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A Case of Bilateral Decidualized Endometriomas during Pregnancy: Radiologic-pathologic Correlation

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Clinical differentiation between decidualized endometrioma and malignant transformation still poses difficulties as both are intracystic vascularized excrescences of an endometrial cyst and exhibit similar characteristics on color-flow Doppler sonography. This is a characteristic sonographic finding associated with ovarian cancer, but MRI can provide further information about mural excrescences that can aid in their differential diagnosis; for example, the signal of decidualized endometriomas is isointense with the placenta within the uterus on all sequences and the apparent diffusion coefficient is higher than that of malignant mural nodules. Thus, MRI should be an aid in deciding whether to intervene during pregnancy. However, considering that it is not yet possible to clearly differentiate decidualized endometriomas from ovarian cancer, surgery or watchful observation may still be needed to exclude the possibility of malignancy.

INTRODUCTION

The incidence of persistent ovarian masses detected in pregnancy is estimated to be between 1% and 2% (1-3). The incidence of surgically proven endometriomas during pregnancy is typically low (11%–20%) (4,5) because endometriosis is a cause of infertility. Furthermore, decidualization of endometriomas during pregnancy appears to be very rare, with only a handful of reported cases (6-11), although specifics of its pathogenesis and frequency remain unknown. Most cases are unilateral. On imaging, decidualized endometriomas appear as solid nodules within cystic ovarian masses, mimicking malignant transformation (12). On MRI, mural nodules composed of ectopic decidua are isointense with the nomotopic endometrium within the uterus on all sequences; however, the signal intensity on each sequence can resemble that of malignant transformation. Apparent diffusion coefficient (ADC) values of the mural nodules may also be a clue to differentiate decidualized endometriomas from malignant transformation of endometriomas (11). Here we report a case of bilateral decidualized endometriomas in a woman in the 12th gestational week, in which the ADC value of mural nodules was very close to that of the placenta.

CLINICAL CASE

A 34-year-old woman (0 Para 0 Gravida) with a chief complaint of amenorrhea consulted an obstetrician and was simultaneously diagnosed as 12 weeks pregnant and with an approximately 10cm adnexal mass. She had regular 25-day menstrual cycle and no past medical history.

Transvaginal ultrasound (TVUS) revealed a monolocular cystic mass with homogeneous low-level internal echoes. There were mural excrescences within the mass suggestive of a malignant ovarian tumor. The mural excrescences were smoothly lobulated mural nodules that did not demonstrate visible internal vascularity on color and spectral Doppler images (Figure 1).

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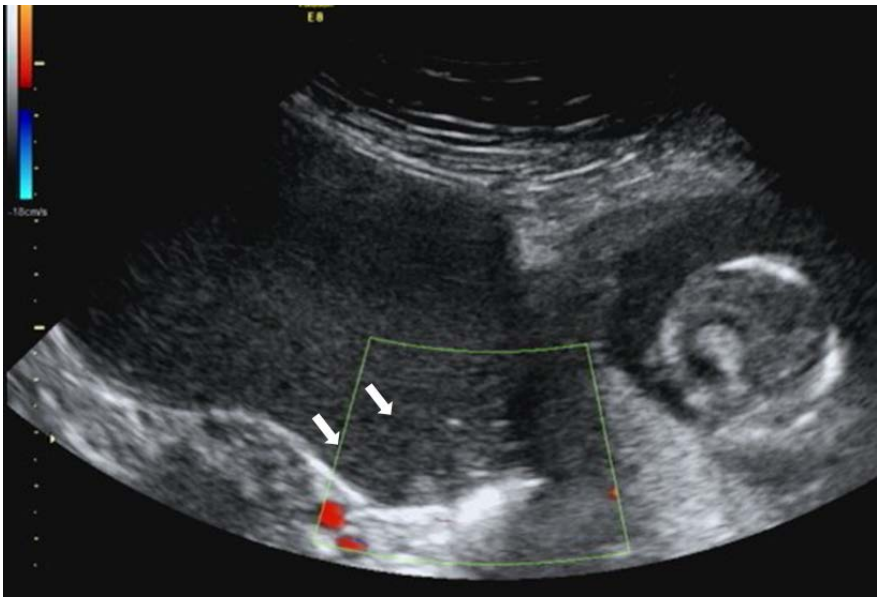

Figure 1.
Follow-up transabdominal ultrasound of left ovarian cyst at 14 weeks of gestation showed mural excrescences within the cyst. The mural excrescences were smoothly lobulated mural nodules that did not demonstrate visible internal vascularity on color and spectral Doppler images (arrows).

