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Case Report

A Giant Breast Hamartoma: A Case Report and Review of the Literature

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Abstract

Mammary hamartomas are benign and infrequent lesions that grow slowly; imaging is generally specific, based essentially on mammography where it presents as a breast in the breast.

We present the case of a 42-year-old woman, recently diagnosed with CCI of the left breast, who presented with a large palpable mass in the right breast. The imaging findings were typical for a breast hamartoma.

The unusual presentation of breast hamartomas justifies histological confirmation by biopsy to exclude a tumour. The incidence of malignancy is low, but cases of malignancy have been reported. Characteristic radiological imaging can minimise unnecessary surgery and morbidity.

Keywords: *Breast; Hamartoma; Mammography; Ultrasonography; Magnetic Resonance Imaging (MRI)*

Introduction

Hamartoma is a benign pseudotumour that can develop in various organs, particularly the lung, kidney, skin or more rarely the breast. It consists of circumscribed masses, often encapsulated, composed of a mixture of tissues from the neighbouring organ [1].

Hamartoma accounts for 4 - 8% of benign breast tumours of women, with rare cases described for men [2,3].

As in the present case, hamartoma is often asymptomatic.

Case Report

A 42-year-old woman, multiparous, followed in our department for infiltrating ductal cancer of the left breast, who presented with a painless tumefaction of the right breast.

Clinical examination revealed a large right breast mass occupying the internal quadrants, well limited, with soft consistency, movable in the superficial and deep areas, with no signs of inflammatory disease or nipple retraction.

Mammography showed a large, oval, well-limited mass in the inner quadrants of the right breast, with regular contours and heterogeneous density: radiopaque of glandular tissue and radiolucent of fatty tissue, surrounded by a thin radiopaque border (Figure 1).

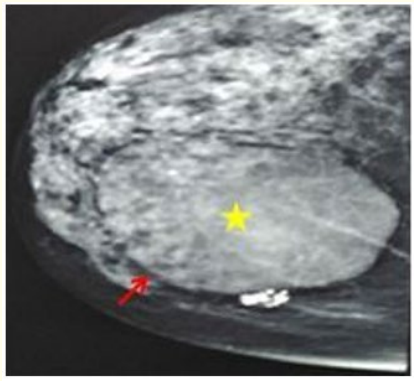

Figure 1: *Craniocaudal mammogram showing a well-defined mass (⭐) containing a substantial amount of fat (radiolucent) and fibroepithelial components (radiopaque) surrounded by a radiolucent halo (red arrow) defining the margin of the mass in the inner quadrants of the right breast, measuring: 65x48mm, giving the appearance of a breast within a breast.*

Ultrasound revealed a voluminous, well-encapsulated, mass with a long axis parallel to the skin, heterogeneously structured with hypoechoic and hyperechoic areas, non-attenuating, pushing back against neighbouring breast tissues, measuring: 65 x 48 mm, without any suspicious axillary adenopathy (Figure 2).

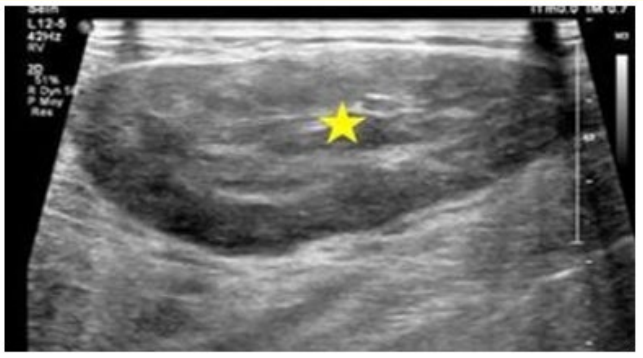

Figure 2: *Ultrasound shows a solid oval mass (⭐) with an inhomogeneous echo structure reflecting the presence of hypoechoic and hyperechoic tissue components, pushing back the surrounding structures, measuring: 65 x 48 mm.*

Breast MRI, as part of extension studies in search of multifocality due to the presence of evolving cancer, revealed a large, oval, well-encapsulated mass in the inner quadrants of the right breast, with a dual component: glandular and fatty, gradually enhancing according to a type 1 curve (Figure 3).

