Case Report

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Radiologic Findings of Mesonephric-Like Adenocarcinoma of the Uterine Corpus: A Case Report 자궁 체부에 발생한 중신 유사 선종의 영상 소견: 증례 보고

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According to the 2020 World Health Organization classification, mesonephric-like adenocarcinoma (MLA) is newly categorized as a subtype of endometrial carcinoma and remains a relatively unknown disease owing to its rarity. To the best of our knowledge, radiological findings of MLA have not been reported in the English literature. The uterine MLAs show a worse clinical prognosis and a more aggressive biological behavior than the usual endometrial carcinoma. Herein, we present the imaging findings of a 65-year-old female with a MLA in the uterine corpus. The tumor was a solid endometrial mass with deep myometrial invasion, poor contrast enhancement, and moderate diffusion restriction.

Index terms Neoplasm; Uterus; Computed Tomography, X-Ray; Magnetic Resonance Imaging

INTRODUCTION

Mesonephric-like adenocarcinoma (MLA) was described as a new subtype of endometrial carcinoma in the 2020 World Health Organization classification of female genital tumors (1). MLA comprises 1% of all endometrial carcinomas and shares similar morphological, molecular, and immunophenotypic characteristics with mesonephric adenocarcinoma arising in the uterine cervix (2). MLA is a more aggressive subtype of endometrial cancer than the com-

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This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (https://creativecommons.org/ licenses/by-nc/4.0) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited. mon subtypes, with a propensity for early recurrence and distant metastases (3). Several case studies have reported the prognosis and clinicopathological findings of MLAs (2-5). However, to the best of our knowledge, imaging features of MLAs in the uterine corpus have seldom been reported. Here, we report a surgically confirmed case of MLA, focusing on its radiologic findings and clinicopathological features.

CASE REPORT

A 65-year-old female, gravida 2 and para 2, presented to the gynecology department, complaining of dysfunctional uterine bleeding for 1 month. The mean age at menopause onset was 56 years. She had never had a Pap-smear before and had no recorded medical history. Physical examination revealed a soft abdomen with no palpable masses. Laboratory findings, including cancer antigen (CA) 125, CA 19-9, urine analysis and complete blood count, were normal.

Initial contrast-enhanced CT revealed a heterogeneously hypodense mass of 5 cm on the left side of the uterine body (Fig. 1A). There was no evidence of ascites, lymph node enlargement, or distant metastases. Pelvic MRI was performed to further evaluate the uterine mass. Pelvic MRI revealed a lobulated mass with intermediate signal intensity on T2-weighted imaging, involving both the endometrium and myometrium (Fig. 1B). The mass showed less enhancement than the myometrium on contrast-enhanced T1-weighted imaging (Fig. 1C). The mass showed high signal intensity on diffusion-weighted imaging and low signal intensity on the apparent diffusion coefficient map (Fig. 1D). Based on CT and MRI findings, the preoperative diagnosis included advanced endometrial cancer with deep myometrial invasion and uterine sarcomas.

The patient underwent a total hysterectomy, bilateral salpingo-oophorectomy, bilateral pelvic lymphadenectomy, and para-aortic lymphadenectomy. On the cut section, the tumor was a pale yellowish solid mass involving the endometrium and the deep myometrium (Fig. 1E). Microscopically, the mass was composed of a compact proliferation of small glands and tubules containing intraluminal eosinophilic secretions and was negative for the estrogen receptor (ER) and progesterone receptor (PR) (Fig. 1E). Immunoreactivity for GATA-binding protein 3 (GATA3) (Fig. 1E) and thyroid transcription factor-1 (TTF-1) was observed. The final pathologic diagnosis was MLA of the uterine corpus with International Federation of Gynecology and Obstetrics (FIGO) stage IB. Therefore, postoperative adjuvant chemotherapy with paclitaxel and carboplatin has been administered three times to date. The patient is being seen regularly in the outpatient clinic without evidence of recurrence or metastasis 18 months after surgery.

This study was approved by the Institutional Review Board of our institution (IRB No. 2022GR0331). Informed consent was waived due to the retrospective nature of the study.

DISCUSSION

Mesonephric adenocarcinoma is a rare malignancy that most frequently occurs in the uterine cervix and is thought to arise from embryonic remnants of the mesonephric ducts and tubules (5). MLA is a recently defined rare tumor that occurs in the uterine corpus and ovaries.

