

World Journal of *Clinical Cases*

World J Clin Cases 2021 September 6; 9(25): 7292-7613



EDITORIAL

- 7292 Radiation oncology practice during COVID-19 pandemic in developing countries
Abuhijla F, Abuhijlih R, Mohamad I

OPINION REVIEW

- 7297 Complete mesocolic excision and central vascular ligation in colorectal cancer in the era of minimally invasive surgery
Franceschilli M, Di Carlo S, Vinci D, Sensi B, Siragusa L, Bellato V, Caronna R, Rossi P, Cavallaro G, Guida A, Sibio S
- 7306 Fecal diversion in complex anal fistulas: Is there a way to avoid it?
Garg P, Yagnik VD, Dawka S

MINIREVIEWS

- 7311 Regulatory roles of extracellular vesicles in immune responses against *Mycobacterium tuberculosis* infection
Yan Z, Wang H, Mu L, Hu ZD, Zheng WQ
- 7319 Aortic stenosis and Heyde's syndrome: A comprehensive review
Lourdusamy D, Mupparaju VK, Sharif NF, Ibebuogu UN

ORIGINAL ARTICLE**Retrospective Study**

- 7330 Key determinants of misdiagnosis of tracheobronchial tuberculosis among senile patients in contemporary clinical practice: A retrospective analysis
Tang F, Lin LJ, Guo SL, Ye W, Zha XK, Cheng Y, Wu YF, Wang YM, Lyu XM, Fan XY, Lyu LP
- 7340 Long-term outcome of pancreatic function following oncological surgery in children: Institutional experience and review of the literature
Bolasco G, Capriati T, Grimaldi C, Monti L, De Pasquale MD, Patera IP, Spada M, Maggiore G, Diamanti A
- 7350 Efficacy of arbidol in COVID-19 patients: A retrospective study
Wei S, Xu S, Pan YH
- 7358 Characteristic analysis of clinical coronary heart disease and coronary artery disease concerning young and middle-aged male patients
Peng KG, Yu HL
- 7365 Quantitative analysis of early diabetic retinopathy based on optical coherence tomography angiography biological image
Shi Y, Lin PY, Ruan YM, Lin CF, Hua SS, Li B

- 7372 Mucin 1 and interleukin-11 protein expression and inflammatory reactions in the intestinal mucosa of necrotizing enterocolitis children after surgery

Pan HX, Zhang CS, Lin CH, Chen MM, Zhang XZ, Yu N

Observational Study

- 7381 Research on the prognosis of different types of microvessels in bladder transitional cell carcinoma

Wang HB, Qin Y, Yang JY

- 7391 Is burnout a mediating factor between sharps injury and work-related factors or musculoskeletal pain?

Chen YH, Tsai CF, Yeh CJ, Jong GP

- 7405 Role of international normalized ratio in nonpulmonary sepsis screening: An observational study

Zhang J, Du HM, Cheng MX, He FM, Niu BL

Randomized Controlled Trial

- 7417 Clinical effectiveness of adding probiotics to a low FODMAP diet: Randomized double-blind placebo-controlled study

Turan B, Bengi G, Cehreli R, Akpınar H, Soytürk M

SYSTEMATIC REVIEWS

- 7433 Association between COVID-19 and anxiety during social isolation: A systematic review

Santos ERRD, Silva de Paula JL, Tardieux FM, Costa-e-Silva VN, Lal A, Leite AFB

CASE REPORT

- 7445 Avascular necrosis of the first metatarsal head in a young female adult: A case report and review of literature

Siu RWH, Liu JHP, Man GCW, Ong MTY, Yung PSH

- 7453 Successful treatment of solitary bladder plasmacytoma: A case report

Cao JD, Lin PH, Cai DF, Liang JH

- 7459 Pseudomyxoma peritonei originating from intestinal duplication: A case report and review of the literature

Han XD, Zhou N, Lu YY, Xu HB, Guo J, Liang L

- 7468 Agranulocytosis following injection of inactivated Japanese encephalitis vaccine (Vero cell): A case report

Wang L, Zhang X, Liu YT

- 7472 Importance of clinical suspicion and multidisciplinary management for early diagnosis of a cardiac laminopathy patient: A case report

Santobuono VE, Guaricci AI, Carulli E, Bozza N, Pepe M, Ranauro A, Ranieri C, Carella MC, Loizzi F, Resta N, Favale S, Forleo C

