



Case Report **Breast Imaging**

Ductal carcinoma *in situ* arises from microglandular adenosis and atypical microglandular adenosis in a young woman

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ABSTRACT

Microglandular adenosis (MGA) and atypical microglandular adenosis (AMGA) are intensely rare and distinctive forms of adenosis of the breast, usually occurring in middle-aged women. Carcinoma arising in MGA is an extremely rare subtype of breast carcinoma, and most reported cases are of invasive carcinoma. Ultrasound and magnetic resonance imaging are accurate imaging modalities for diagnosing these abnormalities. Our goal in this article was to report a rare instance of ductal carcinoma *in situ* (DCIS) arising from MGA and AMGA in a very young Vietnamese woman who presented with a palpable mass in her right breast for 1 month. During clinical examination and imaging, suspected lesions were found and categorized as BI-RADS 4a. The final histopathological findings confirmed DCIS arising from MGA/AMGA. In this patient, the disease was detected and managed early when the lesion was localized in the duct and there were no signs of invasive ductal carcinoma.

Keywords: Microglandular adenosis, Atypical microglandular adenosis, Ductal carcinoma *in situ*, Breast, Magnetic resonance imaging, Ultrasound

INTRODUCTION

Microglandular adenosis (MGA) is a rare neoplastic lesion of the breast. MGA is characterized by the disordered proliferation of small open glands comprised bland epithelial cells and retains the basement membrane but lacks myoepithelium, making it difficult to distinguish from breast carcinoma (BC).^[1] Atypical microglandular adenosis (AMGA) refers to a lesion with the characteristics of MGA and the additional characteristics of structural complexity and the presence of atypical cells.^[2] Although MGA and AMGA are very rare, accounting for <0.1% of all lesions in BC,^[1] carcinoma arising in MGA (MGACA) is reported to comprise 27% of all MGA cases.^[2,3] MGA is considered a non-obligate precursor of triple-negative (progesterone [PR]-, estrogen [ER]-, and human epidermal growth factor receptor 2 [Her2]-negative) BC, a subtype with a poor prognosis.^[2,3] Therefore, understanding the characteristics of MGA/AMGA as well as MGACA is necessary for the early diagnosis and treatment. The purpose of this article is to present the clinical, imaging, and histological features as well as the treatment and follow-up plan of a very rare case of ductal carcinoma *in situ* (DCIS) arising from MGA/AMGA in a 21-year-old woman.

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CASE PRESENTATION

A 21-year-old Vietnamese woman with an unremarkable personal medical history and family history presented to the hospital complaining of a palpable tumor in her right breast. An irregular solid mass in the right breast without ipsilateral axillary lymph nodes was discovered during a clinical examination.

A 35 × 12 × 39 mm hypoechoic lesion with an irregular shape, indistinct margin, and parallel orientation was visible on ultrasound (US) at 9 o'clock position, 4 cm from the right nipple. The lesion was subsequently categorized as BI-RADS 4A [Figure 1]. A 19 × 38 × 31 mm lesion was seen on magnetic resonance imaging (MRI) at 9 o'clock position, 3.8 cm from the nipple. The lesion was non-mass and mildly hyperintense, with a focal distribution, on STIR imaging [Figure 2a]. The lesion was hyperintense on diffusion-weighted imaging and hypointense on the apparent diffusion coefficient map [Figure 2b and c]. On dynamic T1-weighted imaging with contrast, the lesion exhibited a fast initial rise followed by a plateau enhancement during the delayed phase [Figure 2d and e]. Thereafter, a core biopsy of the right breast lesion was performed, and the histopathological results indicated atypical ductal hyperplasia. The patient was treated with conservative surgery.

Post-operative histopathology revealed lesions ranging from MGA to AMGA and DCIS [Figure 3a] that showed no signs of infiltrating the basement membrane of the invasive ductal carcinoma (IDC). All margins were negative for DCIS, and the shortest distances from DCIS to the posterior and anterior margins were 0.1 mm and 0.15 mm, respectively; the anterior margin was MGA and the posterior margin was AMGA. Immunohistochemistry results revealed an ER-, PR-, and Her2-negative MGA phenotype. S100 was strongly positive and pervasive in all MGA/AMGA regions [Figure 3b and c]. Moreover, p63 negativity indicated a lack of myoepithelium in the MGA/AMGA [Figure 3d]. Thereafter, the patient underwent re-resection due to the appearance of MGA and AMGA in the margins and sentinel lymph node biopsy due to the presence of DCIS. Finally, all margins, as well as the sentinel lymph nodes, were negative.

