Magnetic resonance imaging findings of endosalpingiosis: a case report

Shinya Fujii1, Chie Inoue2, Naoko Mukuda1, Atsushi Murakami1, Daisuke Yamaji1, Hiroto Yunaga1 and Kanae Nosaka3

Abstract
Endosalpingiosis is characterized by the presence of glands lined by benign tubal-type epithelium outside the fallopian tube. It is usually an incidental finding and rarely occurs as a tumor-like mass lesion. Here, we describe the magnetic resonance imaging findings of endosalpingiosis that presented as a paraovarian multicystic lesion. It exhibited iso to low intensity on T1-weighted images and inhomogeneous high intensity on T2-weighted images. The septa presented relatively iso to slight high intensity on T2-weighted images and strong contrast enhancement on dynamic contrast-enhanced imaging. Endosalpingiosis should be considered as a differential diagnosis in cases of paraovarian multicystic lesions along the uterine serosa.

Keywords
Magnetic resonance imaging, endosalpingiosis, uterus, uterine

Introduction
Endosalpingiosis is a disease characterized by the presence of glands lined by benign tubal type of epithelium outside the fallopian tube.1 Endosalpingiosis occurs in as many as 12.5% of women, with a wide age range1 in both premenopausal and postmenopausal women.2 It is usually an incidental finding,1 and this condition rarely occurs as a tumor-like mass lesion. Herein, we present the magnetic resonance (MR) imaging findings of pelvic endosalpingiosis.

Case report
A 37-year-old woman complained of lower abdominal pain and visited a hospital where a left adnexal mass was identified, after which she was referred to our hospital for further examination. Her CA 125 level was slightly elevated to 36.2 U/mL.

MR imaging revealed a left paraovarian multicystic lesion along the uterine serosal surface, which exhibited iso to low intensity on T1-weighted images (WI) and inhomogeneous high intensity on T2WI (Fig. 1(a) and (b)). The septa showed iso to slight high intensity on T2WI and strong contrast enhancement on dynamic contrast-enhanced imaging (Fig. 1(c) and (d)). The septa of the lesion demonstrated high intensity on diffusion-WI (DWI) with b value of 1000 and showed relatively high intensity on the apparent diffusion coefficient (ADC) map (Fig. 1(e) and (f)). The lesion was adjacent to the left ovary (Fig. 1(b)). The lesion showed a multilocular cystic mass without calcification on computed tomography (CT). Preoperatively, she was diagnosed with a left ovarian mucinous borderline tumor.

The lesion was considered a subserosal uterine tumor during the operation. Consequently, a...
hysterectomy and left salpingo-oophorectomy were performed. On gross examination, a mass with a smooth surface was adherent to the uterus (Fig. 2(a)). Microscopically, the multilocular cystic mass was located beneath the uterine serosa and slightly invaginating the myometrium. Similar multicystic lesions were found in the uterine subserosa and myometrium (Fig. 2(b)). The lesion in the myometrium was composed of dilated glands surrounded by stromal cells, analogous to endometriosis (Fig. 2(c)). Cysts were covered by a single-layered epithelium, including cuboidal to columnar epithelium, with or without cilia. The cyst walls were composed of loose spindle stromal cells, such as myofibroblasts, similar to the endosalpinx stroma (Fig. 2(d)). The lesion was pathologically diagnosed as endosalpingiosis.

**Discussion**

Endosalpingiosis was first reported by Sampson in 1930 and is characterized by the presence of glands lined by benign tubal-type epithelium outside the fallopian tube. Regarding clinical presentation, 44.5% of the patients present with chronic pelvic pain. In premenopausal patients, 62.7% of the patients present abnormal uterine bleeding, whereas, in premenopausal patients, 45.5% of the patients present with postmenopausal bleeding. Although the etiology of endosalpingiosis is still unclear, celomic metaplasia, implantation during surgery, and müllerianosis are considered.

Endosalpingiosis is most commonly seen along the pelvic organs, such as the uterine serosa, fallopian tube serosa, ovaries, round ligament, and bladder. In the present case, the lesion was found along the uterine serosa. Additionally, it also occurs in the retroperitoneal lymph nodes and appendix. Endosalpingiosis is a potential risk factor for the development of serous borderline ovarian tumors.

Imaging findings of mass-forming pelvic endosalpingiosis are limited. According to previous reports, the lesions show a multilocular cystic mass in the uterine myometrium or along the uterine serosa with infiltration into the uterus. It can be demonstrated as a unilocular cystic or diffusely infiltrating solid and cystic masses. Calcification can be observed as multiple calcified granular nodules in the pelvic peritoneal cavity on CT. Regarding the imaging findings in extrapelvic sites, appendiceal and splenic endosalpingiosis have been reported. In appendices, the lesions showed thin-walled cystic masses. Splenic endosalpingiosis showed a multiloculated cystic lesion with scattered wall calcification. Thus, we should consider the possibility of endosalpingiosis in the case of a paraovarian multicystic mass along the uterine serosa as well as in the other abdominal regions.

