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Case Report

Ectopic breast tissue presenting as an enlarging abdominal mass

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ABSTRACT

Ectopic breast tissue (EBT) is an uncommon entity that occurs in about 6% of the population, more frequently in Asian people. It manifests as a nonspecific soft tissue mass that can develop in any location along the “milk-line,” with the axilla being the most frequently reported location. As with normal breast tissue, both benign and malignant processes can arise from EBT. Therefore, imaging plays an important role in the adequate assessment of these soft tissue lesions, characterizing its shape, borders, internal components, and vascularity, as well as its relationship with the deep fascia and adjacent structures. Here we present a case of a 33-year-old female Indian patient with an 8 cm soft tissue mass in the upper left abdominal quadrant with a recent increase in size. Clinical and imagiological evaluation were nonspecific but established the nonaggressive behavior of the lesion. To ultimately exclude malignancy, an ultrasound-guided biopsy was performed providing the histological diagnosis of EBT. This case report illustrates the value of clinical, radiological, and histological correlation in the approach of indeterminate soft-tissue tumors, with special focus on lesions in close proximity to the native breasts, where EBT should not be forgotten amongst differentials.

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Introduction

Ectopic breast tissue (EBT) occurs in about 0.4% to 6% of the population [1–3], with the axilla being the most common anatomical location [1,2,4]. It usually results from an involutional failure of the embryological mammary ridge, also

known as the “milk line,” which extends from the axilla down to the groin, leading to possibility of its occurrence in any location along this track [1–4]. EBT usually presents as a soft tissue subcutaneous mass, ranging from nonpalpable to palpable and tender, that can slowly grow [1,4,6,7]. The nonspecific presentation often leads to patient’s concern and diagnostic challenges. Imaging modalities allow not only an accurate measurement and localization of the lesion, but also provide crucial information regarding its nature, such as lack of aggressive/infiltrative features and its relationship with the adjacent tissues and deep fascia [1,4,7]. We present a case of a

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Fig. 1 – Ultrasound image showing a well-defined soft tissue mass located in the upper left anterior abdominal wall of a 30-year-old female Indian patient. The lesion demonstrated a lobulated contour and a heterogeneous solid internal component, predominantly hypoechoic.



Fig. 2 – Ultrasound image with Doppler interrogation demonstrating a soft tissue mass with areas of internal vascularity.

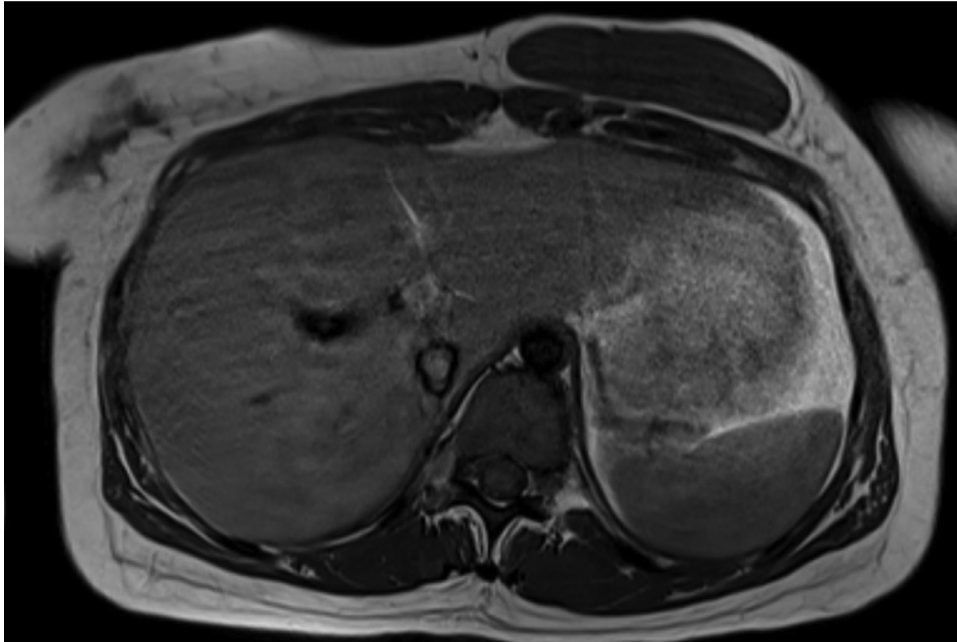


Fig. 3 - Axial T1 image demonstrating a well-circumscribed, mildly lobulated subcutaneous lesion, at the level of the upper inner abdominal quadrant. The lesion appeared predominantly of low signal with some foci of hyperintense areas, particularly on the deep outer portion. There was no evidence of deep tissue invasion, with a clear fat plane standing in between the lesion and the muscle compartment.

