

CASE REPORT

Giant tubular adenoma of the breast in a child female – Case report

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PALABRAS CLAVE

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Abstract Tubular adenoma of the breast is one of the rarest benign tumors and little is known about this entity. Some cases of giant tubular adenoma of breast have been reported, but till the moment the largest lesion described in literature was a case of a 18 cm tubular adenoma in 31-year-old women. In this case report of a 12-year-old patient, we present the largest giant tubular adenoma of the breast ever described. It was an atypical presentation, not only due to the tumor size, but also due to the absence of a clear cleavage plan with the breast tissue in the physical exam and its rapid growth.

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Adenoma tubular gigante de mama en mujer adolescente – caso clínico

Resumen El adenoma tubular de mama es uno de los tumores benignos más raros y poco se conoce sobre esta entidad. Se han reportado algunos casos de adenoma tubular gigante de mama, pero hasta el momento la lesión más grande descrita en la literatura fue un caso de adenoma tubular de 18 cm en una mujer de 31 años.

En este caso clínico de una paciente de 12 años, presentamos el adenoma tubular gigante de mama más grande jamás descrito. Fue una presentación atípica, no solo por el tamaño del tumor, sino también por la ausencia de un plan de clivaje claro con el tejido mamario en el examen físico y su rápido crecimiento.

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Introduction

Tubular adenoma of the breast is an epithelial tumor that represents 0.13 to 1.7% of all breast benign lesions,¹⁻⁴ being one of the rarest benign tumors.¹

Little is known about this tumor, since in the current literature only some case reports or small series have been described.¹⁻⁵

Classically it occurs in young females of reproductive age¹⁻⁴ and is clinically and imagological difficult to distinguish it from fibroadenoma, one of the most common benign breast lesions.^{2,4-6}

It usually presents as a painless well-circumscribed and freely movable breast mass, with slow growing, and without skin or nipple alterations.^{2,3,5} Nevertheless, rarely some suspicious features can be present.^{1,4}

The mean tumor size of tubular adenomas ranges from 1 to 7.5 cm, rarely exceeding 5 cm.^{1-3,6} However, some cases of giant tubular adenoma of breast have been reported, but till the moment the largest lesion described in literature was a case of a 18 cm tubular adenoma in a 31-year-old woman.⁷

The preoperative diagnosis of tubular adenoma is not possible, since the cytological examination isn't reliable, and only the histology can make a definitive diagnosis.¹⁻⁵ For that reason, and to prevent it from growing, surgical excision is recommended when there is any doubt on possible malignancy or the size or the growing is considerable.^{2,5}

Case report

A twelve-year-old girl was admitted to our institution in July 2020 with marked asymmetry of breast development, which became evident since the menarche in January 2020. She was a healthy girl, with no smoke habits or regular medication intake and with no relevant family history. She previously has had a non-presential appointment with her family doctor which reassured her, telling her that some asymmetry is normal during the breast development.

On physical examination, the left breast was significantly enlarged (Fig. 1), with a palpable 15 cm solid mass, without clear cleavage plan with the breast tissue.

On ultrasound, left breast was completely occupied by a large solid mass, not measurable by ultrasound. A needle biopsy was performed, which revealed the presence of filiform fragments with fibrous stroma, CD34+, rich in vascular clefts and lobe hypertrophy, being suggestive of a "Pseudo angiomatous stromal hyperplasia (PASH)".

Clinically, there was an increase in the mass dimensions and surgery was performed in December 2020, with breast tumor excision and left reduction mammoplasty, with the collaboration of plastic surgery. The excised lesion weighted 1016 g and measured 19x17x15cm.

The histological exam revealed a well circumscribed lesion with compact areas of tubular structures with myoepithelial outer cell layer (p63+) and inner layer of epithelial cells (EMA+) and a variable amount of fibrous stroma separating those areas (Fig. 2). Those aspects were compatible with the definitive diagnosis of a tubular adenoma of the breast.