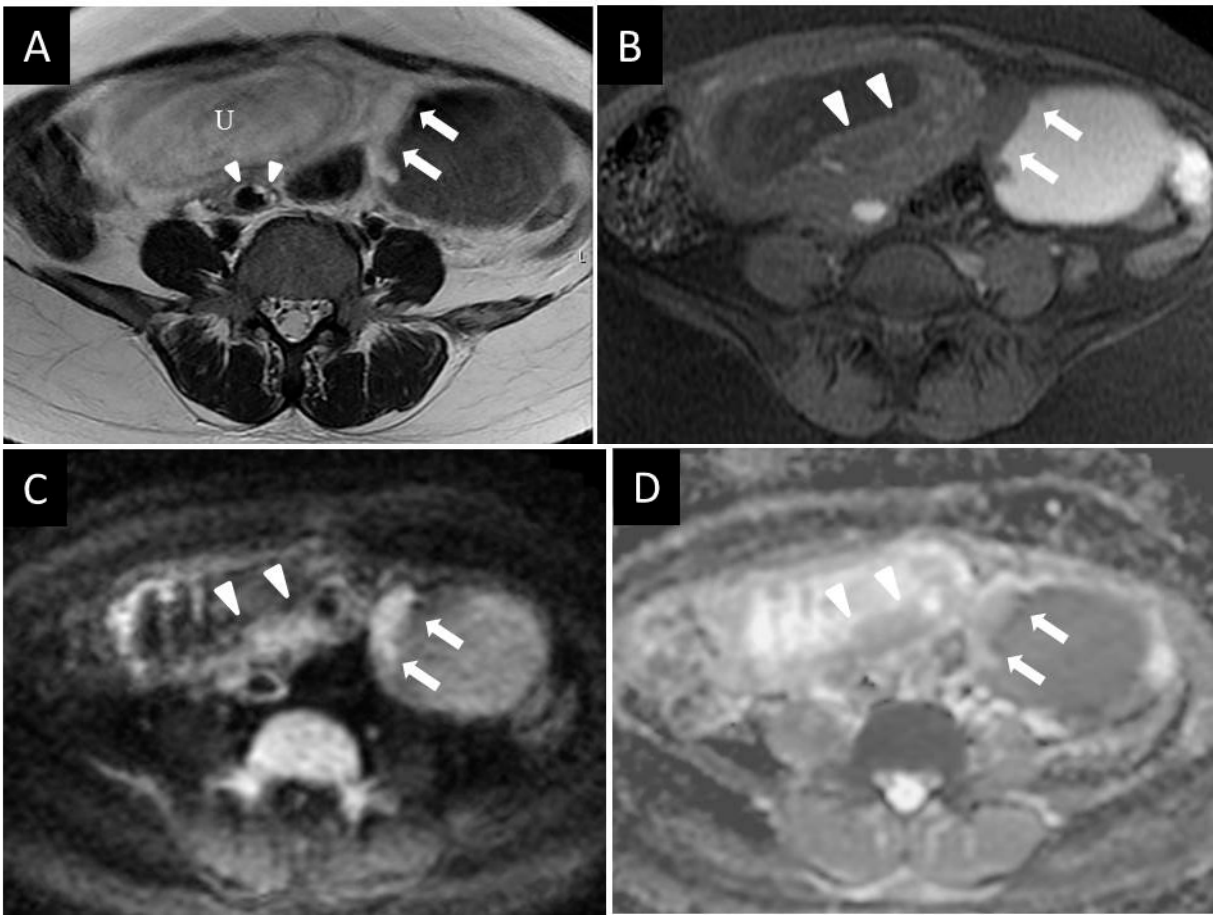

Figure 2.
A. Axial T2-weighted image. Large unilocular cystic mass including hypointense fluid with very hyperintense mural nodules (arrows) depicted on the left side of the uterus (U). A right ovarian cyst with excrescence is also shown behind the uterus (arrowheads). B. Axial fat-suppressed T1-weighted image. Cyst fluid is hyperintense and mural nodules (arrows) demonstrate low signal intensity (SI) similar to that of the placenta (arrowheads). C. Axial high b-value DWI. The mural nodules (arrows) show very high SI. The SI of mural nodules (arrows) is also similar to that of the placenta (arrowheads). D. ADC map. ADCs of the mural nodules (arrows) and placenta (arrowheads) are 2.27×10^{-3} mm^2/s and $2.24\times10^{-3}mm^2/s$, respectively.

Serum cancer antigen (CA) 125, a commonly used marker for epithelial ovarian carcinoma, was 283U/ml (normal range, 0–35 U/ml). However, this was not considered notable enough as CA125 can be elevated in non-malignant conditions such as during the first trimester of pregnancy, postpartum (13), or in endometriosis. CA19-9 was 113 U/ml (normal range, 0–37 U/ml). TVUS showed that the fetus was growing well.