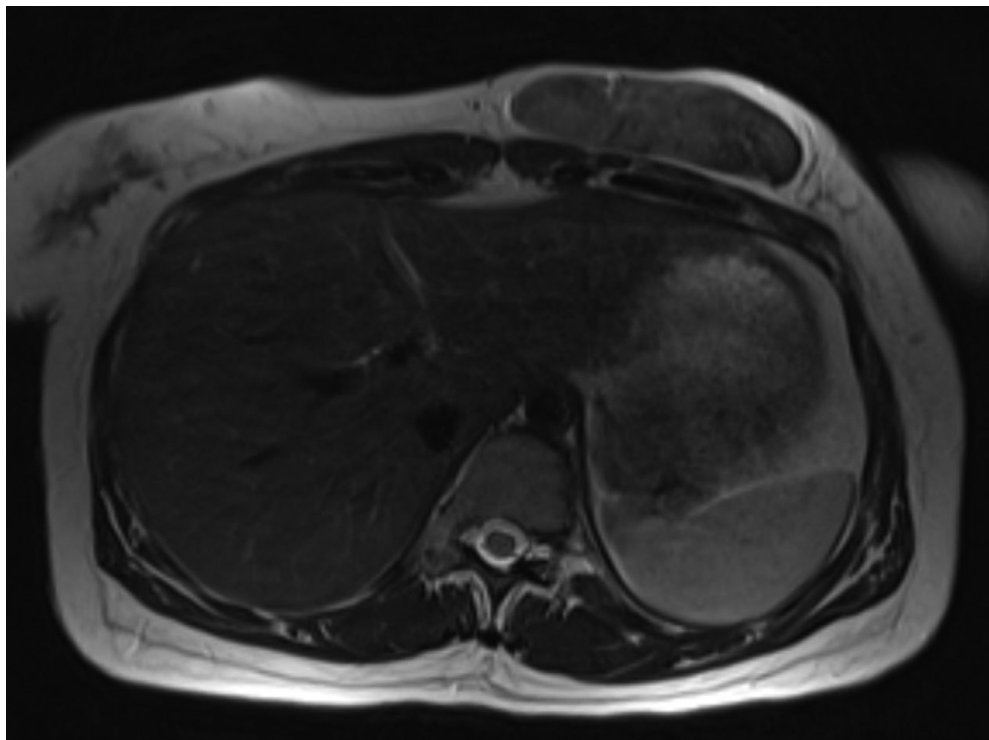


Fig. 4 - Axial T2 image depicting a heterogeneous lesion with intermediate-to-high signal when compared to the adjacent muscle and fat. Some high signal strands were also noted within the mass.