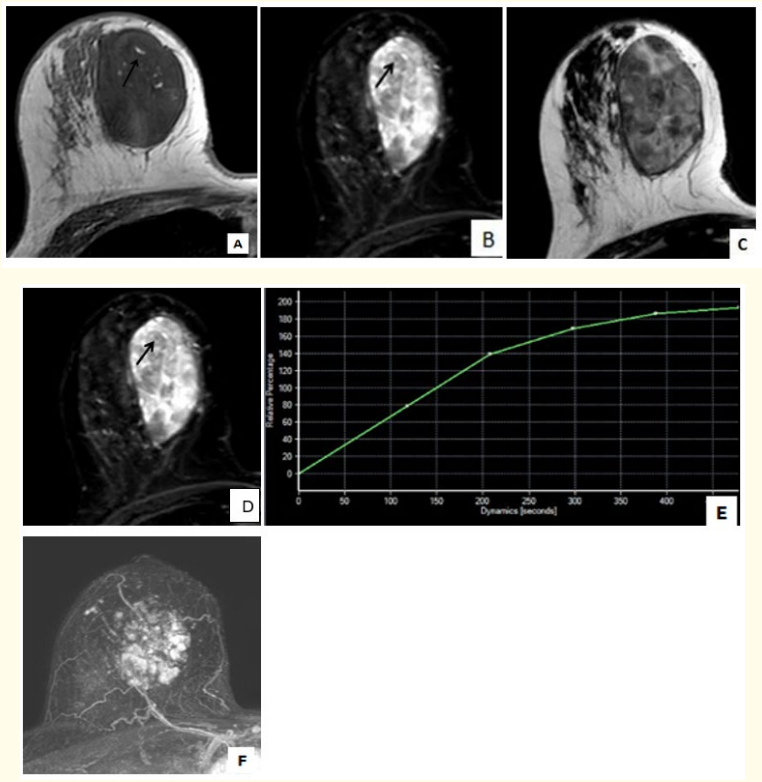

Figure 3A-3F:

Figure 3A: Spin-echo weighted T1 sequence without fat suppression showing a well encapsulated mass with heterogeneous signal predominantly hyposignal with fine hypersignal trabeculae (black arrow) which fade on the STIR sequence: Short T1 Inversion Recovery (Figure 3B) (black arrow) in relation to a fatty contingent.

Figure 3C: T2 spin echo sequence without fat suppression showing the mass with a heterogeneous signal.

Figure 3D: T1-weighted sequence with dynamic injection of Gadolinuim showing heterogeneous and progressive enhancement of the mass according to a type 1 curve (Figure 3E) confirmed by dynamic contrast on maximum intensity projection (MIP) images (Figure 3F).

Discussion

Breast hamartomas are benign tumours first described as mastomas by Prym in 1928 [4] and have been reported as adenolipomas, fibroadenolipomas or lipofibroadenomas [5]. The breast hamartoma was described for the 1st time as a hamartoma in 1971 by Arrigoni., *et al*. [6] and was introduced in the World Health Organization classification in 1981 [5].

The breast hamartoma is a rare benign tumour or more accurately a pseudotumour, well encapsulated and formed to varying degrees by glandular and adipose tissue [7]. The hamartoma is a slow-growing tumour with a variable diameter between 2 and 5 cm, but it can

sometimes be much larger [8]. Breast hamartomas are seen mainly in women with an average age of over 35 years [9,10] in the peri-menopausal period, with an incidence around 4.8% of all benign tumours. The incidence of hamartomas is clearly increasing due to the breast cancer screening programme [10]. Hamartomas are rarely seen in ectopic tissue in the axillary or inguinal region and are also rarely detected in men [11].

Hamartomas are usually asymptomatic, occasionally associated with unilateral mastodynia without a palpable mass [5]. Clinical examination usually reveals a well-circumscribed, mobile, smooth, oval mass with a consistency similar to normal breast tissue; hamartomas are rarely as firm as adenofibromas. The diagnosis of breast hamartoma is difficult to make clinically, and requires a combination of radiology and histology.