- 7478 First case of forearm crisscross injury in children: A case report

Jiang YK, Wang YB, Peng CG, Qu J, Wu DK

- 7484** Octreotide-induced acute life-threatening gallstones after vicarious contrast medium excretion: A case report
Han ZH, He ZM, Chen WH, Wang CY, Wang Q
- 7490** Acute deep venous thrombosis induced by May-Thurner syndrome after spondylolisthesis surgery: A case report and review of literature
Yue L, Fu HY, Sun HL
- 7498** Successful treatment of refractory lung adenocarcinoma harboring a germline *BRCA2* mutation with olaparib: A case report
Zhang L, Wang J, Cui LZ, Wang K, Yuan MM, Chen RR, Zhang LJ
- 7504** Effective treatment of polyneuropathy, organomegaly, endocrinopathy, M-protein, and skin changes syndrome with congestive heart failure: A case report
Fu LY, Zhang HB
- 7512** Awake craniotomy for auditory brainstem implant in patients with neurofibromatosis type 2: Four case reports
Wang DX, Wang S, Jian MY, Han RQ
- 7520** Coexistence of tuberculosis and squamous cell carcinoma in the right main bronchus: A case report
Jiang H, Li YQ
- 7527** Is simultaneous presence of IgG4-positive plasma cells and giant-cell hepatitis a coincidence in autoimmune hepatitis? A case report
Tan YW, Wang JM, Chen L
- 7535** Surgical treatment of delayed cervical infection and incomplete quadriplegia with fish-bone ingestion: A case report
Li SY, Miao Y, Cheng L, Wang YF, Li ZQ, Liu YB, Zou TM, Shen J
- 7542** Neonatal biliary atresia combined with preduodenal portal vein: A case report
Xiang XL, Cai P, Zhao JG, Zhao HW, Jiang YL, Zhu ML, Wang Q, Zhang RY, Zhu ZW, Chen JL, Gu ZC, Zhu J
- 7551** Hemorrhagic transformation after acute ischemic stroke caused by polycythemia vera: Report of two case
Cao YY, Cao J, Bi ZJ, Xu SB, Liu CC
- 7558** Treatment of lower part of glenoid fractures through a novel axillary approach: A case report
Jia X, Zhou FL, Zhu YH, Jin DJ, Liu WX, Yang ZC, Liu RP
- 7564** Trigger finger at the wrist caused by an intramuscular lipoma within the carpal tunnel: A case report
Huang C, Jin HJ, Song DB, Zhu Z, Tian H, Li ZH, Qu WR, Li R
- 7572** Thrombolysis and embolectomy in treatment of acute stroke as a bridge to open-heart resection of giant cardiac myxoma: A case report
Chang WS, Li N, Liu H, Yin JJ, Zhang HQ
- 7579** Breast adenoid cystic carcinoma arising in microglandular adenosis: A case report and review of literature
An JK, Woo JJ, Kim EK, Kwak HY

- 7588** Diagnosis and management of ophthalmic zoster sine herpete accompanied by cervical spine disc protrusion: A case report
Yun G, Kim E, Baik J, Do W, Jung YH, You CM
- 7593** Hemorrhagic pericardial effusion following treatment with infliximab: A case report and literature review
Li H, Xing H, Hu C, Sun BY, Wang S, Li WY, Qu B
- 7600** Wernicke's encephalopathy in a rectal cancer patient with atypical radiological features: A case report
Nie T, He JL
- 7605** Total hip revision with custom-made spacer and prosthesis: A case report
Liu YB, Pan H, Chen L, Ye HN, Wu CC, Wu P, Chen L

ABOUT COVER

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The primary aim of *World Journal of Clinical Cases* (*WJCC*, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING

The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, Scopus, PubMed, and PubMed Central. The 2021 Edition of Journal Citation Reports® cites the 2020 impact factor (IF) for *WJCC* as 1.337; IF without journal self cites: 1.301; 5-year IF: 1.742; Journal Citation Indicator: 0.33; Ranking: 119 among 169 journals in medicine, general and internal; and Quartile category: Q3. The *WJCC*'s CiteScore for 2020 is 0.8 and Scopus CiteScore rank 2020: General Medicine is 493/793.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Yan-Xia Xing; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lai Wang.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

September 6, 2021

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INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>

Breast adenoid cystic carcinoma arising in microglandular adenosis: A case report and review of literature

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Author contributions: An JK contributed to conceptualization and supervision; An JK and Kim EK do the original draft preparation; An JK, Kim EK and Kwak HY do the data curation; An JK, Woo JJ and Kim EK do the formal analysis; all author validate, review and edit the manuscript.

Informed consent statement: This study was reviewed and approved as a retrospective case report by the Institutional Review Board of Eulji Medical Center, Seoul, Korea (EMCIRB 2021-01-003), with informed consent from the patient waived.

Conflict-of-interest statement: The authors have no potential conflicts of interest to disclose.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external

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Abstract

BACKGROUND

Breast adenoid cystic carcinoma (AdCC) is a rare invasive carcinoma composed of epithelial and myoepithelial cells. Microglandular adenosis (MGA) is a rare benign proliferative lesion consisting of small, uniform, and round glands formed by a single layer of epithelial cells and basement membrane without a myoepithelial cell layer. MGA may progress to atypical MGA and carcinoma arising in MGA. Among various invasive carcinomas from MGA, AdCC has been rarely reported. Here, we report a case of AdCC arising in MGA.

