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Case Report

Uterine diverticulum mimicking endometriotic cyst of the ovary ☆,☆☆

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ABSTRACT

Uterine diverticulum is a rare congenital malformation caused by abnormal fusion of the Müllerian ducts. The diagnosis of uterine diverticulum is difficult, and it is often misdiagnosed as a Müllerian duct anomaly, degenerated uterine fibroid, or ovarian cyst. We herein report a case of uterine diverticulum mimicking an ovarian endometriotic cyst. A multiparous woman with a history of normal vaginal delivery underwent magnetic resonance imaging for investigation of lower abdominal pain and fever. A 155-mm cystic lesion was observed on the ventral side of the uterus. The content of the cyst showed high signal intensity on T1- and T2-weighted images with precipitates of low signal intensity on the dorsal side, suggesting an endometriotic cyst of the ovary. Surgical and pathological findings revealed that the cyst was pedunculated from the anterior uterine body and composed of 3 layers: CD10-positive endometrium, a smooth muscle layer, and serosa. A uterine diverticulum was definitively diagnosed.

Introduction

Uterine diverticulum is a rare Müllerian anomaly that can be confused with other pelvic cystic or mass lesions. Diverticula of the uterus are classified as congenital true uterine diverticula (uterine diverticulum) and secondary uterine diverticula (uterine sacculation), which occur after pregnancy or surgery. Secondary diverticulum is a cyst that forms either by internal pressure from the pregnancy on the thinning muscle layer of

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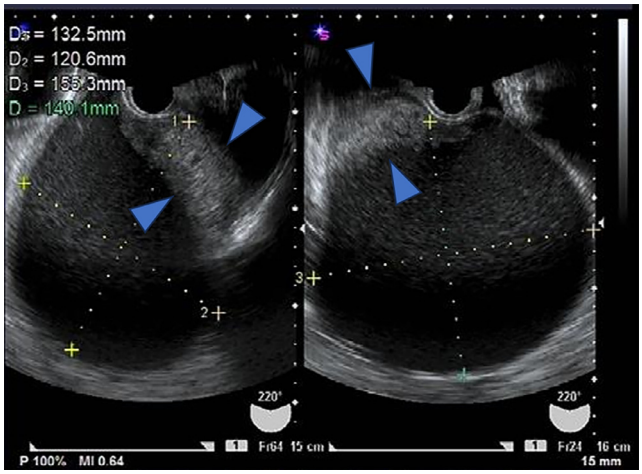

Fig. 1 – Transvaginal ultrasonography shows a cystic lesion (132 x 120 x 155 mm) on the ventral side of the uterus with a solid component (80 x 37 mm, arrowhead).

the cesarean scar or by the growth of chorionic tissue in the muscle layer of the uterus after pregnancy. Congenital uterine diverticulum is an extremely rare anomaly, making preoperative diagnosis difficult. Uterine diverticulum can be misdiagnosed as degenerative myoma, adenomyotic cyst, malignancy, or other uterine malformations on the basis of its imaging findings. In this case report, we describe a uterine diverticulum that mimicked endometriotic cyst of the ovary and discuss the magnetic resonance imaging (MRI) findings of uterine diverticulum.

Case presentation

A woman in her early 40*s* presented to our hospital because of lower abdominal pain and a fever that had persisted for several days. Her temperature was 37.6°C, and she had tenderness in the right lower abdomen. She had an obstetric history of gravida 3 para 2 (13 and 11 years previously, respectively). Both were normal vaginal deliveries, and no gynecological abnormalities including uterine diverticulum were detected during the pregnancies. Abdominal computed tomography performed 7 and 5 years previously for investigation of lower abdominal pain showed diverticulitis in the ascending colon, which was treated conservatively; no abnormalities were observed in the uterus or ovaries. The patient had no history of abdominal surgery or trauma. Her menstrual cycle was regular, and her last menstrual period had occurred 2 weeks previously.

