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Case Report

A rare cause of unilateral breast swelling in a male infant caused by fibrous hamartoma of infancy combined with pseudoangiomatous stromal hyperplasia

Jan Jonckheere, MD, Marian Vanhoeij, MD, Ieva Garkalne, MD, Marijana Antic, MD, Ann Schiettecatte, MD, Johan de Mey, PhD*

Department of Radiology, University Hospital of Brussels, Laarbeeklaan 101, 1090 Brussels, Belgium

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ABSTRACT

We report a case of a male infant who underwent resection of a unilateral breast mass with a histopathological diagnosis of a fibrous hamartoma of infancy (FHI) combined with a pseudoangiomatous stromal hyperplasia (PASH). Breast lumps are uncommon in infants and children, especially in boys. FHI and PASH are very rare causes of breast lumps, especially in infants. To our knowledge, this is the first report of a combination of both pathologies in 1 lesion in the breast of an infant.

Case report

A mother presented with her 14-month-old son for a pronounced left unilateral swelling in the breast. The swelling was first noticed 4 months ago with a rapid increase in growth since 1 month. Palpation revealed a solid retroareolar mass with stretching and protrusion of the areola (Fig. 1).

Ultrasound showed a solitary well-circumscribed heterogeneous hypoechoic mass with a maximal diameter of 3.2 cm behind the left areola (Fig. 2). Color Doppler was not contributive due to agitation of the child with too much movement. No axillary adenopathies were found.

An additional MRI was performed to assess the extent of disease and confirmed a solitary well-circumscribed mass with a larger maximal diameter of 4.5 cm compared to 3.2 cm on ultrasound (Fig. 3). There was a thin hyperintense line between the lesion and the pectoral muscle on T1- and T2-weighted images compatible with fatty tissue, thus no infiltration of the underlying pectoral muscle. The lesion was homogeneously isointense on axial T1 and axial and sagittal T2 images compared to the muscular structures.

The child underwent surgery because of a significant deformation of the breast, with local excision of the lesion and nipple preservation (Fig. 4).

Histopathology revealed the diagnosis of a fibrous hamartoma of infancy in one part of the lesion combined with a

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* Corresponding author.
E-mail addresses: janjonckheere@hotmail.com, jan.jonckheere@uzbrussel.be (J. Jonckheere).

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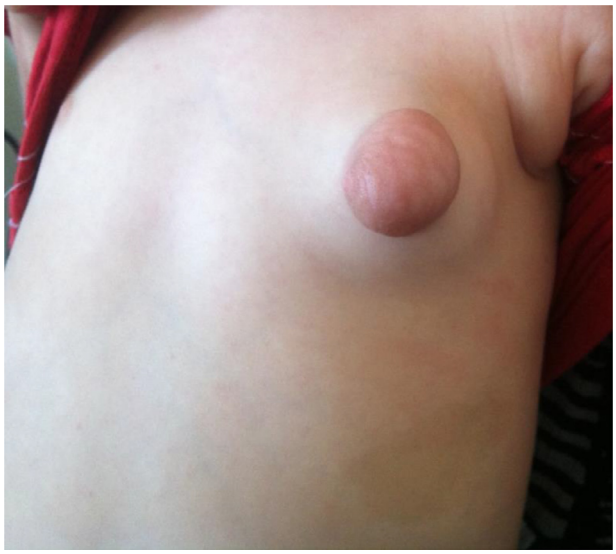

Fig. 1 – Solid left retroareolar mass with stretching and protrusion of the areola.

