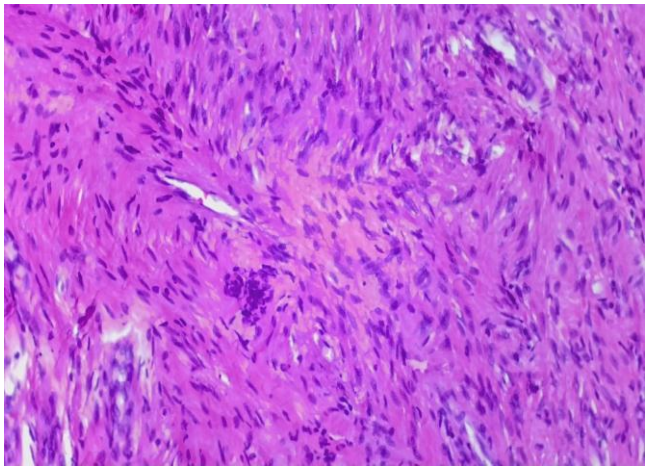

FIG 8

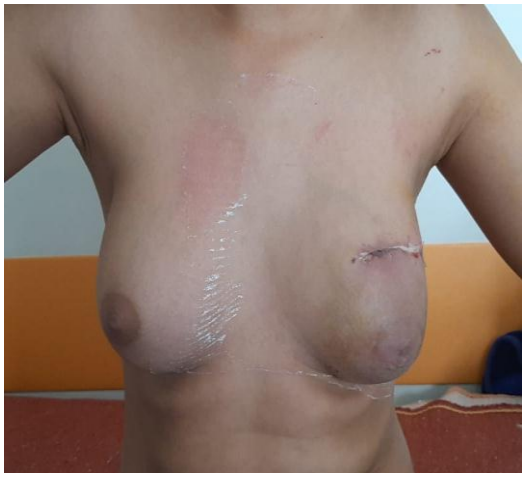

Fig 9

Fig 1 and 2: clinical examination.
Fig 3 and 4: Ultrasound images in favor of a giant fibroadenoma.
Fig 5: Oriented lumpectomy
Fig 6: cut lumpectomy.
Fig 7: Juvenile adenofibroma: epithelial component
Fig 8: Juvenile adenofibroma: stromal component with moderate cellularity without atypia.
Fig 9: D3 Post-op

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