CASE SUMMARY

A 59-year-old woman was diagnosed with a newly developed density on a routine mammogram. The density was similar to or slightly lower than that of the breast parenchyma. Sonography showed an irregular mass with a slightly higher echo than that of fat. Magnetic resonance imaging showed an irregular mass with a similar T1 signal intensity and a slightly higher T2 signal intensity compared to muscles or the breast parenchyma. The lesion showed heterogeneous internal enhancement with an initially slow and delayed persistent enhancing pattern. Microscopically, the tumor was composed of invasive AdCC, *in situ* AdCC, and MGA. AdCC is composed of basaloid and ductal epithelial cells forming cribriform or solid sheets, or haphazardly scattered small cribriform or tubular glands. MGA showed small glands with a single epithelial lining and retained lumen. S-100 staining was strongly positive in MGA area. The patient underwent breast-conserving surgery with sentinel lymph node biopsy.

CONCLUSION

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Manuscript source: Unsolicited manuscript

Specialty type: Radiology, nuclear medicine and medical imaging

Country/Territory of origin: South Korea

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): B
Grade C (Good): 0
Grade D (Fair): 0
Grade E (Poor): 0

Received: March 31, 2021

Peer-review started: March 31, 2021

First decision: June 7, 2021

Revised: June 7, 2021

Accepted: July 29, 2021

Article in press: July 29, 2021

Published online: September 6, 2021

P-Reviewer: Vinh-Hung V

S-Editor: Wu YXJ

L-Editor: A

P-Editor: Xing YX



Breast AdCC arising in MGA showed unique imaging findings that was different from usual invasive cancer.

Key Words: Breast neoplasms; Fibrocystic breast disease; Carcinoma; Adenoid cystic; Case report

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Core Tip: Many pathological or clinical studies have been reported for adenoid cystic carcinoma (AdCC) arising in microglandular adenosis (MGA), but few reports have been reported of radiological findings. In our case, it was characterized by iso- or slight hypo-density in the mammogram and slightly higher echo than that of fat in the ultrasound examination with higher T2 signal intensity and a persistent enhancing pattern in breast magnetic resonance imaging. Although AdCC shows a favorable prognosis and MGA has long been considered a benign entity, there is a risk of MGA becoming malignant and a complete resection should be performed.

Citation: An JK, Woo JJ, Kim EK, Kwak HY. Breast adenoid cystic carcinoma arising in microglandular adenosis: A case report and review of literature. *World J Clin Cases* 2021; 9(25): 7579-7587

URL: <https://www.wjgnet.com/2307-8960/full/v9/i25/7579.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v9.i25.7579>

INTRODUCTION

Adenoid cystic carcinoma (AdCC), a rare breast cancer, accounts for 0.1%-3.5% of all breast tumors. It is known to have a good prognosis[1-5]. AdCC is composed of two main neoplastic cells: (1) Altered myoepithelial cells grouped in nests or outlining characteristic cribriform space; and (2) Epithelial cells lining small ductule-like true lumina[6,7]. AdCC belongs to 'rare and salivary gland-type tumors' among epithelial tumors of the breast in World Health Organization (WHO) classification[2]. Recently, it has been reported that microRNA expression profile of breast AdCC differs from that of salivary gland AdCC[7]. It helps us understand different clinical behaviors of tumors depending on the original site.

Microglandular adenosis (MGA) is a rare benign proliferative lesion consisting of small, uniform, and round glands formed by a single layer of epithelial cells[8,9]. These glands are surrounded by basement membranes without myoepithelial cell layers. Absence of the myoepithelial layer is a characteristic feature that distinguishes MGA from ductal and lobular histology of the breast. Infiltrative proliferation of these glands and absence of the myoepithelial layer may mimic invasive breast carcinoma [8]. Although MGA is a benign entity, it may progress to atypical MGA and carcinoma arising in MGA[10-14]. Various invasive carcinomas arising in MGA have been reported. However, AdCC and its imaging findings have been rarely reported[12,15]. In this study, we report a case of AdCC arising in MGA.

CASE PRESENTATION

Chief complaints

A 59-year-old woman visited our hospital for a routine breast checkup. Since she first had a breast examination at our hospital in 2017, she has been undergoing regular checkups every year. Last year, her breast examination showed negative findings. However, a mammography (MG) performed this year revealed a new density.

History of present illness

She is currently on medication for high blood pressure and diabetes, and has no other breast symptoms.

History of past illness

She had no history of breast surgery.

Personal and family history

She had no family history of cancer, including breast cancer.

Physical examination

The breast lesion was not clearly palpable on physical examination.

Laboratory examinations

No specific laboratory examinations were performed for breast lesion in this patient.

Imaging examinations

In the MG, focal asymmetry was noted in the right upper outer peripheral breast (Figure 1). The density was similar to or slightly lower than that of the breast parenchyma. It was not accompanied by distortions or microcalcifications. In the ultrasound (US) examination, the lesion was shown as a 1.5 cm, irregularly shaped mass with an angular margin (Figure 2). The echo was slightly higher than that of fat. There was no increased vascularity in the color Doppler study. In breast magnetic resonance imaging (MRI), the lesion was shown as a 1.3 cm mass with an irregular shape and margin located in the right upper outer peripheral breast, about 6 cm from the right nipple. The mass showed a similar T1 signal intensity and a slightly higher T2 signal intensity compared to muscles or the breast parenchyma (Figure 3). The lesion exhibited reduced diffusivity with hyperintensity in diffusion-weighted images and hypointensity in the apparent diffusion coefficient map compared to normal breast tissues. In the contrast enhancement study, the lesion showed heterogeneous internal enhancement with an initial slow and delayed persistent enhancing pattern. There was no axillary lymph node metastasis.