To the best of our knowledge, DWI findings of endosalpingiosis have not been reported. In the present...
case, the septa of the lesion demonstrated high intensity with a relatively high ADC value. Consequently, the high intensity on DWI is suggested to be due to T2 shine through effect. The high ADC value is considered to reflect the benign nature of the lesion.

Differential diagnoses include uterine subserosal leiomyoma (LM) with cystic degeneration, adenomatoid tumors, ovarian mucinous cystic tumors, and peritoneal cystic tumors, such as multicystic mesothelioma. Cystic degeneration of LM is observed in approximately 4% of LMs. LM with cystic degeneration demonstrates a low-intensity mass accompanied by large or small, well-demarcated cystic spaces showing the signal intensity of serous fluid on T2WI. Adenomatoid tumors are rare benign tumors of mesothelial origin. It is usually very small but is rarely identified on MR imaging. The MR imaging findings are highly similar to those of LMs. However, adenomatoid tumors can show multilocular cystic features. In the present case, the septal parts showed a relatively high intensity on T2WI. However, the septal parts of endosalpingiosis demonstrated low intensity on T2WI in some case reports. Thus, it is occasionally difficult to differentiate these tumors from endosalpingiosis. Ovarian mucinous cystic tumors usually show a multilocular cystic mass with stained-glass appearance. Unidentification of the ipsilateral normal ovary is important for differentiation. Multicystic mesothelioma is a rare benign primary peritoneal neoplasm of mesothelial origin. It typically shows nonenhancing unilocular or multilocular thin-walled cystic lesions. The solid part in endosalpingiosis showed contrast enhancement in the present case and a reported case. Therefore, it may be possible to differentiate between them.

In conclusion, we described the MR imaging findings of a case of endosalpingiosis. Endosalpingiosis should be considered in cases of paraovarian multicystic lesions along the uterine serosa.

Acknowledgement
We thank Hiroaki Komatsu, Shinya Sato, Tetsuro Oishi, for their help in the tumor board.

Declaration of Conflicting Interests
The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.
Funding
The author(s) received no financial support for the research, authorship, and/or publication of this article.

ORCID iDs
Shinya Fujii https://orcid.org/0000-0002-0048-0396
Hiroto Yunaga https://orcid.org/0000-0001-8300-9542
Kanae Nosaka https://orcid.org/0000-0003-0344-5470

References
1. Malpica A, Baker PM, Cheung AN, et al. WHO classification of tumours editorial board. WHO classification of tumours of female reproductive organs. Lyon: International Agency for Research on Cancer, 2020, p. 204.
2. Prentice L, Stewart A, Mohiuddin S, et al. What is endosalpingiosis? Fertil Steril 2012;98:942–947.
3. Sampson J. Postsalpingectomy endometriosis (endosalpingiosis). Am J Obstet Gynecol 1930;16:443–480.
4. Nixon KE, Kenneth Schoolmeester J, Bakkum-Gamez JN. Florid cystic endosalpingiosis with uterine preservation and successful assisted reproductive therapy. Gynecol Oncol Rep 2018;25:8–10.
5. Tudor J, Williams TR, Myers DT, et al. Appendiceal endosalpingiosis: clinical presentation and imaging appearance of a rare condition of the appendix. Abdom Radiol (NY) 2019;44:3246–3251.
6. Cil AP, Atasoy P, Kara SA. Myometrial involvement of tumor-like cystic endosalpingiosis: a rare entity. Ultrasound Obstet Gynecol 2008;32:106–110.
7. Taneja S, Sidhu R, Khurana A, et al. MRI appearance of florid cystic endosalpingiosis of the uterus: a case report. Korean J Radiol 2010;11:476–479.
8. Yang M, Li Y, Chen M, et al. Uterine endosalpingiosis: case report and review of the literature. Taiwan J Obstet Gynecol 2019;58:324–327.
9. Katre R, Morani AK, Prasad SR, et al. Tumors and pseudotumors of the secondary müllerian system: review with emphasis on cross-sectional imaging findings. AJR Am J Roentgenol 2010;195:1452–1459.
10. Bazot M, Vacher Lavenu MC, Bigot JM. Imaging of endosalpingiosis. Clin Radiol 1999;54:482–485.
11. Nguyen BD, McCullough AE. Gastrointestinal: cystic endosalpingiosis of the spleen: CT, MR, and US imaging. J Gastroenterol Hepatol. 2017;32:1911.
12. Ueda H, Togashi K, Konishi I, et al. Unusual appearances of uterine leiomyomas: MR imaging findings and their histopathologic backgrounds. Radiographics 1999;19:S131–145.
13. Kim JY, Jung KJ, Sung NK, et al. Cystic adenomatoid tumor of the uterus. AJR Am J Roentgenol 2002;179:1068–1070.
14. Lee R, Tong A, Kurtis B, et al. Benign multicystic peritoneal mesothelioma: AIRP best cases in radiologic-pathologic correlation. Radiographics 2016;36:407–411.