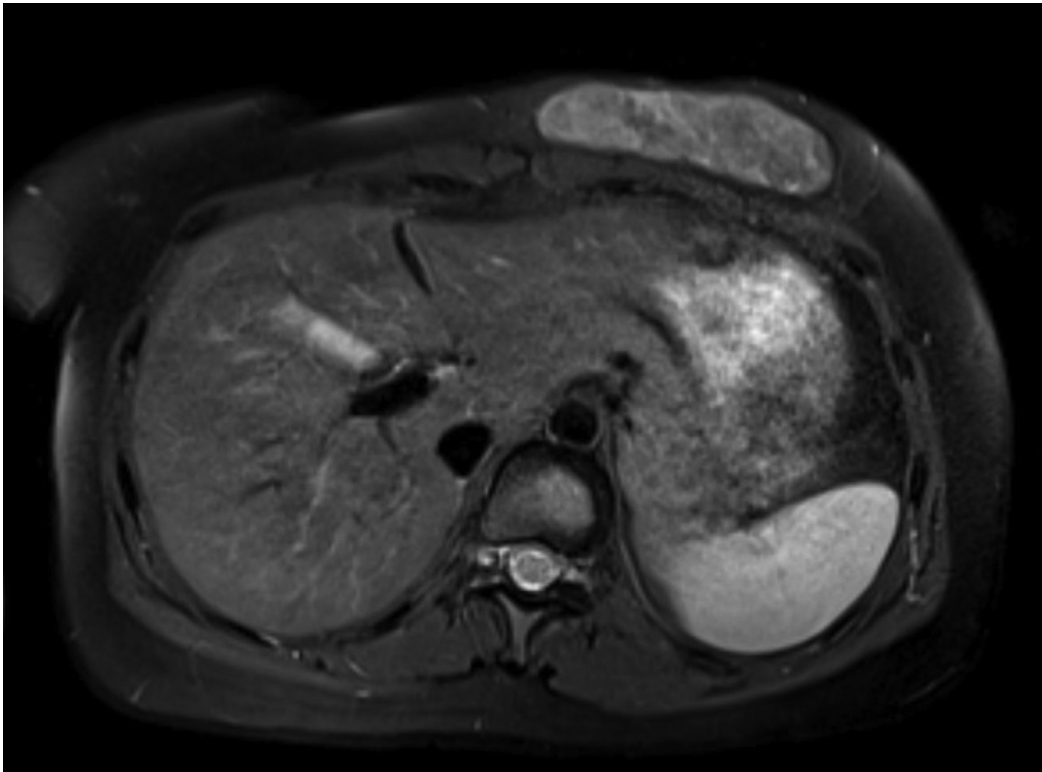


Fig. 5 – Axial Proton Density (PD) image with fat saturation confirming the heterogeneity of the lesion, with a predominantly high signal, but with some interposed components of low intensity.

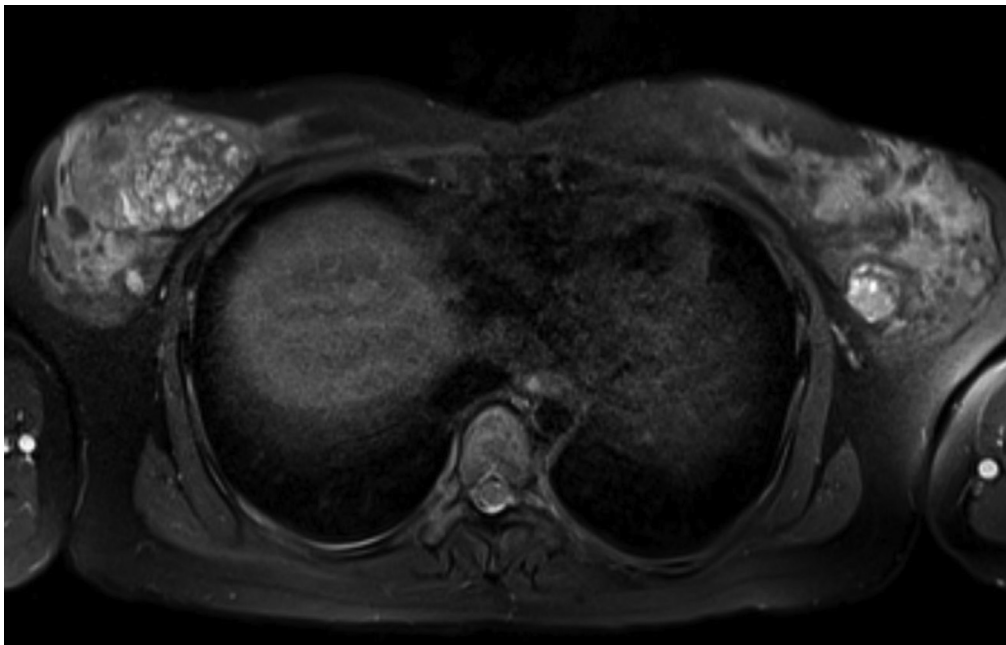


Fig. 6a – Axial and sagittal PD sequence with fat saturation demonstrating the presence of multiple bilateral breast heterogeneous masses, in keeping with fibroadenomas.