Pathological examination

The patient underwent breast-conserving surgery and sentinel lymph node biopsy. Grossly, an ill-defined whitish tumor of roughly 1.6 cm × 1.4 cm was observed. Microscopically, invasive cancer showed diffuse infiltrating nests of cribriform or solid sheets composed of two types of cells: basaloid and ductal epithelial cells (Figure 4A and B). The cribriform space was filled with bluish or pinkish mucoid materials that were Periodic acid Schiff-positive (Figure 4C and D). Surrounding breast tissue also showed extensive intraductal proliferation of tumor cells forming small cribriform or tubules. Small round glands lined by benign-looking uniform cells were noted at the periphery of the intraductal carcinoma, suggesting *in situ* AdCC arising in the background of MGA (Figure 5). Benign small glands of MGA were p63 negative but S-100 positive. However, *in situ* AdCC was p63 positive but S-100 negative, similar to that of invasive element (Figure 6A). Immunohistochemical staining revealed myoepithelial differentiation with strong p63 positivity (Figure 6B) and epithelial differentiation with cytokeratin (CK) (Figure 6C), CD117, and epithelial membrane antigen (EMA). Triple markers of estrogen receptor (ER), progesterone receptor (PR), and human epidermal growth factor receptor2 (HER2) were negative. The Ki-67 proliferation index was low (less than 17%). There was no lymphovascular or perineural invasion. There was no tumor metastasis in sentinel lymph nodes.

FINAL DIAGNOSIS

The lesion was confirmed as adenoid cystic carcinoma arising in microglandular adenosis.

TREATMENT

After the breast-conserving surgery and sentinel lymph node biopsy, adjuvant radiation therapy was performed at the site of excision.

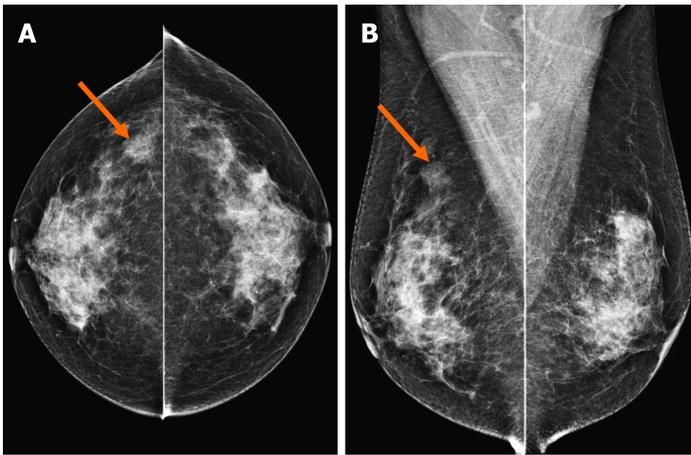


Figure 1 Mammography showing a focal asymmetry. On cranio-caudal (A) and medio-lateral-oblique (B) view, the lesion was located in the right upper outer peripheral breast (arrows). The density was similar to or slightly lower than that of the breast parenchyma. There were no associated parenchymal distortions or microcalcifications.

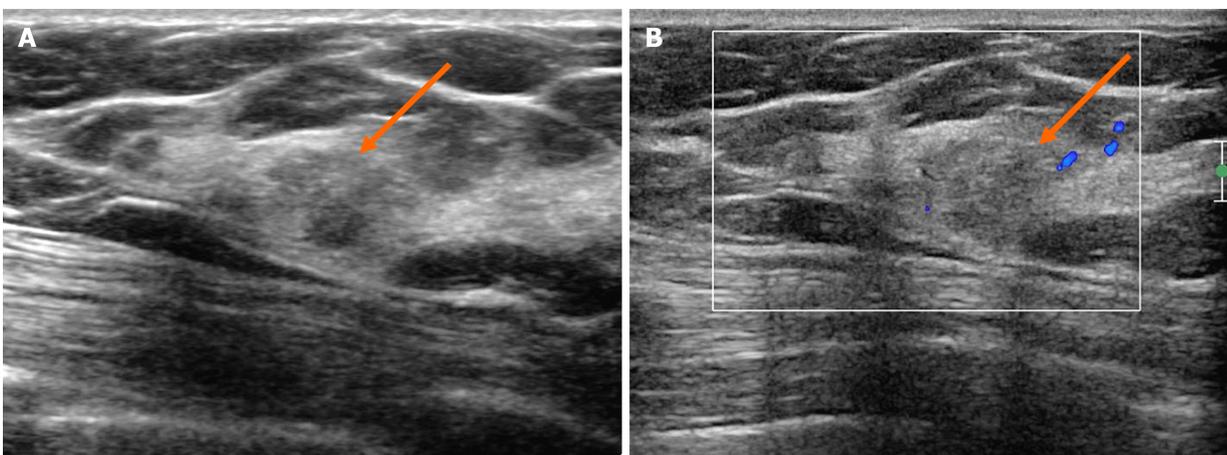


Figure 2 Ultrasonography showing an irregular mass. A: On B-mode ultrasonography, the lesion showed an irregular shape and angular margin (arrow). The echogenicity was slightly higher than that of fat; B: In the color Doppler study, there was no internal or rim vascularity (arrow).