DISCUSSION

The first detailed description of MGA was published in 1968 by McDivitt. All previously reported MGA patients were women in the age range of 28–82 years, usually aged 40–55 years.^[1] Patients often presented to the hospital with symptoms such as a palpable breast mass but was sometimes detected by screening. Physical examination and imaging revealed lesions that were usually 3–4 cm in size but sometimes went up to 20 cm.^[4] In our case, the lesion was detected early (when the patient was just 21 years old)

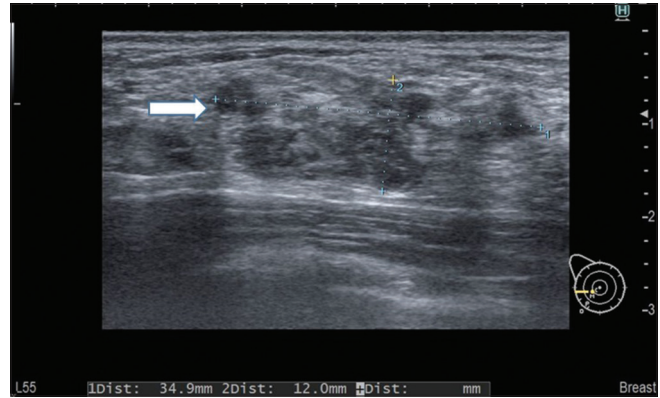


Figure 1: A 21-year-old Vietnamese woman with a palpable tumor in her right breast. An irregularly shaped hypoechoic mass (arrow) in the left breast was detected using color-Doppler mode ultrasound.

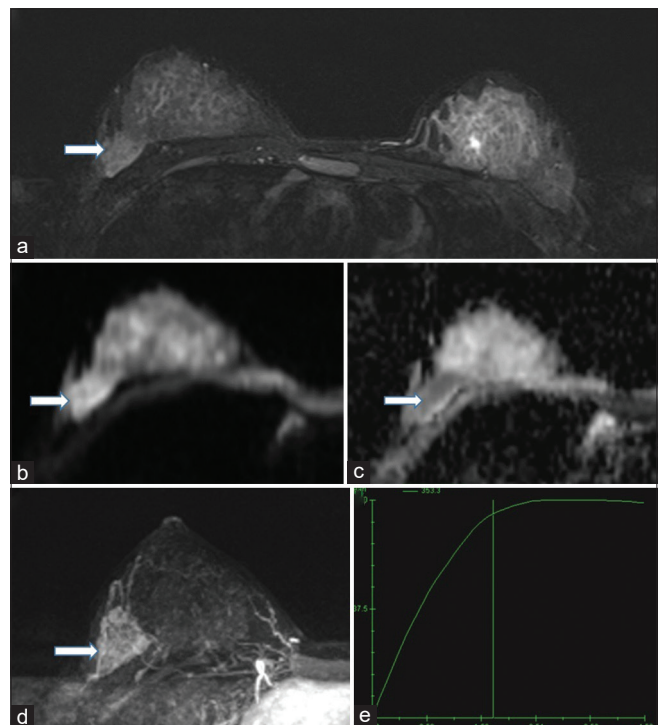


Figure 2: A 21-year-old Vietnamese woman with a palpable tumor in her right breast. A hyperintense lesion (arrow) was seen on short tau inversion recovery (a). A restricted diffusion lesion was seen via diffusion-weighted imaging (b) and the apparent diffusion coefficient map (c). The lesion (arrow) showed a fast-initial increase on dynamic T1-weighted imaging with contrast enhancement, followed by a plateau enhancement in the delayed phase (d and e).

compared to previous reports, with a palpable breast mass over 3 cm in size.

