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Case Report

Giant adolescent tubular adenoma of the breast mimicking giant fibroadenoma: a case report

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ABSTRACT

Tubular adenomas are a rare benign breast neoplasm, often clinically indistinguishable from fibroadenomas or phyllodes tumors. We present a case of the largest adolescent tubular adenoma reported in the literature, measuring 12.5 cm. Radiological evaluation and histopathological assessment of the core biopsy sample was consistent with a giant fibroadenoma. However, the final histopathological analysis of the excisional biopsy sample confirmed a tubular adenoma. This case highlights the clinical conundrum of reaching the definitive diagnosis in a patient with a tubular adenoma. A contemporary review of the clinical features, radiology, histopathology and management options for tubular adenoma of the breast was undertaken.

Keywords: Breast, Tubular adenoma, Adolescent

INTRODUCTION

Tubular adenoma is a rare breast pathology, constituting 0.13 percentages to 1.7 percentages of benign breast tumors.[1] Over the past decades, the literature has described only approximately 100 cases of tubular adenoma, finding that tumor size typically ranged between 1 to 10 cm and very rarely exceeded 5 cm.[2,3] With the rarity of the pathology comes diagnostic dilemmas, especially due to the almost identical clinical features when compared with a fibroadenoma or phyllodes tumor.

We present a case of a giant tubular adenoma, the largest tumor size to be reported to our knowledge, especially in the adolescent age group. We provide a contemporary review on the clinical features, radiology, pathology and management of tubular adenomas.

CASE REPORT

A 14-year-old female presented with a 12-month history of a progressively palpable lump of the left breast with bluish discoloration over the large mass. She denied tenderness or nipple discharge. She had no significant past medical history and no previous history of breast-related investigations. There was no known family history of breast cancer. She experienced menarche at the age of 11 years old and had a regular menstrual cycle. There was no contraceptive use.

On examination, marked asymmetry was noted with her left breast being markedly expanded by a large firm mass in the upper and central aspect of her left breast which spanned at least 12 cm. The left areola was inferiorly displaced. Venous distention was noted in the skin over the left breast, giving a blue appearance (Figure 1).

Figure 1: Preoperative clinical photo demonstrating marked enlargement left breast.

She underwent an ultrasound of both breasts and axillae, which revealed normal right breast tissue with a heterogeneous mammary layer throughout, consistent with the patient's age. The left breast was markedly increased in volume due to a gently lobulated, but well-defined hypoechoic mass centro-superior to the nipple line, displacing normal breast tissue inferiorly and toward the upper outer quadrant. The lesion was encompassed by thin septae and contained occasional sonographic clefts. The mass measured 11.9×5.3×12.0 cm sonographically (Figure 2.) Left axillary lymph nodes showed mild reactive changes only. From the ultrasound imaging, the appearances were most consistent with a giant fibroadenoma of adolescence, with the differential of a phyllodes tumor.

A core biopsy was performed, which revealed closely packed small glands surrounded by myoepithelial cells in a background of fibrotic stroma, consistent with a fibroadenoma exhibiting pericanalicular architecture. There was no epithelial hyperplasia, cytologic atypia or malignancy seen. The stroma showed no identified overgrowth, cytologic atypia or mitotic activity. Features on core biopsy were consistent with juvenile fibroadenoma formation, in concordance with imaging features.

Due to the large asymmetric size of the lesion and its somewhat indeterminate nature, complete excision of the mass was undertaken via a laterally placed incision over the left breast. The lesion itself appeared at the time to be well demarcated and was able to be removed with macroscopically clear margins (Figure 3).

Surgery proceeded uneventfully; upon closure an excellent cosmetic outcome was achieved, and the patient made an uncomplicated recovery.