OUTCOME AND FOLLOW-UP

The patient had the first follow-up six months after surgery. There were no specific abnormalities other than postoperative changes.

DISCUSSION

Breast AdCC is classified into three subtypes: classic AdCC, solid-basaloid AdCC (SB-AdCC), and AdCC with high-grade transformation[2]. Classic AdCC shows a central cribriform area and a peripheral area with a tubular architecture. Both areas have the same cellular compositions of epithelial and myoepithelial cells. The glandular space is lined by epithelial-type cells that produce mucins. The pseudolumina is filled with stromal matrix, including basal membranes. Classic AdCC lacks nuclear atypia and necrosis. It shows a low mitotic count. An *in situ* component is rarely seen in classic AdCC. In addition to classic features of AdCC, SB-AdCC is characterized by solid nests composed of basaloid cells with marked nuclear atypia, high mitotic count, and necrosis[16]. Breast AdCC associated with high grade carcinoma is rare. It is classified into AdCC with high-grade transformation[2]. Classic AdCC shows favorable behavior. SB-AdCC and AdCC with high-grade transformation show worse prognosis than classic AdCC[2]. In immunohistochemistry, epithelial cells are positive for CK7, CK8, and EMA. In some cases, they are positive for CK5/6[17]. Myoepithelial cells show positivity for CK14, CK5/6, P63[18], and myoepithelial markers such as heavy-

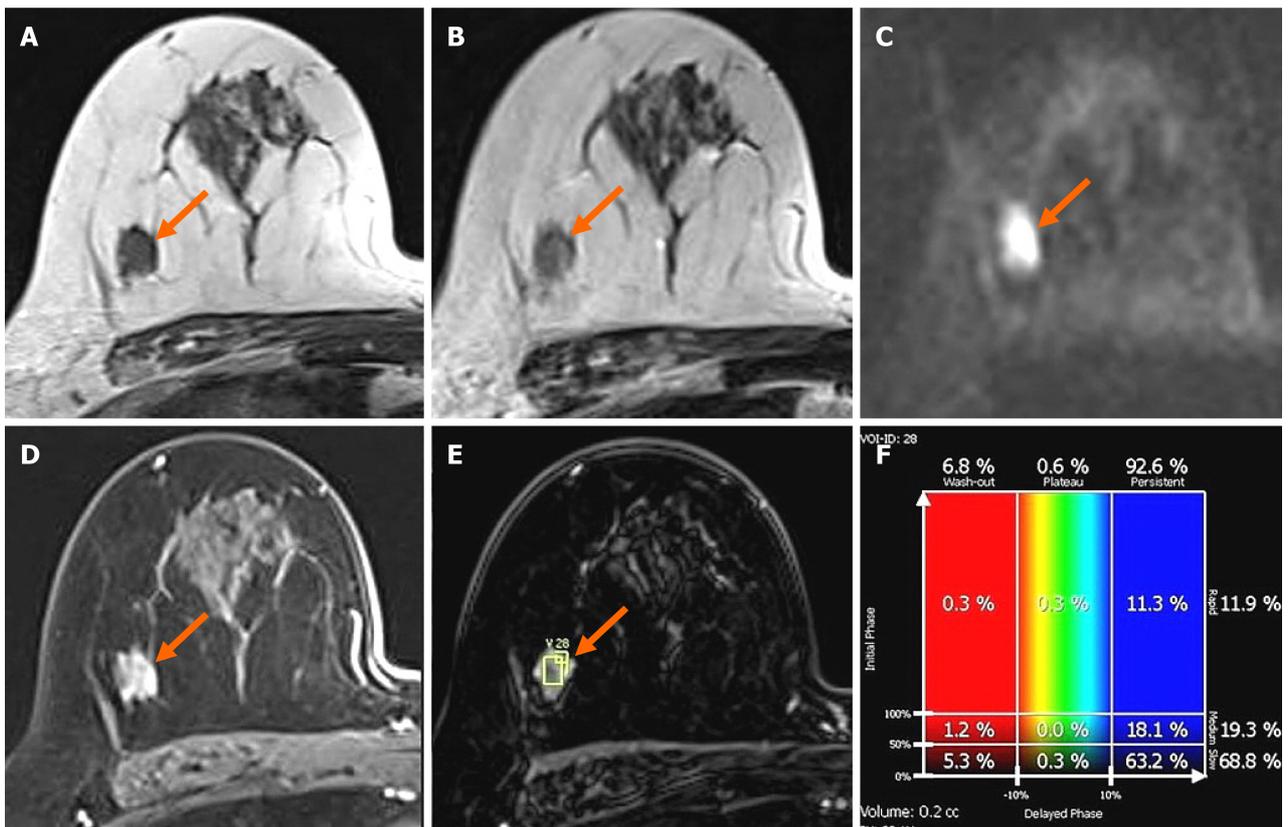


Figure 3 Breast magnetic resonance imaging showing an irregular mass in right breast. In breast magnetic resonance imaging, the lesion showed similar T1 signal intensity (A) and slightly higher T2 signal intensity (B) compared to muscles or the breast parenchyma. The lesion showed reduced diffusivity with hyperintensity in diffusion-weighted images (C). In the contrast enhancement study, the lesion showed heterogeneous internal enhancement (D) with an initial slow and delayed persistent enhancing pattern (E and F).