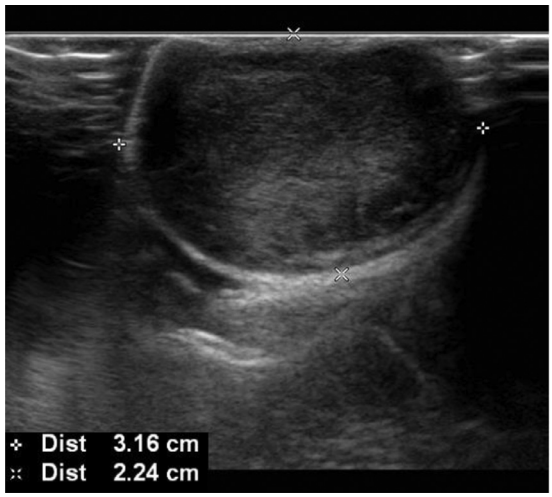

Fig. 2 – Ultrasound shows a solitary well-circumscribed heterogeneous hypoechoic mass with a maximal diameter of 3.2 cm behind the left areola.

pseudoangiomatous stromal hyperplasia in another part of the lesion.

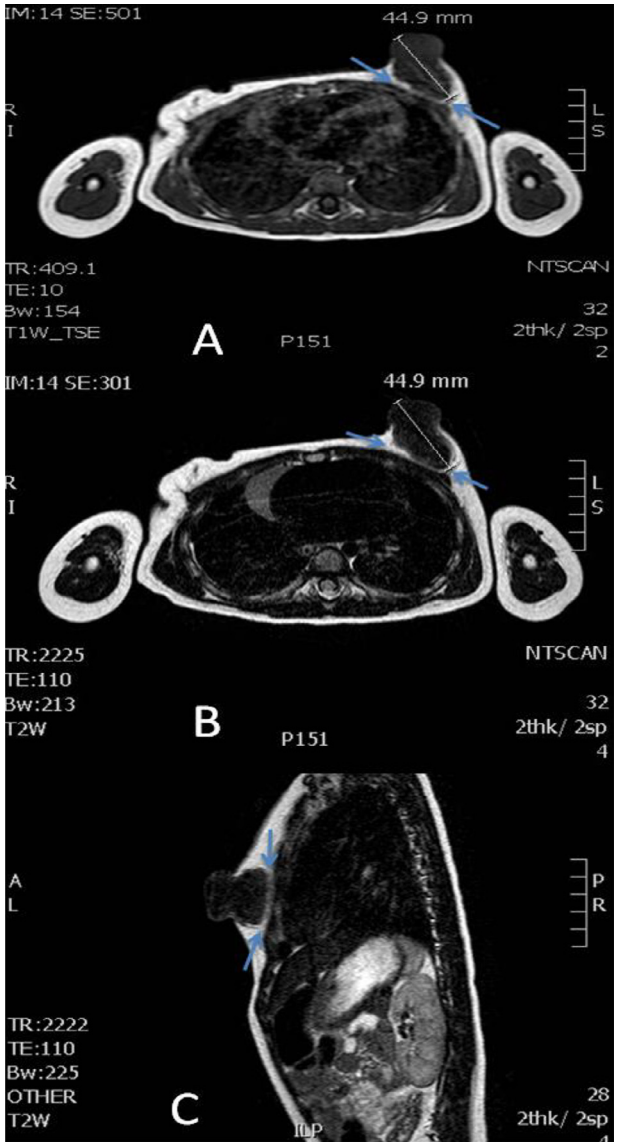

Fig. 3 – MRI with (A) axial T1, (B) axial T2 and (C) sagittal T2 sequences shows a solitary well-circumscribed lesion of 4.5 cm in the left breast, isointense compared to the muscular structures. without invasion of the pectoral muscle. A thin hyperintense line between the lesion and the pectoral muscle on T1- and T2-weighted images (blue arrows) corresponds to fatty tissue, thus no infiltration of the underlying pectoral muscle. (Color version of figure is available online.)

Discussion

Pediatric breast lesions like fibrous hamartoma of infancy (FHI) and pseudoangiomatous stromal hyperplasia (PASH) are very rare, especially in male infants, and are mostly benign. However, these lesions can often be alarming to medical staff, patients, and their family. Therefore, it is important to be aware of the existence of these lesions and their diagnostical work-out. Key factors that aid in diagnosis include the length of time that the mass has been present, associated pain or other symptoms, whether the mass affects 1 breast or both, how rapidly the mass is growing, and family history of breast disease [1].