Blood tests revealed a high white blood cell count of 11,210/μL, a C-reactive protein level of 1.54 mg/dL, and normal serum levels of carcinoembryonic antigen, CA19-9, and CA125. Transvaginal ultrasonography revealed a 132- × 120- × 155-mm cystic lesion with an 80- × 37-mm solid component on the ventral side of the retroflexed uterus (Fig. 1). A subserosal leiomyoma (43 × 34 mm) was observed on the posterior wall of the uterus. MRI showed that the cystic lesion had high signal intensity on both T1- and T2-weighted images, and there was no signal decrease on fat-suppressed T1-weighted images, suggesting bloody or protein-rich contents (Fig. 2). A component that was suspected to be an old clot due to preexisting hemorrhage was seen on the dorsal aspect of the cyst. The cyst wall was up to 5 mm thick and isointense to the normal myometrium. Diffusion-weighted images and apparent diffusion coefficient maps showed some diffusion restriction in the cyst. The cyst appeared to be contiguous with the right ovary. An intramural uterine leiomyoma was observed on the dorsal cervix. No congenital uterine, renal, or urinary tract malformations were observed. Based on the MRI findings, we initially diagnosed an endometriotic cyst of the right ovary with infection.

Antibiotics were administered after admission, and elective surgery was planned. However, urgent surgery was performed on the fifth day of hospitalization because of worsening of the abdominal pain and inflammatory reaction. Through a median incision in the lower abdomen, a large mass was found immediately below the peritoneum. Aspiration of the liquid content in the cyst to secure an adequate view yielded 1600 mL of old hemorrhagic fluid. The mass was stemming from the anterior wall of the uterus (Fig. 3) and growing into the right broad ligament. The bilateral ovaries and fallopian tubes were normal and showed no continuity with the mass. The root of the mass was ligated and dissected. Because the patient had a uterine leiomyoma and no desire for future fertility, total hysterectomy with preservation of the bilateral ovaries was performed. The surgery was completed without complications. The postoperative course was uneventful, and the patient was discharged on the eighth postoperative day.

Pathological examination revealed that the cyst surface was covered with a serous membrane. The middle layer of the cyst wall was positive for α-smooth muscle actin, a marker of smooth muscle, and the inner lumen was positive for CD10, an endometrial stromal marker; these findings suggested that the cyst wall consisted of uterine tissue (Fig. 4). The surgically removed uterus had 2 normal cornua, and no morphological abnormalities such as bicornuate uterus, septate uterus, or uterus didelphys were observed. The renal and urinary systems also had normal morphology. Cystic adenomyosis was ruled out because the native uterus had no findings suggestive of adenomyosis on MRI or histology. On the basis of these findings, we concluded that this lesion was a uterine diverticulum.

A retrospective review of the MRI scan showed an area of high signal intensity between the anterior wall of the uterine cervix and the cystic wall on T2-weighted images (Figs. 2A and B), which indicated the connection between the uterine cavity and the cyst.

Discussion

We experienced a case of uterine diverticulum that appeared more than 10 years after normal vaginal delivery. Uterine diverticulum is a rare morphological abnormality of the uterus. It is identified on MRI or ultrasound as a cystic lesion adjacent to the uterus or a mass lesion that contains an internal fluid component, and its lumen is continuous with the nor-