chain myosin, calponin, S100, and CD10. CD117 is usually strongly positive in the luminal component of AdCC. The tubular component of classic AdCC, unlike MGA, is composed of two cell types: epithelial cells and myoepithelial cells. Tubules contain mucins and basement membrane materials.

MGA has the following histologic findings: A single layer of cuboidal epithelium, clear to eosinophilic cytoplasm, rare mitotic nuclei figures, and well-preserved gland lumen and basement membranes[8,13,14,19]. Atypical MGA has features of architectural complexity, cellular expansion in the lumina, prominent nucleoli, and vesicular nuclei. *In situ* carcinoma from MGA shows frequent mitotic figures, obvious cytologic atypia, and obliteration of the gland lumen. Invasive carcinoma arising in MGA usually forms a microscopic solid tumor mass with infiltrative growth, desmoplastic reaction, severe cytologic atypia, and increased mitosis[13-15]. MGA, atypical MGA, and carcinoma from MGA have common immunohistochemical profiles of ER, PR, HER2, and CK5/6 negativity but CK8/18, S-100, and epithelial growth factor receptor positivity[11,13,15]. Khalifeh *et al*[11] have reported that Ki 67 and P53 expression could be reliable markers to distinguish MGA from atypical MGA or carcinoma from MGA because their positivity tends to increase as the lesion progresses to malignancy.

Acs *et al*[15] have reported 17 cases of AdCC coexisting with MGA. They suggested that AdCC might develop in a background of MGA or from MGA. They described morphological spectra of lesions with MGA, atypical MGA (also called AdCC *in situ*), and invasive AdCC. Altered myoepithelial cells appear to be the major neoplastic element in both AdCC and atypical MGA. Cells were stained similarly to what was seen in the myoepithelium of the breast with three markers: S-100 protein, SMA, and vimentin. MGA, atypical MGA, and AdCC show similar growth patterns with expansile or infiltrative patterns. They have a very limited metastatic capacity, showing a favorable prognosis. Rico *et al*[12] have reported that among carcinomas arising in MGA, not otherwise specified ductal carcinoma, metaplastic carcinoma, and salivary gland type carcinoma account for 63.4%, 35.2%, and 27.4%, respectively. Among salivary gland type carcinomas, AdCC was the most common carcinoma arising in MGA.