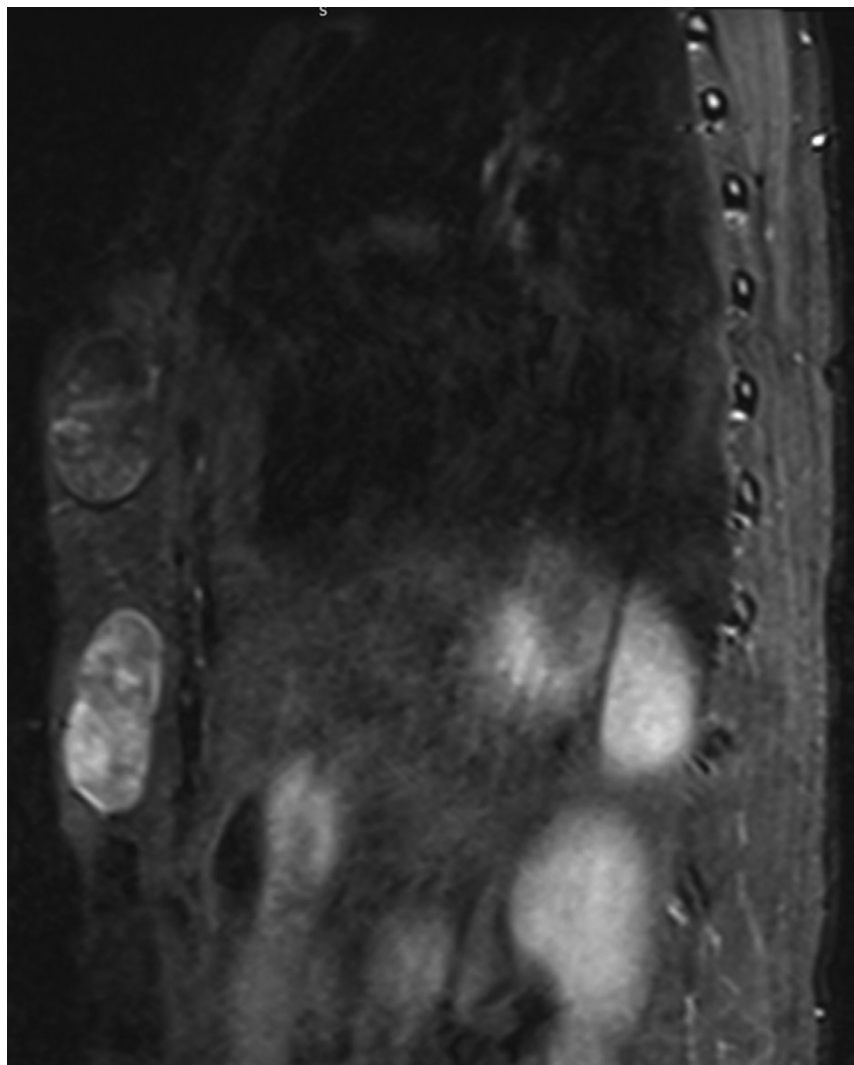


Fig. 6b – Axial and sagittal PD sequence with fat saturation demonstrating the presence of multiple bilateral breast heterogeneous masses, in keeping with fibroadenomas.

33-year-old patient with a large and slow growing mass in the upper abdominal wall histologically proven to be EBT.

Case report

A 33-year-old Indian female patient presented with a slowly enlarging mass in the upper left abdominal wall. Though the lesion had been present for at least 5 years, the patient reported perception of recent enlargement. There was no relevant clinical or family history. Physical examination revealed a 5 cm mobile, nontender, mild-to-moderate compressible mass located in the left upper abdominal quadrant. Laboratory results were unremarkable.