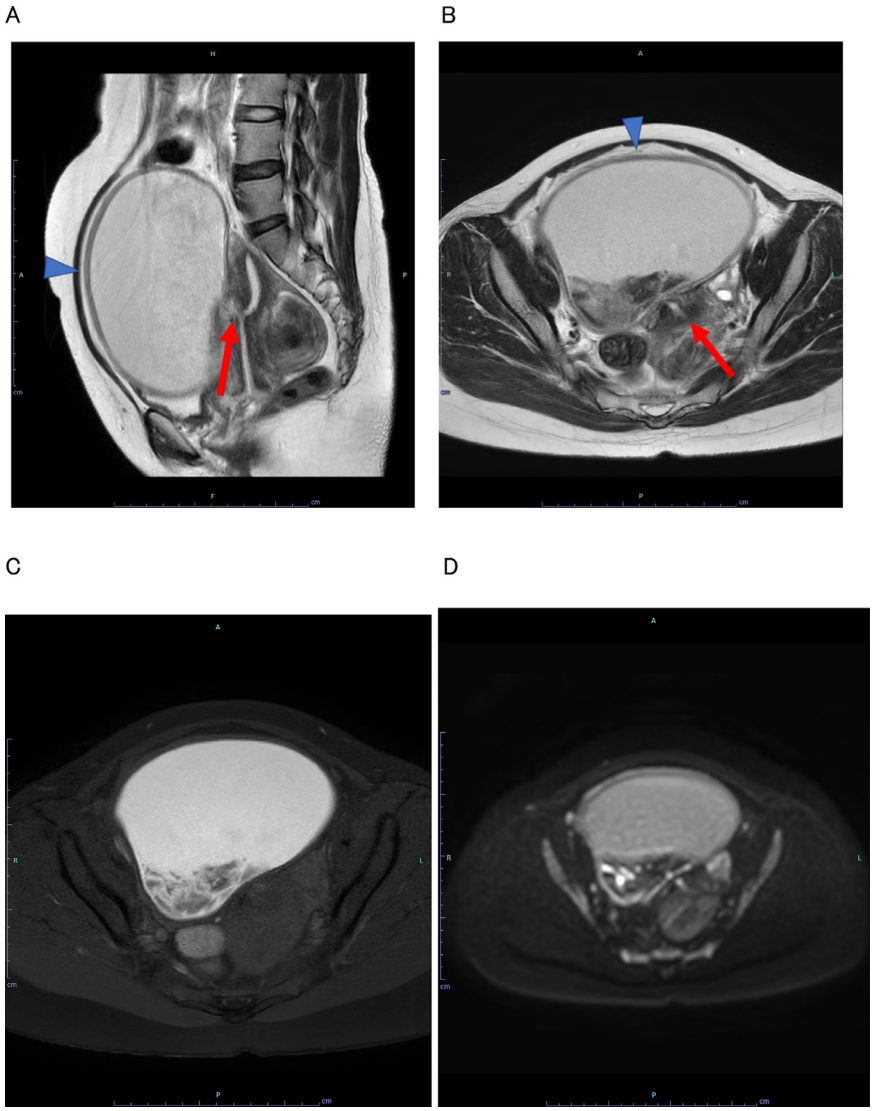

Fig. 2 – (A) Sagittal and (B) axial T2-weighted images. (C) Fat-suppression T1-weighted image. (D) Diffusion-weighted image. A cystic lesion was observed in front of the uterus and was filled with fluid showing high signal intensity on T1- and T2-weighted images. The dorsal sediments of the cyst showed low signal intensity on T1- and T2-weighted images and partial high signal intensity on diffusion-weighted images, suggesting chronic retracted blood clots with high concentrations of protein and hemosiderin. The wall of the cystic lesion was isointense to the uterine myometrium (arrowhead). Communication between the uterine cavity and the cyst was observed as a high-signal elongated structure (arrow) on T2-weighted images.

mal uterus cavity. Twenty-three case reports of uterine diverticulum have been published to date [1–16]. Uterine diverticulum is sometimes confused with uterine sacculation, a type of secondary diverticulum. Uterine sacculation forms at sites of myometrial thinning caused by uterine surgery, including cesarean section, and becomes temporarily enlarged because of the pressure associated with pregnancy. By contrast, uterine diverticulum is thought to form during uterine development. It has been suggested that the diverticulum forms from myometrium that has become vulnerable because of a fusion defect between the bilateral Müllerian ducts [16]. Considering this mechanism, diverticula should appear more frequently in the middle portion of the uterus. However, in previous reports, the site of the lesion was not only on the midline but also anterior, posterior, or lateral to the uterine body or cervix. Therefore, the development of uterine diverticula is not solely due to the aforementioned mechanisms. Another report hypothesized that the diverticulum arises from a localized, unilateral, distal duplication of the Müllerian duct [8]. However, the exact process of uterine diverticulum development remains inconclusive. A uterine diverticulum is sometimes huge, with some cases reportedly as large as 18 cm [2,6,16]. The thickness of the diverticulum also varies, with some cases reportedly as thick as 1.5 cm [6]. Another report stated that the wall of the diverticulum was as thick as the native uterus [15]. The wall of the diverticulum has a homogenous intensity [2], and the signal intensity of the wall is similar to that of normal myometrium, reflecting the fact that it is composed of normal myometrium.