A magnetic resonance (MR) examination was performed with informed consent at the 14th gestational week using a 1.5 T superconducting magnet (Signa Excite HD, GE Healthcare, Milwaukee, Wisconsin, USA). MR images depicted bilateral ovarian cystic lesions. On axial fat-suppressed T1-weighted images (T1WI), a monolocular cystic mass including hyperintense fluid was located on the left side of the uterus. The size was 10cm. There were several hypointense mural nodules and irregular wall thickening. The signal intensity (SI) of the cystic fluid was not suppressed on fat-suppressed T1WI (Fig. 2B). T2-weighted images (T2WI) revealed that the SI of the cystic fluid was much lower than that of urine, whereas the mural nodules and irregularly thickened wall had markedly high SI like the placenta on T2WI (Fig. 2A) and low SI like the placenta on fat-suppressed T1WI. On axial diffusion-weighted images (DWI), they demonstrated high SI and a high b-value (b = 800 s/mm^2) (Fig.2c), and on the apparent diffusion coefficient (ADC) map, they demonstrated slightly high SI similar to that of normal endometrium. On the ADC map, the ADCs of the mural nodules and placenta were 2.27 × 10^{-3} mm^2/s and 2.24 × 10^{-3} mm^2/s respectively (Fig.2D). A right ovarian cyst with excrescence is also shown behind the uterus (Fig. 2A; arrowheads).

A right ovarian cyst measuring 4cm was located behind the uterus and had a similar appearance on all MR sequences (Fig. 3 A-D).