An ultrasound and magnetic resonance imaging (MRI) were requested. Ultrasound demonstrated a well-defined subcutaneous solid lesion, measuring $7.9 \times 1.9 \times 5.1$ cm, of slightly heterogeneous echotexture, predominantly hypoechoic to the surrounding fat, with no evidence of deep fascia or muscle in-

vasion (Fig. 1). Power Doppler evaluation depicted some internal vascularity (Fig. 2). On MRI, the lesion was well defined, with circumscribed mildly lobulated margins, located in the subcutaneous tissue, below the left inframammary fold, not extending through the deep fascia or infiltrating to the adjacent structures. The mass presented a predominately low T1 signal (Fig. 3), and a heterogeneous intermediate-to-high T2 signal (Figs. 4 and 5). These signal characteristics together with the ultrasound features were not consistent with lipoma. In addition, MRI denoted multiple bilateral breast lesions suggestive of fibroadenomas, which were not far from the lesion (Figs. 6a and 6b). No lymphadenopathy was noted. To further characterize the mass an ultrasound-guided biopsy was performed. The specimen's histopathological analysis revealed the presence of fibrocollagenous tissue, which included clusters of benign glandular epithelium and associated myoepithelial cells arranged in a lobular architecture. Some of the glandular elements showed columnar cell change and epithelial hyperplasia, with areas of focal secretory changes (Fig. 7). There was no evidence of atypia or invasive malignancy. Im-

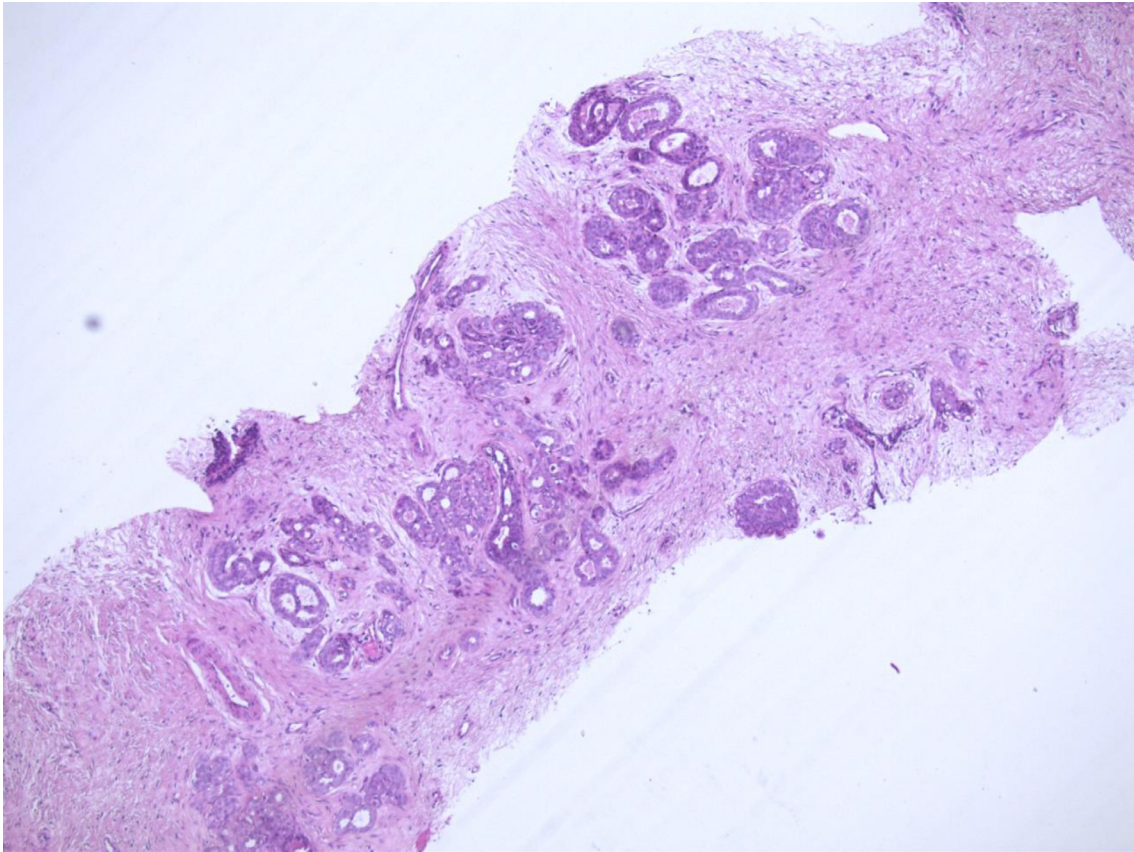


Fig. 7 – Low magnification (x50) microphotograph of a biopsy fragment with H&E staining. Some terminal ductal lobular units with acini can be appreciated surrounded by a fibroconnective tissue. No clear pseudocapsule is seen, neither sheet-like epithelial cells nor abundant fibrous tissue that would be consistent with fibroadenoma.

munohistochemistry demonstrated estrogen receptor positivity (Fig. 8). The findings were then compatible with healthy breast tissue.