The recovery period after surgery went well, and the surgical scars had a good evolution. The patient did a follow-up ultrasound in May 2021 which revealed only a cicatricial residual hypoechoic area measuring 9 mm in the lower inner quadrant of the left breast, without new nodular lesions.

Discussion

The presented case corresponds to the largest tubular adenoma of the breast described in the literature to our knowledge till the moment.

The clinical presentation in this young girl wasn't the typical one due to the tumor size, the absence of a clear cleavage plan with the breast tissue in the physical exam and its rapid growth.

Despite the impossibility of a definitive diagnosis based on radiological assessment,^{3,4} the usual radiologic features of tubular adenomas are well-circumscribed lesions without calcifications, resembling fibroadenomas.^{1,2,4} However, in some cases, radiology or even cytology, fails in excluding possible malignant nature of the lesions.⁴ In this case, the ultrasound wasn't helpful due to the lesion dimensions and the biopsy, despite being suggestive of a benign lesion, wasn't concordant with the posterior definitive diagnosis.



Fig. 1 Breast asymmetry.

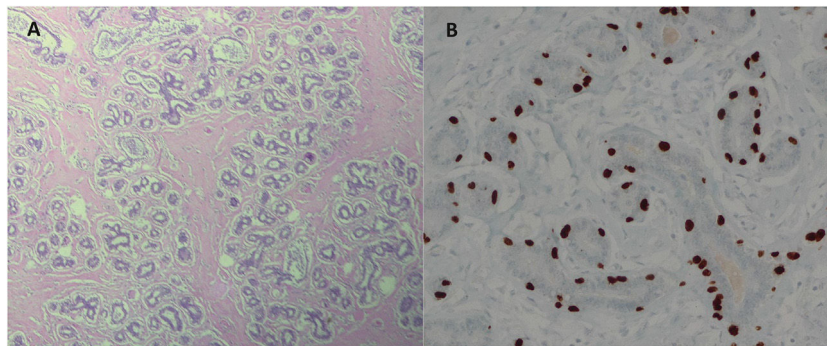


Fig. 2 Photomicrograph of histopathology, stained with HE (A) and immunohistochemistry for p63 (B).

Preoperative diagnosis of tubular adenoma is impossible in most cases and surgical excision is necessary to obtain a definitive diagnosis^{2,4} since histopathology remains the gold standard for final confirmation.³

Histologically, tubular adenoma is characterized by the occurrence of tightly packed homogenous tubular and acinar epithelial components with scant stroma surrounding the ducts, in contrast with fibroadenoma which contains large amount of stroma.² The round tubules are composed by two cell layers: luminal epithelial cells and abluminal myoepithelial cells.⁵

Immunohistochemical analysis confirms the presence of myoepithelial cells, by the identification of myoepithelial markers such as p63 protein, SMA, cytokeratin 14 (CK14), CK5/6 and calponin. Detection of vimentin or CD34 highlights the stromal component of the tubular adenoma.¹

The differential diagnosis of tubular adenoma includes fibroadenoma, nipple adenoma, lactating adenoma, sclerosing adenosis, microglandular adenosis, tubular adenosis, ductal adenoma and tubular carcinoma.²⁻⁶

The use of myoepithelial and stromal cell markers allows in ruling out the hypothesis of breast carcinoma and other benign breast lesions that can be associated with high cancer risk.¹

Lactating adenoma can only be differentiated from tubular adenoma by history data. These neoplasms also show lactational changes in the lining epithelium of glands.⁴

According to the available literature, tubular adenoma is a benign entity, with no reported incidence of recurrence or increased risk of malignancy.^{1,3-5} Nevertheless, we cannot ignore that there are cases described of coexistence of tubular adenomas and carcinoma of the breast.

To conclude, despite being a benign entity, in some rare cases tubular adenoma of the breast can grow to considerable dimensions and alarm both patients and physicians. Surgery should be performed for a correct definitive diagnosis.

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Ethical considerations

The authors declare that they have followed the protocols of their center on the publication of patient data.

Patient consent

The patient gave her informed consent.

Declaration of Competing Interest

The authors declare that they have no conflict of interests.

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