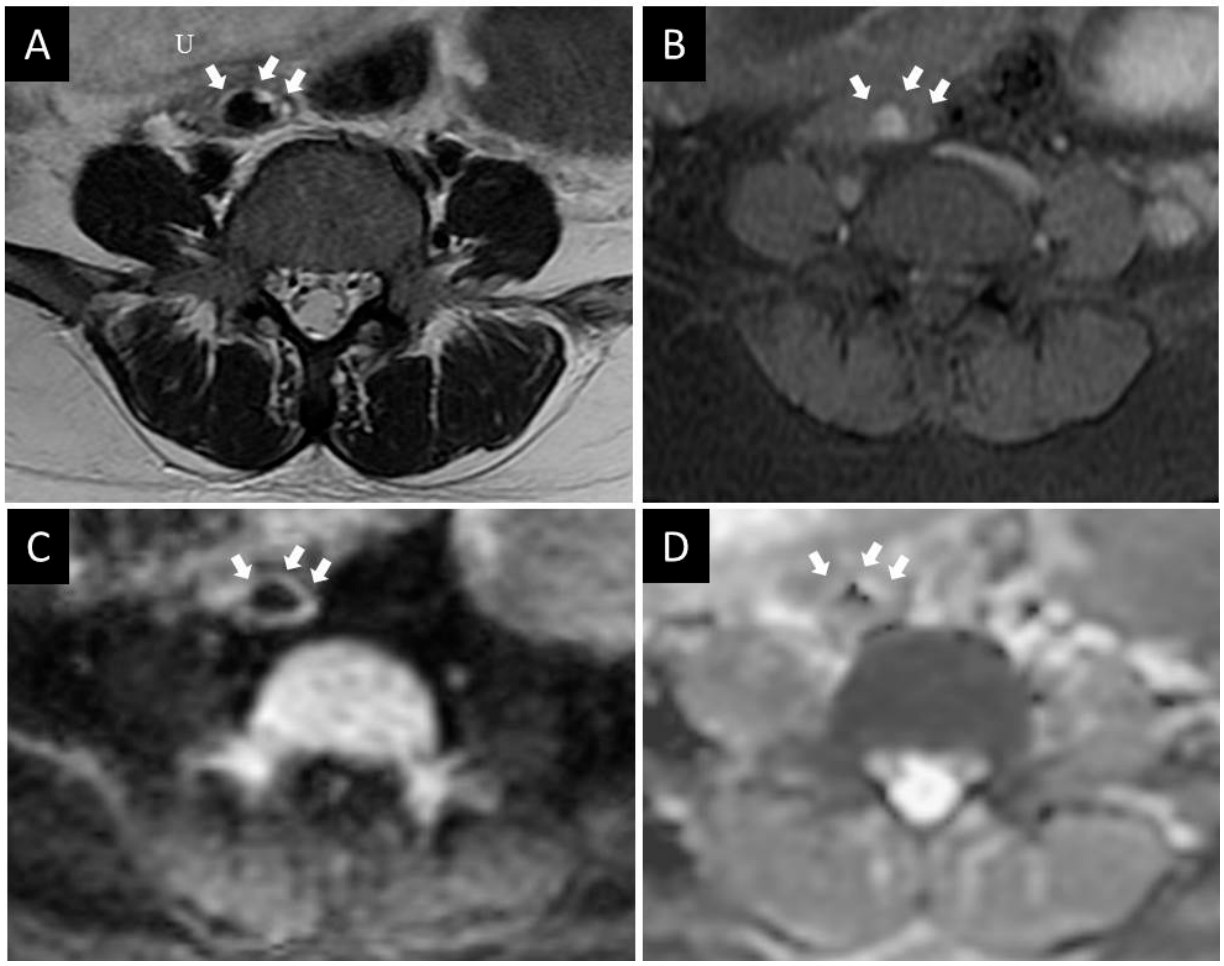

Figure 3.
A. Axial T2-weighted image. Small unilocular cystic mass including hypointense fluid with very hyperintense mural nodules (arrows) depicted behind the uterus (U). B-D. The cyst fluid shows hyperintensity on the fat-suppressed T1-weighted image. The mural nodules (arrows) show low signal intensity on the fat-suppressed T1-weighted image (B) and high signal intensity on both the DWI (C) and the ADC map (D), and has a similar appearance to the left ovarian cyst with excrescences on all MR sequences.

Gadolinium was not administered as it is contraindicated during pregnancy. Since the mural nodules and irregularly thickened wall were isointense with the placenta on all MR sequences, bilateral decidualized endometriomas were considered more likely as a preoperative imaging diagnosis. However, ovarian malignancy was also included in the differential diagnosis as it may not be possible to clearly differentiate between decidualized endometrioma and ovarian cancer arising from endometriosis.

At 15 weeks of gestation, a left adnexectomy was performed by laparotomy. Intraoperative frozen section analysis showed no evidence of malignancy.