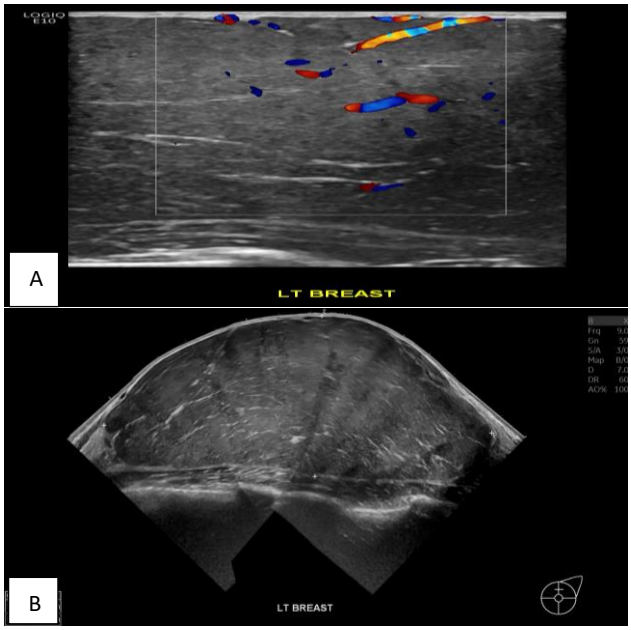

Figure 2 (A and B): Ultrasound left breast showing large well defined mass lesion occupying the centro-superior aspect of the left breast.

Figure 3: Left breast tumour after complete excision.

Histopathological assessment revealed closely packed tubules with sparse intervening stroma, with overall features consistent with a tubular adenoma (Figure 4).

The pathologic size of the lesion was measured at 125×115×70 mm. There was no evidence of atypical epithelial hyperplasia, in situ or invasive malignancy.

On follow-up assessment 12 months after surgical excision, a repeat ultrasound examination demonstrated benign appearing glandular tissue in both breasts with no evidence of recurrence of the tubular adenoma on the left side.

The surgical defect in the left breast was noted to have been largely filled in by developing breast tissue.

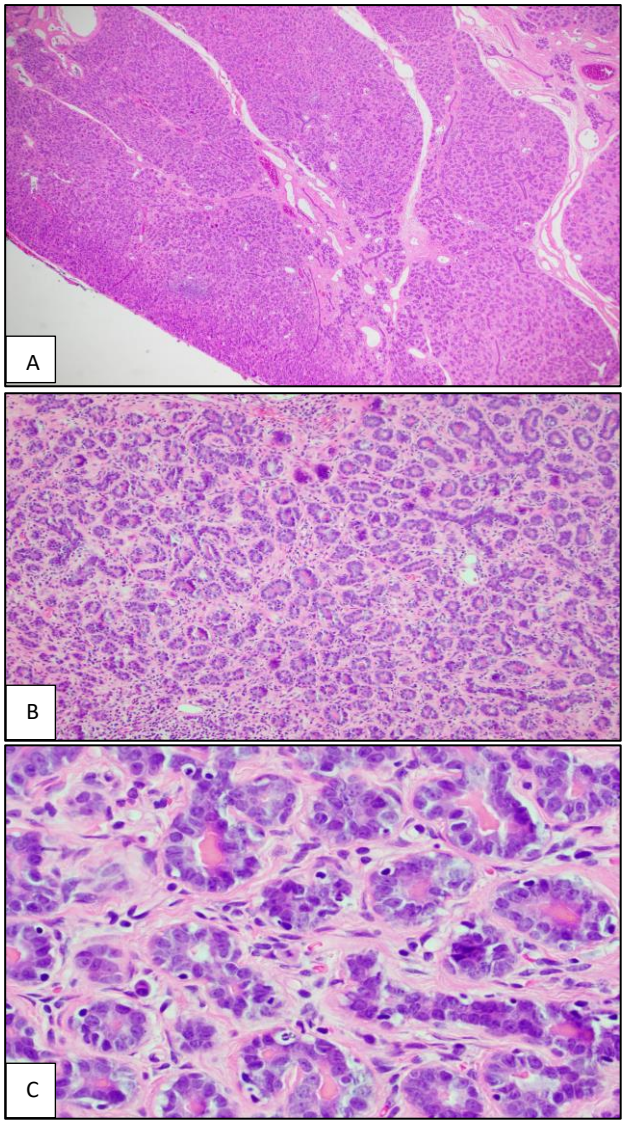

Figure 4 (A-C): Well-circumscribed, cellular lesion comprising lobules of closely packed small acinar structures (×20 magnification). Individual lobules are composed of closely packed rounded acini with only a small amount of intervening stroma (×40 magnification). Small, round and regular acinar structures exhibiting prominent bimodal pattern with peripheral myopethilelial cells and central luminal epithelial cells and minimal intervening stroma (×400 magnification).