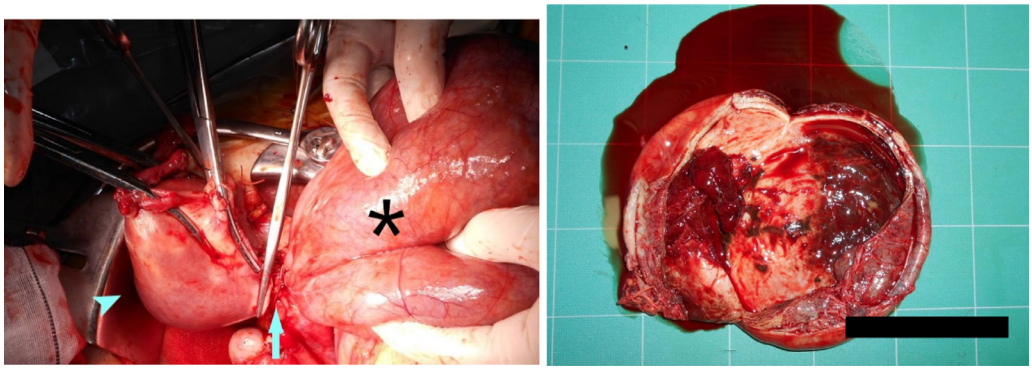

Fig. 3 – The cyst (asterisk) was stemming from the anterior wall of the uterus (arrowhead). The root (arrow) of the cyst was ligated and dissected. The lesion was a thick-walled cystic structure with an internal reservoir of old blood.

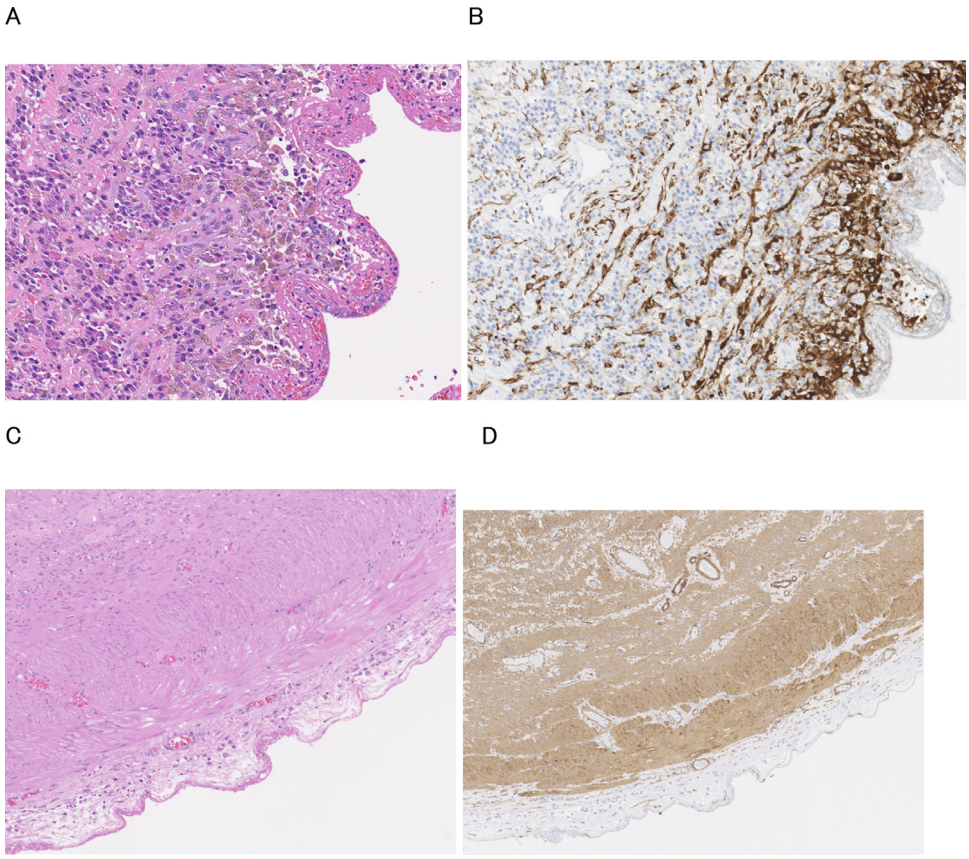

Fig. 4 – (A) Hematoxylin–eosin staining and (B) CD-10 staining of the inner layer of the cyst. The inner layer was lined with CD-10-positive endometrium. (C) Hematoxylin–eosin staining and (D) α-smooth muscle actin staining of the outer layer of the cyst. The middle layer was α-smooth muscle actin-positive, suggesting that it was composed of smooth muscle. The surface was covered with serosa.