Mesonephric-Like Adenocarcinoma of the Uterine Corpus

Fig. 1. A 65-year-old female with mesonephric-like adenocarcinoma of uterine corpus.

A. Axial contrast-enhanced CT image shows a heterogeneous hypodense mass lesion (arrows) of approximately 5 cm on the left side of uterine body.

B. Axial and sagittal T2WI demonstrate a lobulating mass with intermediate SI (arrows) involving both the endometrium and myometrium.

C. Axial fat saturated T1WI (right) and axial fat saturated CET1 (left) demonstrate lesser enhancement of the lesion (arrows) than the myometrium.

CET1 = contrast-enhanced T1WI, SI = signal intensity, T1WI = T1-weighted imaging, T2WI = T2-weighted imaging





Fig. 1. A 65-year-old female with mesonephric-like adenocarcinoma of uterine corpus.

D. High b-value (b = 1000) DWI (right) demonstrates high SI (arrows) with low SI (arrows) in the ADC map (left) indicating moderate diffusion restriction.

E. Gross specimen shows a pale yellowish solid mass (arrows) of uterus. The mass originated from the endometrium, invading the myometrium. The microscopic image with H&E stain (\times 400) shows a variety of architectural patterns with intraluminal eosinophilic secretions. On immunohistochemistry, the tumor cells are negative for ER (\times 200) and demonstrate a diffuse GATA3 (\times 100) positivity.

ADC = apparent diffusion coefficient, DWI = diffusion-weighted imaging, ER = estrogen receptor, GATA3 = GATA-binding protein 3, H&E = hematoxylin and eosin, SI = signal intensity



MLA has morphological, immunophenotypic, and molecular features similar to those of mesonephric adenocarcinoma. Despite the similarities between MLA and mesonephric adenocarcinoma, it is still uncertain whether MLAs originate from mesonephric remnants or from Mullerian neoplasms, such as endometriosis and adenomyosis (2). Due to these histogenetic uncertainties, the term "MLA" has been used (6).

The pathologic diagnosis of MLA can be challenging because of their rarity and various

histological patterns. Endometrioid endometrial carcinoma (EEC) with tubular and glandular growth pattern is the primary differential diagnosis (7). An important diagnostic clue for MLA is the presence of eosinophilic secretions in the tubules. Additional immunohistochemical staining can be performed when this morphology is observed. Immunohistochemically, EECs are positive for ER and PR, whereas they are almost always absent in the MLA. GATA3 and TTF are useful markers for mesonephric remnants and hyperplasia. Therefore, MLA also shows positive staining for GATA3 and/or TTF-1 (6, 7).

MLAs are rare tumors of the female genital tract, comprising 1% of all endometrial carcinomas (2). In the literature, the mean age of the patients was 61 years (range, 26–91 years) (2). In our case, the patient was 65 years old. Patients with MLA had nonspecific clinical presentation. In a few reported cases, the patients were mostly asymptomatic, with other symptoms including vaginal bleeding, vaginal discharge, and abdominal pain or discomfort (5). In the present case, the patient complained of vaginal bleeding.

Although the radiologic findings of endometrial carcinomas are well known, the specific details and distinctions in the radiologic features of each subtype remain unclear. Furthermore, imaging features of the MLA in the uterine corpus are not well established, and only clinical and histopathological characteristics have been reported in previous studies (2-5). Advanced endometrial carcinoma is usually seen as a solid endometrial mass with deep myometrial invasion, less enhancement than the myometrium, and moderate diffusion restriction (8). Our case was also depicted as an advanced endometrial carcinoma. Na and Kim (4) reported gross findings of uterine MLA in 11 patients. Three types were observed: tumors protruding into the endometrial cavity but did not clearly invade the myometrium, tumors with deep infiltration into the myometrium like intramural leiomyoma, and tumors infiltrating the entire uterine wall. In our case, the tumor had infiltrated the entire uterine wall.

The differential imaging diagnosis for MLA includes uterine sarcomas, such as leiomyosarcoma, endometrial stromal sarcoma, and undifferentiated endometrial sarcoma. Uterine sarcomas usually show more contrast enhancement than endometrial carcinomas, which is helpful for distinguishing these tumors (9).

MLA is biologically more aggressive than other common types of endometrial carcinomas. MLA is considered a high-grade carcinoma, although it has confusingly low-grade morphology (3). Xie et al. (5) reported that 82% of patients presented with FIGO stage IB at initial diagnosis. Euscher et al. (3) reported that 48% of patients were at FIGO stage III or IV disease. Thus, uterine MLA typically presents as an advanced-stage disease at initial diagnosis. In contrast, other common types of endometrial cancers are diagnosed at an early stage (61% in stage IA) (10). The patient was diagnosed with FIGO stage IB disease. Furthermore, MLA tend to recur early and metastasize to distant organs, especially to the lungs (2).