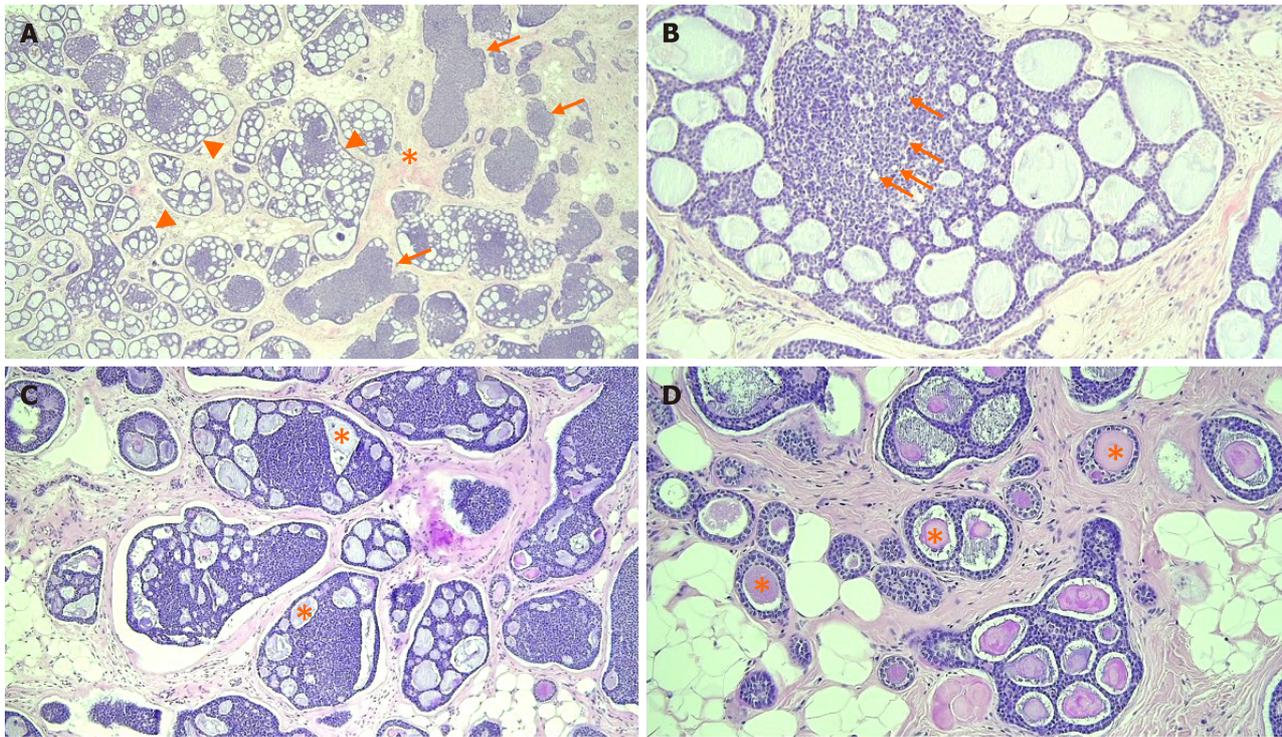


Figure 4 Photomicrographs of the adenoid cystic carcinoma. A: The tumor was composed of cribriform (arrowheads) and solid nests (arrows) in the fibrous stroma (asterisk), [hematoxylin and eosin (H&E) staining, magnification: 40 ×]; B: Myoepithelial cells formed cribriform or solid basaloid growth, whereas ductal epithelial cells were lining small ductule-like lumen (arrows) (H&E staining, magnification: 200 ×); C and D: The cribriform space contained Periodic acid Schiff positive basement membrane-like materials (asterisk) in invasive (H&E staining, magnification: 100 ×) and *in situ* components (H&E staining, magnification: 200 ×).

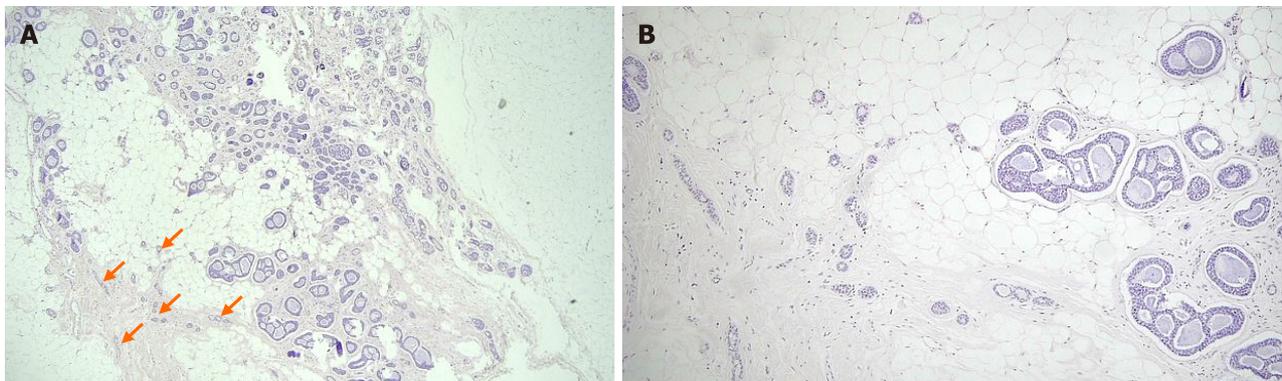


Figure 5 Photomicrographs of *in situ* adenoid cystic carcinoma and microglandular adenosis. A: *In situ* adenoid cystic carcinoma haphazardly scattered in fat tissue, showing cribriform or tubular proliferation of tumor cells. Just next to the *in situ* components, remaining microglandular adenosis (MGA) showed small tubular glands (arrows) [hematoxylin and eosin (H&E) staining, magnification: 40 ×]; B: *In situ* adenoid cystic carcinoma was noted in the right. MGA showed small tubular glands with single epithelial lining and retained lumen in the left (H&E staining, magnification: 100 ×).

In our case, the tumor was composed of invasive AdCC, *in situ* AdCC, and MGA. Invasive AdCC showed cribriform or solid sheets composed of two cell types in the desmoplastic stroma, which was a differentiating point with *in situ* AdCC. Loss of basement membrane is also a characteristic of invasive cancer that could be distinguished from *in situ* cancer. Laminin or collagen IV staining could be helpful for the differentiation. *In situ* or atypical AdCC showed haphazardly scattered small cribriform or tubular tumor glands in fat tissue in the same pattern as MGA. Just next to the *in situ* components, small tubular glands with single epithelial lining and retained lumen were present, suggesting remaining MGA. In this area, S-100 staining was strongly positive, but p63 negative, compared to S-100 negative and p63 positive of *in situ* or invasive AdCC.