The diverticular cavity is filled with fluid showing high signal intensity on T2-weighted images [10,11,13,15,16]. We found no cases in the literature in which the cavity showed high signal intensity on T1-weighted images.

Because of its rarity, accurate early diagnosis of uterine diverticulum can be difficult unless it is listed as a differential diagnosis. Differential diagnoses include degenerated uterine fibroids [16], torsion of ovarian cysts [6], unicornuate uterus [2], uterine bicornis unicollis [10], and incomplete septate uterus [14]. In our case, the preoperative diagnosis was an endometriotic cyst of the ovary because MRI showed a cystic lesion with internal hemorrhagic effusion and a rather thick wall that was contiguous with the right ovary. The following criteria are suggested to establish the diagnosis of uterine diverticulum: the surface is covered with peritoneum, the wall is composed of myometrium, a tract is present between the diverticular cavity and the uterine lumen, and the interior is lined with endometrium or decubitus membranes [5]. Differential diagnoses for a cystic lesion composed of endometrium and myometrium are unicornuate uterus and an accessory

cavitated uterine mass (ACUM). Unicornuate uterus is a uterine malformation. In this condition, a rudimentary horn usually exists on the contralateral side of the unicornuate uterus. If the rudimentary horn has functional endometrium, it is observed as a cavitated mass and sometimes enlarges because of hematometra, which may resemble a uterine diverticulum. This rare condition can be ruled out by the presence of the Fallopian tubes and ovaries on the uterus and their absence on the surface of the cyst on gross examination. In patients with uterine diverticulum, no other coexisting Müllerian anomaly is present, and MRI should therefore show a uterus cavity with 2 normal cornua. An ACUM can also be confused with uterine diverticulum. This is a rare pelvic lesion proposed in 2010, and the diagnostic criteria are as follows: (1) an isolated accessory cavitated mass usually located under the round ligament; (2) normal uterus, fallopian tubes, and ovaries; (3) excision of a mass with pathological examination; (4) an accessory cavity lined by endometrial epithelium with glands and stroma; (5) chocolate brown-colored fluid contents; and (6) no adenomyosis in the uterus (if resected), although tiny foci of adenomyosis may be present in the myometrium of the accessory cavity because of increased intracystic pressure [17]. An ACUM is similar to a uterine diverticulum in that it is a mass coexisting with a normal uterus and its cavity is lined with endometrium; the communication with the original uterine lumen is the key to differentiation.

On MRI examination of a uterine diverticulum, the tract between the diverticulum and the uterus is observed as a high-signal region on T2-weighted images, crosses the myometrium, and continues into the uterine cavity. Thin-section images may be useful to detect communication; such images are also useful in differentiating other pelvic masses and cysts as well as ACUMs. Hysterosalpingography can reveal the tract between the diverticulum and the uterine cavity [7]. However, MRI is particularly advantageous because it is a less invasive examination and provides a picture of the entire pelvic morphology, including the outer uterus, ovaries, and fallopian tubes. In the present case, abdominal computed tomography performed 7 and 2 years previously (4 and 9 years after the last delivery, respectively) showed no abnormality in the uterus or ovaries. This suggests that the enlargement of the uterine diverticulum was not caused by pregnancy. Although a case of uterine diverticulum enlargement with pregnancy has been reported [2], we found no previous reports of rapid enlargement of the diverticulum over a period of several years. It is speculated that retrograde menstruation causes the diverticulum to enlarge [6] or that the contents of the diverticulum increase secondary to accumulation of the shedding membrane of the diverticulum itself. Nevertheless, the trigger for progressive enlargement of the diverticulum remains unknown. Our patient had a subserosal leiomyoma on the posterior wall of the uterus; however, given its size, morphology, and location, it was unlikely to obstruct the menstrual passage and cause enlargement of the diverticulum.

In summary, we experienced a case of a congenital uterine diverticulum that mimicked an endometriotic cyst of the ovary. The possibility of a uterine diverticulum should be considered when a cystic lesion is observed in the pelvis and appears to be contiguous with or adherent to the uterus.

Patient consent

The patient outlined in this case report provided written informed consent for publication of the case.

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