On gross pathological inspection, the left adnexal specimen was a dark red 9-cm unilocular cystic tumor that contained chocolate-like brown fluid indicative of endometrioma. Its internal surface was edematous and papillary excrescences protruding into the lumen of the tumor were found (Fig. 4).

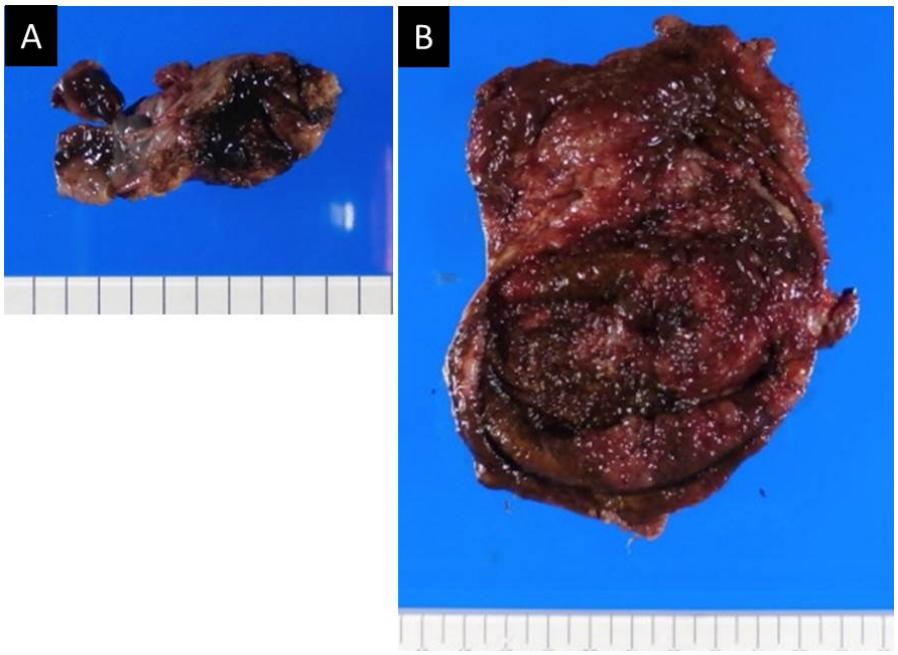

Figure 4.
Gross appearance of resected specimens of right (A) and left (B) ovarian cysts. Both adnexal specimens were dark red unilocular cystic tumors and contained chocolate-like brown fluid indicative of endometrioma. The internal surface was edematous and papillary excrescences protruding into the lumen of the tumor were found.

Microscopically, these excrescences consisted of round cells with small nuclei and abundant eosinophilic cytoplasm without cytologic atypia, characteristic of decidual cells (Fig. 5A). Marked edema was observed in part of the excrescence (Fig. 5B). The final histopathologic examination revealed ovarian endometrioma with marked decidualization and no evidence of malignancy.

At 37 weeks of gestation, a 2960-g healthy male infant was delivered by elective cesarean section due to total placenta previa. Right ovarian cystectomy for the lesion that had been identified by MRI was performed at the same time. The resected specimen, which measured 4 cm, had a strikingly similar appearance to the left ovarian lesion microscopically as well as macroscopically (Fig. 4). Although endometrial glands in the wall of right endometrial cyst were surrounded by many endometrial stromal cells, not all of them were decidualizing (Fig. 5C). The cytoplasm of decidualized stromal cells was positive for CD10 on staining (Fig. 5D). Thus, the diagnosis of right decidualized ovarian endometriotic cyst was finally made. The patient had an uneventful postoperative course without any detectable complications.