Hamartoma is diagnosed by mammography, showing a well-circumscribed, rounded or oval mass separated from the breast parenchyma by a thin radiopaque pseudocapsule, giving it the appearance of a breast within a breast [9,12,13]. The degree of opacity is proportional to the percentage of fat in the breast parenchyma. When it contains a large portion of fat, it appears similar to a lipoma, liponecrosis or oily cyst; the history of previous trauma may redetermine the diagnosis in the last two cases [5] and when it contains a greater degree of fibro-glandular tissue, it may be mistaken for a fibroadenoma.

The ultrasound presentation of the hamartoma is highly variable and depends on its adipose and fibrous tissue content. The most typical appearance is that of a solid, well encapsulated, oval mass, parallel to the skin, often of heterogeneous structure containing both hypoechoic and hyperechoic areas in the shape of a band or nodule, without hypervascuarisation on colour Doppler. Diagnosis becomes more difficult if the mass is small and poor in adipose tissue or when the pseudocapsule is incomplete [3,14]. Hamartomas rich in adipose tissue may appear as lipomas.

Data in the literature relating to the elastographic characteristics of hamartomas are limited, and compressibility and consistency depend on the amount of adipose tissue, which is variable [14]. In the majority of cases, the colour scale shows a mass that is less elastic than the surrounding tissue, with an Italian elastographic score of 3 or 4 [15].

The typical appearance of a breast hamartoma without suspicious criteria on mammography or ultrasound is not an indication for MRI, although hamartomas may be found incidentally on MRI. Hamartomas on MRI are well-demarcated masses with heterogeneous structure and intensity, showing glandular and adipose components with a thin pseudocapsule. Gadolinium injection shows progressive enhancement according to a type 1 curve. If suspicious features are seen on mammography or ultrasound, MRI with Gadolinium injection may be useful in making the diagnosis [16,17].

Due to its characteristic appearance on imaging, the diagnosis of hamartoma is generally straightforward. In rare cases, the diagnosis is difficult, and the diagnosis is differentiated from a fibroadenoma or a phyllodes tumour (both of which do not contain internal fat), or from a lipoma (homogeneous fat with an imperceptible soft tissue component) [5,13,18].

Pathologically, hamartoma is composed of normal breast tissue with mammary ducts, lobules, fibrous stroma, adipose tissue and varying degrees of smooth muscle [6]. Diagnosis is generally made by fine needle cytopuncture or ultrasound-guided biopsy.

Surgical resection identifies hamartomas and allows all tissue components to be studied [19]. Surgical treatment is indicated for patients with a doubtful hamartoma or a firm diagnosis of hamartoma [20].

Breast hamartomas are benign tumours and not precancerous lesions. However, due to the presence of glandular tissue, they can develop malignant architectural changes within the normal tissue of the hamartoma, which explains why a definitive histopathological

diagnosis is imperative. The incidence of malignancy in normal breast tissue is 0.1%. One study showed lobular cancers *in situ* and invasive carcinomas when an excisional biopsy was performed on irregular microcalcifications initially suspicious on mammography [21]. However, lesions with an abnormal growth pattern and atypical features require biopsy, as breast carcinoma can develop within a hamartoma [13]. Surgical excision with a resection margin is necessary because of the risk of recurrence and, rarely, of occult foci of malignancy within the lesion [20].

Conclusion

Breast hamartomas are uncommon, slow-growing benign tumours that are often easily diagnosed when the appearance is typical on mammography and ultrasound, and do not require any other diagnostic means. When the appearance is less typical, the differential diagnosis may be fibroadenoma, lipoma, phyllodes tumours or cancer.

Characteristic radiological imaging can reduce morbidity and avoid unnecessary surgery.

The unusual presentation and low risk of synchronous cancer mean that a biopsy is required to confirm the diagnosis.

Author's Contributions

All the authors contributed to study concept, data analysis and writing the paper. All authors read and approved the final version of the manuscript.

Conflicts of Interest

The authors declare that there are no conflicts of interest regarding the publication of this manuscript.

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