Given the anatomical location and the radiologic-pathologic correlation, the diagnosis of ectopic/aberrant breast tissue was made.

Discussion

EBT is an embryological abnormality that occurs around 0.4% to 6% of the global population [1–3,5], with its incidence varying according gender, race, and genetics. It is more common amongst females compared to males (5.19% vs 1.68%) [4], highest in Japanese and lowest in white people [1,4,5]. Most cases are sporadic, but hereditary dominant trait with incomplete penetrance have also been reported (6%) [1,2,7]. The axilla is the most common location followed by vulva [4], but it can also occur anywhere along the thoraco-abdominal region [1,2,6], usually just under the inframammary sulcus, more frequently on the left side of the body [8].

By the 4th to 6th week of the normal embryological development, 2 symmetrical folds of thickened ectoderm form along the ventral side of the embryo, running from the axillary fold to the inguinal region, known as the mammary ridges or

“milk line.” During normal development, these ridges of galactophoric ducts regress with the exception of the pectoral regions [1–3]. Therefore, one of the theories for EBT is a failure in the regression of the mammary ridges, leading to the expected occurrence of EBT anywhere along the track, with the axillary folds being the most common location [1,2,7]. However, some rare cases of EBT outside the milk line have been reported, such as face, ear, neck, dorsal scapula, knee, lateral thigh, and buttock [1–3,6,7]. For this reason, 2 other theories have been suggested, one relating to breast differentiation from modified apocrine sweat glands and other considering its random occurrence due to the migration of primordial breast cell nests outside the milk line [1].

EBT or accessory breast tissue is encompassing and consensual terms, referring not only to supernumerary breasts but also to aberrant breast tissue [1]. The former corresponds to the presence of a breast unit along the milk line, composed by an organized ductal system that communicates with the overlying skin [7]. The latter refers to the presence of breast tissue islands located near normal breasts, but with a non-organized secretory system [1] and no communication with the skin. EBT can present with the same elements as the normal breast (glandular tissue, areola, and nipple) in variable associations [2–4,7], so, even in the absence of a nipple, EBT should not be excluded from the diagnosis [4,5]. While supernumerary nipples are present at birth, EBT maybe only be noticeable