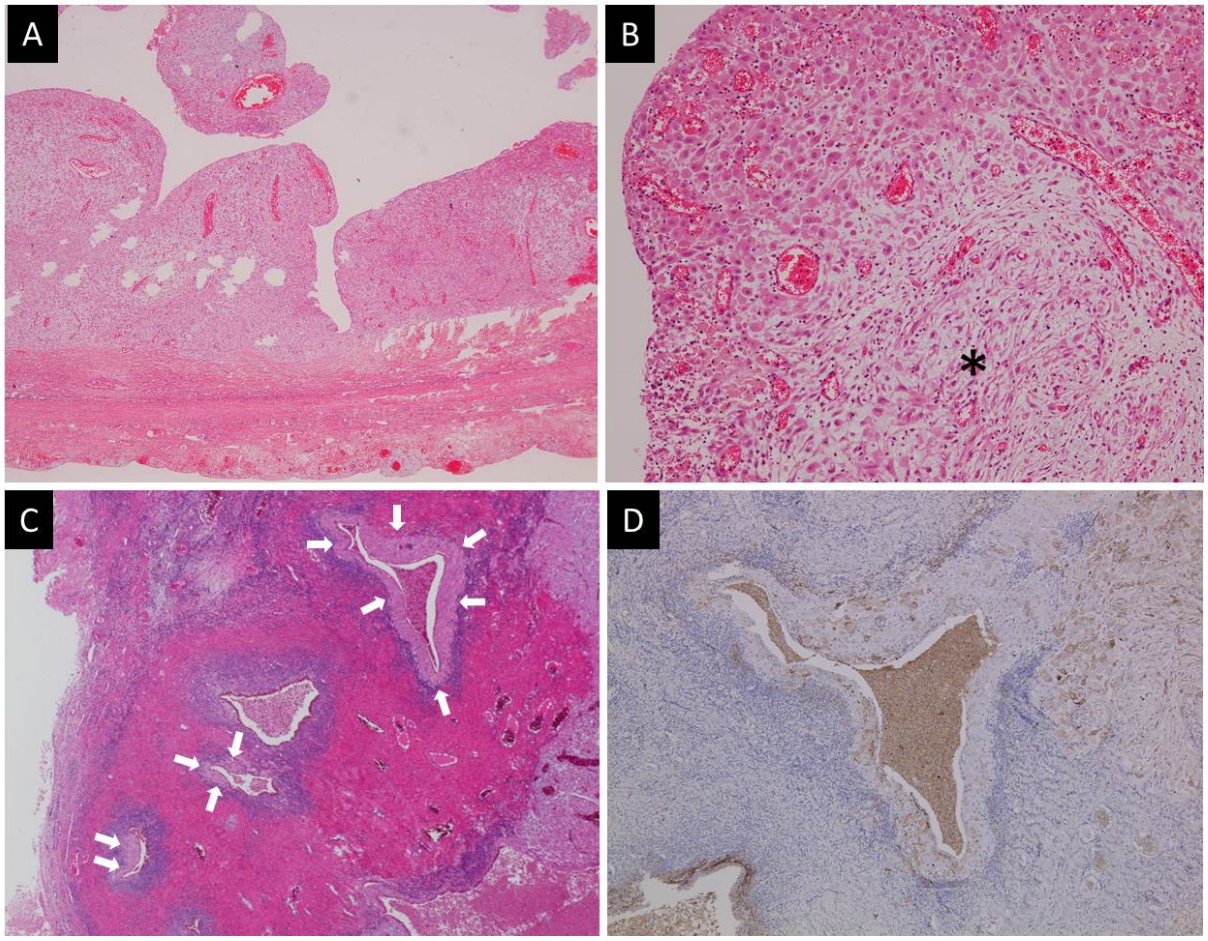

Figure 5.
A. A photomicrograph shows a papillary excrescence protruding into the lumen of the left ovarian cyst. B. Photomicrograph of the papillary excrescence. The excrescence is composed of stromal cells with small nuclei and abundant eosinophilic cytoplasm, characteristic of decidual cells. Marked edema was observed in part of the excrescence (asterisk). C. Ectopic endometrial glands in the wall of the right endometrial cyst. Not all endometrial stromal cells around glands are decidualized. The areas occupied by decidualized endometrial stroma are indicated by arrows. D. CD10 immunostaining in the ectopic decidualized endometrial gland. The cytoplasm of decidualized stromal cells was positive for CD10.

