Case Report

Ultrasonographic features of adenomyoepithelial adenosis: a case report and literature review

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Received February 18, 2014; Accepted March 21, 2014; Epub April 15, 2014; Published April 30, 2014

Abstract: Adenomyoepithelial adenosis of the breast is extremely rare. We treated a 35-year-old woman with two small painless hard lumps in her breasts. On ultrasonography, the lesion in the right breast (upper inner quadrant) showed heterogeneous echogenicity and irregular, well-defined, uniformly hypoechoic nodules separated by funiform hyperechoic areas. The entire lesion had unclear boundaries. Excisional biopsy showed adenomyoepithelial adenosis. The mass in the left breast was a fibroadenoma. Adenomyoepithelial adenosis can show local recurrence and malignant degeneration, therefore, preoperative ultrasonographic diagnosis is important for surgical planning. The differential diagnosis includes common adenosis, fibroadenoma, adenomyoepithelioma and adenomyoepithelial carcinoma.

Keywords: Adenomyoepithelial adenosis, breast, ultrasonography, adenosis, adenomyoepithelioma

Introduction

Myoepithelial cells are widely present in the breast, and their hyperplasia may result in a spectrum of diseases ranging from myoepitheliolysis to myoepithelial carcinoma. Adenomyoepithelial adenosis (AMEA) is an extremely rare type of adenosis with highly proliferative glandular epithelial and myoepithelial cells [1]. In this paper, we describe the case of a 35-year-old woman with AMEA of the right breast and fibroadenoma of the left breast. The ultrasonographic features and differential diagnosis of AMEA are discussed, along with a review of the literature.

Case report

A 35-year-old woman presented with a 2-year history of bilateral breast masses. Two years ago, a small painless mass measuring 2 cm × 1 cm was incidentally discovered in her left breast, and simultaneously, a lump measuring 1 cm × 1 cm was found in her right breast. A physical examination revealed a hard, palpable mass (2 cm × 1 cm) at the medial border of the areola of her left breast and a soft, elastic lump (4 cm × 3.5 cm) in the upper inner quadrant of the right breast. Both masses had clear borders and were mobile. There was no lymph node swelling, local tenderness and redness or orange peel-like appearance of the skin. Nipple retraction and discharge were not observed. Ultrasonography displayed an isolated oval hypoechoic solid mass measuring 2.2 cm × 1.3 cm (wider than tall) in her left breast. The boundary of this mass was clear with lateral shadows at the margins. The capsule of the mass was thin, and the internal echo was uniform. Color Doppler flow imaging (CDFI) showed minor flow in the mass (Figure 1A). Imaging of the right breast showed a 4.2 cm × 3.6 cm complex mass with heterogeneous echo and ovoid or irregular hypoechoic nodules separated by funiform hyperechoic areas. The nodules had clear boundaries and uniform echo, and the largest nodule measured 1.9 cm × 0.9 cm (wider than tall). There was circuitous flow at the edges of nodules on CDFI (Figure 1B). The boundary of the whole lump was unclear. Several lymph nodes without metastatic chang-
es were found in both the right and left axillary fossae. The ultrasonographic diagnosis was fibroadenoma in the left breast and possible adenomatous hyperplasia, which could not be accurately classified, in the right breast.

Bilateral excision of the breast masses was performed. During the surgery, an isolated firm mass with clear boundaries was found in the left breast, while an ill-defined hard area consisting of multiple well-circumscribed nodules was observed in the right breast (Figure 2). The pathological diagnosis was AMEA of the right breast and fibroadenoma of the left breast. The informed consent was obtained from the patient.

Discussion

AMEA of the breast is a controversial terminology. It was recommended as a distinct form of adenosis by Kiaer et al in 1984 [2], and was included in the 2003 revision of the WHO classification of breast tumors [3] but was deleted in 2012. It is similar in several ways to common adenosis with one significant difference: AMEA involves myoepithelial cells while common adenosis does not [2].

Owing to the limitation of core needle aspiration biopsy and the lack of specific imaging findings, the preoperative diagnosis of AMEA is thought to be very difficult. Moreover, the ultrasonographic features of AMEA have been only rarely described because of the low incidence of this condition [1, 4, 5]. Erel et al. reported a case of AMEA appearing as an irregular speculated hypoechoic mass measuring 1.2 cm × 1.3 cm with a posterior acoustic shadow on ultrasonography [4]. Hiroyuki et al. observed an AMEA that appeared as an irregular hypoechoic mass measuring 5 cm × 5 cm × 1 cm and contained multiple small cysts [1]. Hitoshi et al. also described an AMEA which was irregular in shape and associated with a posterior acoustic shadow on ultrasonography, both of which were strongly suggestive of cancer [5]. A mastectomy was therefore performed for that patient. In the present case, we detected a 4.2 cm × 3.6 cm complex mass with heterogeneous echo and ovoid or irregular hypoechoic nodules separated by funiform hyperechoic areas. Unlike the slow-growing fibroadenoma in the left breast, the AMEA nodules in the right breast showed rapid growth, which suggested that both the glandular epithelial and myoepithelial cells were highly proliferative. This may be a dynamic characteristic of AMEA.

AMEA has a potential for local recurrence and progression to adenocarcinoma or malignant adenomyoepithelioma [2, 4, 5]. Therefore, wide
local excision with negative margins is recommended, which differs from the treatment for common adenosis and fibroadenoma of the breast. Accurate preoperative determination of the extent of the tumor is important for surgical planning. On ultrasonography, AMEA may be confused with other myoepithelial/stromal cell-rich structures that mimic this lesion, such as common adenosis, fibroadenoma, adenomyoepithelioma and adenomyoepithelial carcinoma. Common adenosis appears as a weak-to-low-echoic mass that is wider than tall on ultrasonography. The boundary of the nodules is usually unclear, and there is no envelope or space-occupying effect. On CDFI, a little blood flow can be detected in the nodules. Fibroadenoma is usually elliptical (wider than tall), shows uniform echogenicity and is completely encompassed by a thin, echogenic capsule. Adenomyoepithelioma lacks specific ultrasonographic characteristics, and often appears as a benign nodule with surrounding ductal extension. It can occasionally be confused with an intraductal papilloma, and some tumors show angular margins and spicule signs [6]. Adenomyoepithelial carcinoma is almost never elliptical or gently lobulated. Moreover, it is usually associated with other suspicious findings, such as angular margins, taller-than-wide shape, microlobular, ductal extension and branch pattern [6].

Conclusion

Although AMEA is a rare myoepithelial hyperplastic lesion, it has the potential for local recurrence and malignant degeneration. Preoperative diagnostic evaluation is important to avoid improper treatment. The ultrasonographic appearance of this lesion shows wide variations, and more cases of AMEA need to be assessed in order to accurately summarize the ultrasonographic characteristics of this lesion.

Acknowledgements

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Disclosure of conflict of interest

None.

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