In the past, we have reported that ductal carcinoma *in situ* (DCIS) arising in MGA [20]. However, not many reports on imaging findings of carcinoma arising in MGA

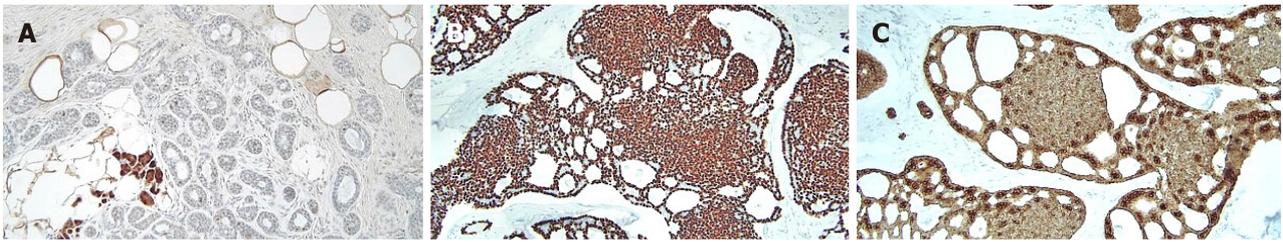


Figure 6 Immunohistochemical staining. A: *In situ* adenoid cystic carcinoma showed S-100 negativity, whereas microglandular adenosis was strongly positive for S-100 staining; B: Immunohistochemical staining for p63 showed diffuse myoepithelial differentiation of the tumor cells; C: Immunohistochemical staining for cytokeratin showed epithelial cell differentiation of the tumor cells. Magnification: 200 ×.

have been published since then. Choi and Bae[10] have reported multifocal invasive carcinoma associated with DCIS and invasive ductal carcinoma associated with encapsulated papillary carcinoma, both arising from MGA. The former showed a small nodular density in the MG and a suspicious, small hypoechoic lesion on US. The latter showed a lobulated, heterogeneous hypoechoic mass on US. Oh *et al*[21] have reported one case of invasive carcinoma arising in MGA. Mammogram showed an irregular isodense mass with a spiculated margin and irregular hypoechoic mass in US examination. Breast MRI showed an irregular mass with heterogeneous enhancement with a delayed washout kinetic curve. Adjacent segmental non-mass enhancement was confirmed as MGA and atypical MGA.

Imaging findings of AdCC of the breast are very diverse. In the MG, AdCC shows irregularly shaped, circumscribed, ill-defined, or microlobulated margins with iso- or hyper-density. It also appears as an architectural distortion or asymmetry[22-26]. In the US examination, it shows an irregular shape, circumscribed or ill-defined margins, and iso-, hypo- or complex echogenicity[22,23,25,27,28]. In MR images, it shows circumscribed round mass with intermediate high T2 SI and persistent enhancing curve[23], increased T2 SI with variable kinetic curves[22], spicules and early phase enhancement[28], and mixed cystic and solid mass with irregular shape and margin, and rapid and heterogeneous enhancement[25]. However, imaging findings of AdCC arising in MGA have not been described yet. To the best of our knowledge, this is the first report of imaging findings of AdCC arising in MGA.

In our case, in the MG, the lesion had no common invasive cancer features such as high density, distortion, spiculation, or suspicious microcalcifications, except for developing density. Therefore, it would be missed unless comparison with previous mammographic images was made. The density was similar to that of the breast parenchyma or slightly less. On US, sonographic echo of the lesion was slightly higher than that of fat. As the contrast between the hyperechoic breast tissue and the lesion was unclear, it was difficult to clearly distinguish the margin of the lesion in B-mode ultrasound, even with a harmonic image. Therefore, our sonographic diagnosis was a low-suspicion lesion with the possibility of a benign lesion, not breast cancer. In breast MRI, a slightly increased T2 signal intensity and a persistent enhancing pattern were seen, similar to other reports on MRI findings of AdCC. MGA might also show imaging findings similar to breast malignancy[29]. Thus, it is currently unclear which characteristics of MGA can affect imaging findings of AdCC arising in MGA. Further case collections and analysis are needed in the future to characterize this rare lesion.

For the treatment of primary AdCC of the breast, Treitl *et al*[1] have reported that radiation therapy or sentinel node excision besides a primary surgical therapy might not be warranted due to the tumor's indolent course. Kulkarni *et al*[30] have reported that AdCC has characteristics of lower grade, hormone receptor negativity, and node negativity compared to an invasive ductal carcinoma. Therefore, AdCC was treated with less axillary surgery, fewer mastectomy, and less chemotherapy and hormone therapy. However, there is no treatment policy for AdCC arising in MGA due to the rarity of this type of malignancy. Although many published studies have suggested a relatively favorable prognosis for carcinomas arising in MGA, MGA may no longer be considered simply a benign proliferative lesion as it can progress to aggressive breast cancer. Resetkova *et al*[31] have emphasized the importance of complete excision of the mass and reported a recurrent case of high-grade *in situ* carcinoma within the residual MGA after primary surgery for this invasive tumor arising in atypical MGA. Therefore, appropriate treatment should be performed according to the patient's clinical status considering the general treatment strategy for invasive cancer with complete excision of MGA.

CONCLUSION

AdCC is a rare breast malignancy with a favorable prognosis. Although MGA has long been considered a benign entity, it has the risk of progression to malignancy. Therefore, in the case of AdCC arising in MGA, an appropriate treatment should be performed according to the clinical stage of the patient. In particular, complete resection should be performed so that no MGA remains. Imaging findings of AdCC occurring in MGA were characterized by iso- or slight hypo-density in the MG and slightly higher echo than that of fat in the US examination with higher T2 signal intensity and a persistent enhancing pattern in breast MRI.

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