DISCUSSION

Decidualization is the hypertrophy of endometrial stroma cells and the formation of the decidua in response to progesterone, which functions to optimally adapt the endometrium for pregnancy (9). Deciduosis in the endometrial stromal cells of endometriosis, including ovarian endometriomas, can occur during pregnancy but has only been reported infrequently. Whether decidualization occurs may depend not only on the presence of endometrial stromal cells but also on other unknown factors associated with them. This is because, as shown in Figure 5C, although endometrial glands in the wall of the right endometrial cyst were surrounded by many endometrial stromal cells, not all of them were decidualizing. Irrespective of endometriosis, decidualization has been reported in areas such as the appendix, the visceral peritoneum, and the cervix (14-17). Therefore, it is believed that deciduosis may be the physiological response to pregnancy of hormone-responsive pluripotent cells in the peritoneal lining of women. Thus, it is speculated that ovarian deciduosis during pregnancy may be caused by two different processes, one being decidualization of a pre-existing ovarian endometriotic cyst and the other being rapid enlargement of ovarian ectopic decidualized endometrial glands differentiated from pluripotential cells in response to pregnancy. This is because Machida et al. described a decidualized ovarian endometriotic cyst filled with clear fluid but with no brown fluid indicative of endometriosis (18), and moreover, Takeuchi et al. reported that a decidualized endometrioma diagnosed clinically by MRI during pregnancy disappeared after delivery (11). In addition, an increase in size of the mass during the course of a pregnancy has been reported in a number of reports (6, 17). These phenomena are not likely to happen to common ovarian endometriomas clinically. In our case, it appears that the bilateral ovarian cysts arose from ovarian endometriotic cysts because they were filled with brown fluid, although they were detected after pregnancy.

The risk of cancer arising in pre-existing endometriomas is estimated to be 0.7% to 1% (19, 20). Therefore, structural changes in ovarian endometriomas detected during pregnancy should not be ignored. However, clinical differentiation of decidualized endometriomas from ovarian cancer still poses difficulties as both are intracystic vascularized excrescences of an endometrial cyst and have similar characteristics on color-flow Doppler sonography (10). On MRI, decidualized mural nodules on the cyst wall were isointense with the placenta within the uterus on all sequences. However, it is common for both malignant mural nodules arising from endometriosis and decidualized endometrial tissues to show high SI on T2WI (12) and high SI on diffusion-weighted MR images. One aspect that can differ is ADC values. The ADC value of decidualized mural nodules is significantly higher than that of malignant nodules of ovarian cancer. It is believed that the edematous, vascularized, and decidualized endometrial tissue with abundant cytoplasm of the stromal cells may cause ADC values to be higher than those of malignant tumors, which have high cellularity and reduced extracellular space that may restrict water movement. The high ADC of the mural nodules in our case is consistent with previous studies (11, 21) and was very close to the ADC of the placenta (Fig. 2D). This suggests that measuring the ADC of decidualized mural nodules may aid in their prospective diagnosis, but further studies will be needed to confirm the facts as the ADC values of benign and malignant cystic adnexal masses have not been consistently shown to differ significantly (22-25). Moreover there remains the limitation of measuring ADC value such as a lack of reproducibility and comparability of ADC values between MR machines if different scan parameters or methods of measurement are applied.

In the management of adnexal masses in pregnancy, surgical intervention is an additional option besides expectant management even for benign adnexal masses, especially when they are large in size (>8–10 cm). This is because even benign adnexal masses can cause symptoms or obstetric complications such as torsion, rupture, and labor dystocia that may necessitate emergency surgery in the third trimester, thus adding an increased risk of complications (26).

When malignancy is suspected, surgical treatment cannot be avoided to confirm the presence of malignancy pathologically (27).

In conclusion, MRI can assist in the prospective diagnosis of decidualized ovarian endometrioma. However, considering that it is not yet possible to clearly differentiate decidualized endometriomas from ovarian cancer, surgery or watchful observation may still be needed to exclude the possibility of malignancy.

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