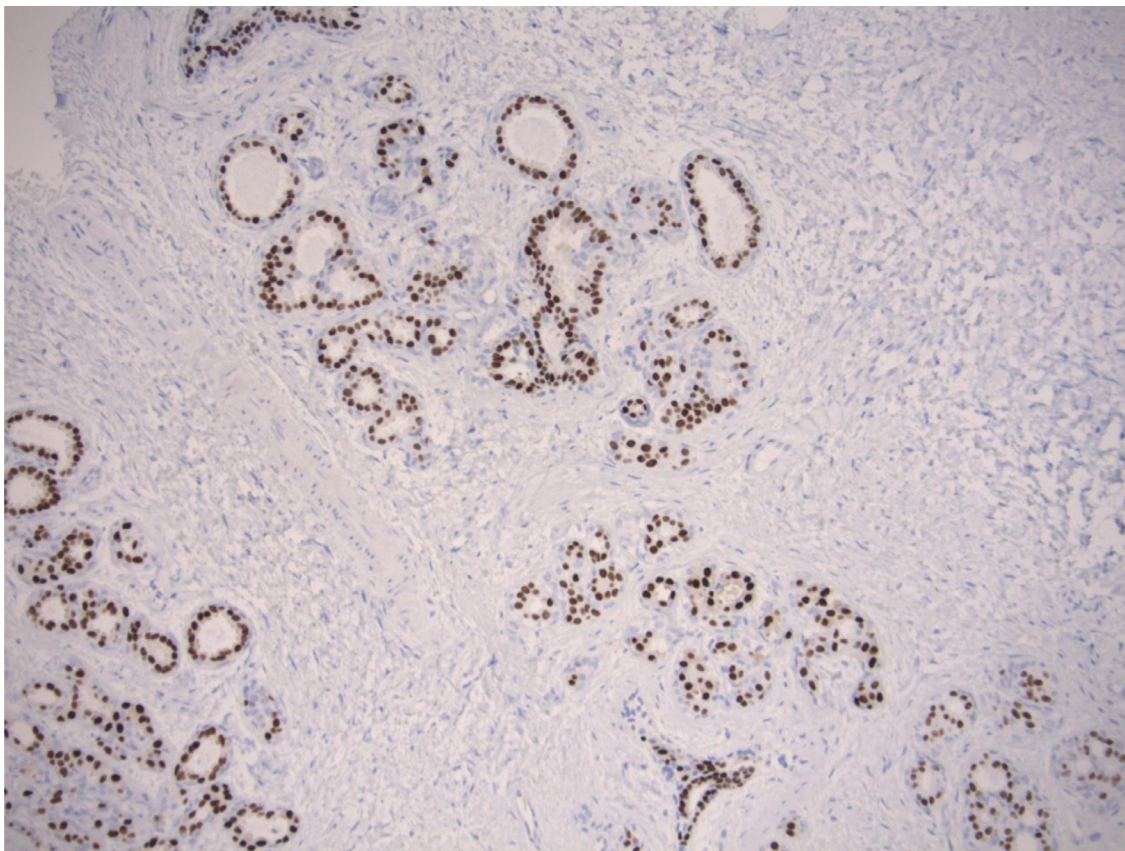


Fig. 8 – Microphotograph of a section stained by immunoperoxidase method using a monoclonal antibody specific for estrogen receptors. Most cells show a strong brown staining of the nuclei in keeping with estrogen receptor positivity.

later in life, since it responds to hormonal stimulation, such as during puberty, pregnancy, and lactation [2–5]. This variability in presentation is partially responsible for the diagnostic challenges of EBT [9].

Supernumerary nipples have an established association with renal abnormalities, hypertrophic pyloric stenosis, cardiac pathologies (congenital heart defects and conduction defects), and urological malignancies (kidneys, bladder, prostate, and testis) [1,3,4]. Skeletal abnormalities such as vertebral anomalies, bifid mandibular condyle polydactyly and clinodactyly have also been reported [2].

EBT behaves like normal breast tissue, which means that the normal spectrum of breast pathology can also occur. This ranges from benign conditions such as mastitis, fibrocystic changes, and fibroadenoma to malignant conditions, such as invasive carcinoma, medullary carcinoma, and more rarely invasive lobular and phyllodes tumor [1,2,7].

Clinical presentation is variable. In some cases, patients are unaware of its presence either because it is nonpalpable or because it lacks accompanying signs and symptoms [1,7]. Periodic tenderness or discomfort over a mobile soft-tissue mass, or even local irritation or restricted movement have also been reported [1,4,6]. Milk secretion or erythema can be accompanying signs [1]. The differential diagnosis includes fibroadenoma, fibroadenolipoma, sebaceous cyst, hidradenitis suppurativa, lymphadenopathy, vascular malformations, and malignancy (lymphoma and metastatic disease) [1,4].

Physical examination should always include assessment of the contralateral side, as EBT can present as bilateral soft tissue masses up to one-third of cases [1,10].

The choice of imaging studies depends on the patient's age and on the presumptive diagnosis raised by the clinician. It usually encompasses an ultrasound evaluation and, if the lesion is axillary, a mammogram should also be performed. Mammography usually depicts a focal asymmetry in the axilla, with fibroglandular densities interspaced by fat, similar to the normal breasts [1,7]. If tissue elongation from the axilla to the native breast is noted then it is referred to as the axillary tail of Spence and not EBT, where both tissues are in discontinuity [1]. Ultrasound is an excellent modality for the initial evaluation of soft tissue masses. It allows measurement, general localization, and vascularity assessment. On ultrasound, EBT resembles the normal breast tissue with evidence of fibroglandular components and fat [1]. MRI appearances are of a subcutaneous lesion with similar signal intensity to the background breast tissue parenchyma, with variable degree of enhancement and variable amount of interposed fat [1,4,7]. MRI provides better soft tissue contrast resolution than any other imaging techniques, which means it is of particular use in the evaluation of nonspecific soft tissue masses, not only portraying the intrinsic signal characteristics of the lesions, but also especially depicting the extension of depth invasion and its