Due to the limited data, no standard treatment guideline exists for uterine MLA, including surgery and chemotherapy. In recent studies, patients underwent hysterectomy with bilateral salpingo-oophorectomy (3). Current studies have also suggested that combination chemotherapy with carboplatin and paclitaxel has a good response to MLA (2).

In conclusion, we present a rare case of MLA in the uterine corpus. Our case revealed an endometrial mass with deep myometrial invasion, poor enhancement and moderate diffusion restriction on CT and MRI; with these findings, we considered advanced endometrial carcinoma. Clinically, uterine MLAs usually present as advanced-stage tumors that frequently recur and metastasize to the lungs. Therefore, familiarization with the radiologic findings and clinicopathologic features of this rare tumor may be helpful for the management and treatment of patients with MLAs.

Author Contributions

Conceptualization, K.H.J., K.K.A.; data curation, C.Y., K.J.W., L.J.; formal analysis, K.H.J., K.K.A., L.C.H.; investigation, K.H.J., K.K.A., L.C.H.; project administration, K.K.A.; resources, L.J.; validation, K.H.J., K.J.W.; visualization, K.H.J.; writing—original draft, K.H.J.; and writing—review & editing, K.K.A.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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REFERENCES

- McCluggage WG. Progress in the pathological arena of gynecological cancers. Int J Gynaecol Obstet 2021; 155(Suppl 1):107-114
- Deolet E, Van Dorpe J, Van de Vijver K. Mesonephric-like adenocarcinoma of the endometrium: diagnostic advances to spot this wolf in sheep's clothing. A review of the literature. J Clin Med 2021;10:698
- 3. Euscher ED, Bassett R, Duose DY, Lan C, Wistuba I, Ramondetta L, et al. Mesonephric-like carcinoma of the endometrium: a subset of endometrial carcinoma with an aggressive behavior. *Am J Surg Pathol* 2020;44: 429-443
- 4. Na K, Kim HS. Clinicopathologic and molecular characteristics of mesonephric adenocarcinoma arising from the uterine body. *Am J Surg Pathol* 2019;43:12-25
- 5. Xie C, Chen Q, Shen Y. Mesonephric adenocarcinomas in female genital tract: a case series. *Medicine (Baltimore)* 2021;100:e27174
- McFarland M, Quick CM, McCluggage WG. Hormone receptor-negative, thyroid transcription factor 1-positive uterine and ovarian adenocarcinomas: report of a series of mesonephric-like adenocarcinomas. *Histopathology* 2016;68:1013-1020
- 7. Koh HH, Park E, Kim HS. Mesonephric-like adenocarcinoma of the ovary: clinicopathological and molecular characteristics. *Diagnostics (Basel)* 2022;12:326
- Faria SC, Sagebiel T, Balachandran A, Devine C, Lal C, Bhosale PR. Imaging in endometrial carcinoma. Indian J Radiol Imaging 2015;25:137-147
- 9. Santos P, Cunha TM. Uterine sarcomas: clinical presentation and MRI features. *Diagn Interv Radiol* 2015; 21:4-9
- 10. Trojano G, Olivieri C, Tinelli R, Damiani GR, Pellegrino A, Cicinelli E. Conservative treatment in early stage endometrial cancer: a review. *Acta Biomed* 2019;90:405-410

자궁 체부에 발생한 중신 유사 선종의 영상 소견: 증례 보고

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중신 유사 선종은 2020년 World Health Organization 분류체계에서 자궁내막암의 한 아형 으로 새롭게 분류되었고, 드물기 때문에 잘 알려지지 않은 질환이다. 저자들이 아는 한, 영어 문헌에서 영상의학적 소견은 아직 보고된 바 없다. 자궁의 중신 유사 선종은 일반적인 자궁 내막암에 비해 예후가 좋지 않고 더 공격적인 생물학적 양상을 보인다. 저자들은 65세 여성 의 자궁 체부에 생긴 중신 유사 선종의 영상의학적 소견에 대하여 보고하고자 한다. 자궁내 막 고형 종괴가 심부 자궁근 침범을 보였고, 조영 증강은 잘되지 않았고 중등도의 확산제한 을 보였다.

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