MGA/AMGA seems to appear as a “vague area of altered echotexture” on US;^[1] therefore, MRI is an effective modality with high sensitivity to evaluate the features and extent

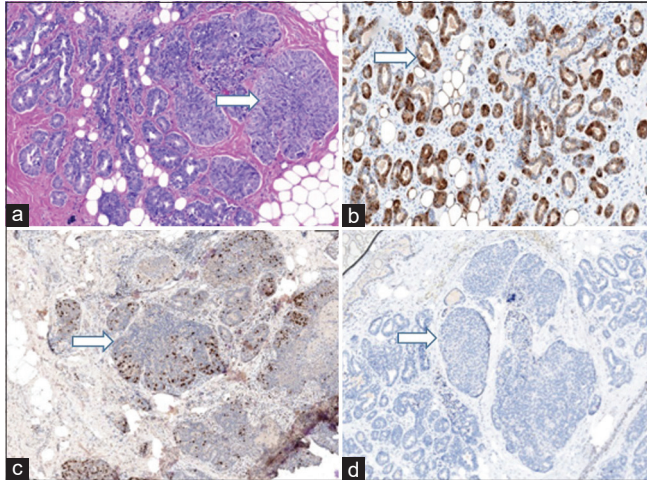


Figure 3: Photomicrographs of the histopathological specimen. Ductal carcinoma *in situ* arising from microglandular adenosis/atypical microglandular adenosis (MGA/AMGA) (H and E $\times 100$) (a). S100 was strongly positive and pervasive in all MGA/AMGA regions (S100 $\times 20$, $\times 50$) (b and c). P63 was negative in the MGA/AMGA regions, indicating a lack of myoepithelium (p63 $\times 20$) (d).

of the lesion. MGA/AMGA often presents as non-mass enhancement lesions on MRI.^[5] With such clinical and imaging features, it is difficult to distinguish MGA/AMGA from DCIS, IDC, or other benign lesions, so a biopsy is often required. In addition, imaging is also a necessary tool for post-treatment follow-up.^[6] In our patient, the lesion appeared mildly hypoechoic, with an indistinct margin on US, and a non-mass enhancement on MRI, which is consistent with the previous reports of MGA/AMGA.

Histologically, MGA is characterized by an erratic infiltration of small, slightly open, and round glands in a thick, hypocellular, and stringy towel or adipose mammary tissue.^[7] Positive pancytokeratin and negative smooth muscle cause it to be confused with ductal carcinoma. The lack of stromal tissue and the appearance of a thickened basement membrane makes MGA different from carcinoma.^[4,8] MGA/AMGA and MGACA have common immunohistochemical features, tending to be negative for ER, PR, Her2, and CK5/6 and positive for CK8/18, S100, and receptor epidermal growth factor. There is also increasing positivity for Ki-67 and p53 as lesions “progress” from MGA to atypical MGA, DCIS, and invasive carcinoma.^[9-11]

MGA is a known pitfall in breast pathology as it may mimic carcinoma clinically, radiologically, and histologically.^[6] MGA is associated with carcinomas in approximately 25–27% of cases, and AMGA has been described as a lesion in transition to carcinoma. It is unclear whether MGA represents a truly benign proliferation or an indolent precursor lesion, so the currently recommended management strategy for MGA is complete excision, with the excision specimens needing to be sampled thoroughly to rule out the possibility of an

associated carcinoma.^[1] During follow-up, most MGA cases have not recurred. However, if MGA/AMGA is associated with DCIS/IDC, the patient is treated according to the current DCIS/IDC management protocol. The prognosis of MGACA is controversial in the literature. In one study, only one patient developed lung metastasis 24 months after surgery, while ten patients remained without recurrence after 10–64 months of follow-up. This suggests that although aggressive histopathological features (high nuclear grade) and immunohistochemistry (triple-negative, high Ki-67 index) are often associated with poor prognosis, most patients with MGACA have a relatively favorable outcome.^[12] In our case, the patient was treated and followed up like other DCIS cases.

CONCLUSION

The clinical features, imaging, as well as pathology of MGA/AMGA are difficult to distinguish from those of BC, so experienced doctors are required for diagnosis and treatment. US and MRI are useful modalities to evaluate the characteristics and extent of the lesion. In the presence of MGA/AMGA, lesions need to be resected and the excision specimens need to be analyzed to detect the accompanying DCIS/IDC component, thereby ensuring appropriate treatment and follow-up. Further studies should be performed to enhance diagnostic accuracy and optimal treatment for AMGCA.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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