relationship with the adjacent structures. The presented case features a good correlation between ultrasound and MRI imaging findings, with coherent findings, such as measurements, contour shape, and heterogeneous solid component. The shape and relative position of the lesion in respect to the muscle compartment on ultrasound could almost infer a noninvasive behavior of the mass. However, this important radiological criterion is much better delineated by MRI, where a clear fat plane between the inferior border of the lesion and the deep tissues is well appreciated. Furthermore, MRI demonstrated similar signal characteristics between the mass and the native breast tissue.

The histopathological analysis of EBT exhibits the presence of glandular columnar epithelium, frequently hyperplastic, with no evidence of cytological atypia. In this case, the absence of a uniformly distributed sheet-like or stellate epithelial cells in a background of abundant and hyperproliferative hypovascular stroma/mesenchymal tissue on histological study [11] excluded fibroadenoma. Hamartoma was also excluded due to the absence of fat.

Management of asymptomatic EBT, after histological confirmation, is usually conservative [3,7]. Clinical and imaging follow-up assessment of EBT should not be completely dismissed [3], particularly in patients who are being screened for or have history of breast cancer [1,6]. Surgical excision may be appropriate for cosmetic reasons or if there is rapid growth or pain [1–5]. In cases of BRCA 1 or 2 mutations, ablation has also been proposed.

Conclusion

EBT is a rare condition that may pose diagnostic challenges. It should always be considered in imaging studies when a subcutaneous mass demonstrates similar imaging characteristics or proximity to the native breast tissue. This case report provides an adequate sequence of imaging studies when manag-

ing soft tissue masses and highlights the importance of rad-path correlation. Radiologists should be aware of this condition when confronted with this kind of lesions, as raising the possibility of a benign lesion might help avoiding unnecessary patient's distress.

REFERENCES

- [1] Defilippis EM, Arleo EK. The ABCs of accessory breast tissue: basic information every radiologist should know. *Am J Roentgenol* 2014;202(5):1157–62.
- [2] Velanovich V. Ectopic breast tissue, supernumerary breasts, and supernumerary nipples. *Southern Med J* 1995;88(9):903–6.
- [3] Srinivasan A, Bose JC, Govindaraj K, Ramalingam P. Fibroadenoma of ectopic breast tissue - a case report. *Int J Surg Res* 2018;5(1):88–90.
- [4] Laor T, Collins MH, Emery KH, Donnelly LF, Bove KE, Ballard ET. MRI appearance of accessory breast tissue: a diagnostic consideration for an axillary mass in a peripubertal or pubertal girl. *Am J Roentgenol* 2004;183(6):1779–81.
- [5] Patel PP, Ibrahim AM, Zhang J, Nguyen JT, Lin SJ, Lee BT. Accessory breast tissue. *Eplasty* 2012;12:ic5.
- [6] Guray M, Sahin AA. Benign breast diseases: classification, diagnosis and management. *Oncologist* 2006;11(5):425–49.
- [7] Lim HS, Kim SJ, Baek JM, Kim JW, Shin SS, Seon HJ, et al. Sonographic findings of accessory breast tissue in axilla and related diseases. *J Ultrasound Med* 2017;36:1469–78.
- [8] Singal R, Mehta SK, Bala J, Zaman M, Singal S. A study of evaluation and management of rare congenital breast diseases. *J Gynecol Obstetr* 2019;7(3):68–71.
- [9] Scanlan KA, Propeck PA. Accessory breast tissue in an unusual location. *Am J Roentgenol* 1996;166(2):339–40.
- [10] Down S, Barr L, Baildam AD, Bundred N. Management of accessory breast tissue in the axilla. *Br J Surg* 2003;90(10):1212–14.
- [11] Ajmal M, Van Fossen MK. *Breast Fibroadenoma*. Stat Pearls. Treasure Island: Stat Pearls Publishing; 2018.