In contrast to adults, mammography is not indicated in children because of the extremely low risk of breast cancer and the increased risk of radiation-induced malignant changes in the young glandular breast [2,3].

Ultrasound is the primary imaging modality used in young patients, aiding in the initial diagnosis, assisting in imaging-guided biopsy when indicated, and offering a safe method of

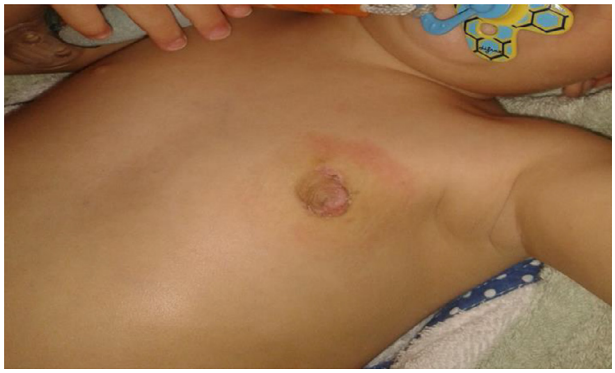

Fig. 4 – Postoperative result after local excision with nipple preservation.

follow-up. MRI in the pediatric patient of the breast is rarely used, though in select cases it may be useful for assessing the extent of disease and surgical planning [4].

FHI is a benign, uncommon fibroproliferative lesion of the dermal and subcutaneous layer occurring in infancy and childhood, especially in boys. The tumor is most frequently found in the axilla, followed by shoulder, inguinal area, and chest wall. The breast has also been described as a rare location for this tumor [5].

Histology shows triphasic elements of mature fibrous tissue, mature adipose tissue, and immature mesenchymal tissue [6].

At ultrasound, FHI is most often a well-circumscribed or lobulated mass and heterogeneously hyperechoic, with a superficial location. A "serpentine pattern" could be present by intervening hypoechoic portions in the hyperechoic mass. The hyperechoic portions can represent fat component and the hypoechoic portions can be the fibrous component on pathology [7].

PASH is a benign, uncommon form of mesenchymal overgrowth within the breast tissue that derives from a possible hormonal aetiology, occurring very rarely in infants [8].

Histology shows stromal proliferation with anastomosing slits mimicking vascular spaces [9].

At ultrasound, PASH is most often a well-circumscribed oval or round mass, often hypoechoic and sometimes slightly heterogeneous. Imaging appearance may be similar to that of fibroadenoma [10].

In this case, there was a pathologically proven combination of FHI in one part of the lesion and PASH in another part of the lesion.

A combination of both pathologies in 1 lesion of the breast in an infant has to our knowledge not been described in the literature and therefore specific ultrasound features are not known. However in this case, ultrasound showed a well-circumscribed heterogeneous mass and these features seem to correlate with the benign ultrasound features of both separate entities.

MRI was performed to evaluate the extent of the disease and to plan the type of surgical excision. There was an underestimation of the total diameter of the lesion on ultrasound (3.2 cm) compared to MRI (4.5 cm) in this case.

FHI and PASH are benign and they are neither associated with malignancy nor considered to be premalignant lesions.

Generally, a conservative approach with imaging follow-up is sufficient for these benign lesions [4]. In this case however, local excision was the treatment of choice because of the severe breast deformity.

In conclusion, breast lesions are very rare in children and are mostly benign. It is important to be aware of these benign lesions as they can often be alarming to medical staff, patients and their family. Ultrasound is the first modality of choice in these young patients and MRI can be used for assessing lesion extension. The combination of both FHI and PASH in 1 lesion of an infant breast has to our knowledge never been described before.

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