DISCUSSION

Clinical features

Tubular adenomas present in women of childbearing age, with very rare occurrences before menarche or after menopause.[1] The patient in this case had experienced menarche at age 11 years, only 3 years prior to her presentation. The cardinal symptom is usually a mobile, well-circumcised palpable breast mass. Our patient presented with a gross mass which occupied virtually her whole breast. In a study exploring 33 cases of tubular adenoma, the size of the lesion was more than 5 cm in only six cases, with the largest being 8.5 cm.[4] The literature has reported tubular adenomas with sizes up to 10 cm.[3] This case presents the largest reported tubular adenoma, especially in an adolescent patient, with a size of 12.5 cm. Tubular adenoma is not associated with axillary lymphadenopathy. Clinically, it is often indistinguishable from other differential diagnoses such as a giant fibroadenoma or phyllodes tumor.

Radiology

Mammographically, the appearance of tubular adenomas is variable; they can appear as masses with microlobulated margins, or as grouped heterogeneous microcalcifications with or without the mass.[5] Sonographically, tubular adenomas appear as well-circumscribed, hypoechoic oval masses with variable vascularity. On magnetic resonance imaging (MRI), tubular adenomas appear as T2-hyperintense, heterogeneously enhancing masses. Given these findings, tubular adenomas are almost indistinguishable from fibroadenomas. Sengupta et al investigated 33 cases of tubular adenoma, comparing clinical, radiological and histological correlation.[4] Clinical and radiological evaluation was only marginally helpful in stratifying whether the lesion was likely to be benign or malignant, and all cases required pathological evaluation to reach a diagnosis.

Histopathology

Tubular adenomas are characterized by the presence of densely packed regular round tubules forming a circumscribed mass, which although being sharply demarcated, does not have a true capsule. These unencapsulated tumours usually have only scant intervening stroma surrounding the ducts.[6] This is in contrast to fibroadenomas, which are usually rich in stroma. Confirmation of tubular adenoma by histopathology is the gold standard for diagnosis.

Fine needle aspiration (FNA) cytology of tubular adenomas is not recommended. The main findings of FNA are small clusters of tubular structures with or without acini.[7] Kumar et al compared FNA smears from 10 cases of confirmed fibroadenomas and 6 cases of confirmed tubular adenomas, and found that initial diagnosis of tubular adenoma was made in only one case from FNA cytology.[8] Furthermore, features such as infarction or degeneration can mimic malignancy in FNA smears, which may lead to unnecessary treatment.[9]

Excisional biopsy is recommended to exclude other more sinister diagnoses, or because of large size or failure to regress.[10] In this case, the large size and mass distortion caused by the lesion warranted an excisional biopsy. The case was interesting in that the histopathological examination of the core biopsy, along with radiological

assessment, was consistent with a diagnosis of juvenile giant fibroadenoma. However, when complete excision of the lesion was undertaken, the formal diagnosis of tubular adenoma was able to be achieved.

Malignant transformation

Malignant transformation is very rare, with only a handful of probable cases reported in the literature. Hill and Miller reported on a case of a breast nodule initially diagnosed as an adenoma, with liver metastasis.[11] Re-examination of histopathology showed presence of invasive carcinoma within the tubular adenoma and liver biopsies that were consistent with metastasis of the breast cancer. Saimura et al reported a rare case of DCIS within a tubular adenoma, with indistinct borders between the two lesions.[12] Domoto et al reported colocalization of a ductal carcinoma *in situ* (DCIS) with a tubular adenoma.[13] However, on histopathological assessment, there was a clearly defined boundary between the two lesions. The authors suggested that while the case may be coincidental existence of two tumors within the same vicinity, malignant transformation could not be excluded.

Management

Complete excision provides a definitive diagnosis, and is often undertaken in the context of an indeterminate breast lesion with the differential diagnosis including a potential phyllodes tumour or a giant fibroadenoma. Due to their benign nature and as tubular adenomas are not associated with an increased risk of breast cancer, ongoing screening can simply be in accordance with local health recommendations.

CONCLUSION

Tubular adenomas are a rare benign tumor of the breast. We present the case of the largest tubular adenoma of adolescence to be reported in the literature measuring 12.5 cm. This case highlights the diagnostic dilemma when differentiating between tubular adenomas and other entities such as fibroadenomas or phyllodes tumors. Histopathological examination remains the gold standard of diagnosis.

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