

## Research Letter

# Sepsis secondary to cesarean scar diverticulum resembling an infected leiomyoma

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Cervical diverticulum is a very rare anomaly and has also been called pregnancy-associated sacculations [1] and previous cesarean delivery scar (PCDS) defect and may impair the drainage of menstrual flow through the cervix producing an accumulation of blood in the pouch. Most reported cases described the presence of a hiatus or diverticulum on the anterior isthmus at the site of a PCDS and only three cases were considered true uterine diverticula [1]. Uterine abscess or pyometra is uncommon and is thought to occur as a result of interference with the natural drainage of the uterus. It is commonly caused by malignant or benign gynecological tumors, radiation cervicitis, atrophic cervicitis, congenital anomalies, and intrauterine device (IUD). We herein describe the first case of PCDS diverticulum complicated by sepsis secondary to abscess formation.

A 41-year-old gravida 2 para 2 was seen with complaints of intermittent fever and progressive lower abdominal pain for 2 weeks. Her history was significant for two cesarean deliveries, an IUD in place for 3 years, pelvic inflammatory disease 5 years previously, and recurrent urinary tract infection in the recent 6 months. Additionally, a lower corpus uterine mass of about 3 cm in diameter was noted 3 years prior, and she experienced postmenstrual spotting occasionally.

On admission, her body temperature was 38.4°C. A palpable mass over the lower anterior aspect of the uterus, profuse odorless vaginal discharge, and cervical motion tenderness were noted on pelvic examination. The abdomen was soft, but mild tenderness without rebounding pain was noted over the lower abdomen. Abdominal ultrasonography revealed a fluid-filled mass over the anterior wall of the uterus and an IUD in the uterine cavity (Fig. 1). Laboratory results revealed an elevated

white blood cell count (16,100/mm<sup>3</sup>, segmented neutrophils 96%) with a left shift and an elevated C-reactive protein level of 124 mg/dL. An infected uterine tumor with malignant change could not be excluded. A dilatation and curettage was performed and the patient was treated with gentamycin and cephadrine. Five days after admission, the blood culture yielded *Escherichia coli* and the histopathological diagnosis of endometrial tissue was endometritis. Computed tomography scan showed a 5 × 6 × 9 cm exophytic tumor in the lower anterior aspect of the uterus with central necrosis (Fig. 2).

Because patient's symptoms were much improved and the infection had been controlled, surgery was arranged 2 weeks later because uterine malignancy change could not be excluded. On explorative laparotomy, a well-defined mass of about 10 cm in diameter was found above the PCDS. The bladder was compressed caudally by the mass. Total abdominal hysterectomy was performed. Grossly, the mass was fibrotic tissue with multiple tracts containing brown purulent discharge and had a narrow connection of about 0.5 cm in diameter with the uterine cavity (Fig. 3).

Pathological examination of the mass showed bundle-like muscle tissue but no findings of degenerated leiomyoma. The cavity of the tumor and connection to the uterus were lined by epithelial cells resembling endocervical epithelium (Fig. 4). An infected cervical diverticulum of the PCDS was diagnosed. The patient was discharged in good condition and has remained free of symptoms during follow-up.

Abnormal uterine bleeding is one of the most common complaints among women in their reproductive years, usually secondary to functional disorders of the menstrual cycle or uterine pathology, such as myomas, polyps, or endometrial hyperplasia. However, another cause of bleeding has recently been described in patients with PCDS with an anatomic defect in relation to the scar [2]. Morris [3] reported 51 patients who underwent hysterectomy because of abnormal bleeding and abdominal pain. None of the patients had any uterine and hormonal pathology and all of them had at least one previous

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Fig. 1. Abdominal ultrasonography revealed a fluid-filled mass over anterior wall of uterine corpus with connection to uterine cavity (arrow) and an intrauterine device in uterine cavity. B = bladder; M = mass.

cesarean delivery. The author found several abnormalities in the specimens in relation to the Cesarean scar, such as distortion and widening of lower uterine segment, congested endometrium, lymphocytic infiltration, and capillary dilation. The blood that accumulates in the pouch may be produced *in situ* and remain there because of impaired drainage.

Our patient had an IUD inserted for 3 years and blood culture showed *E coli*. Intrauterine infections with organisms, such as with *Actinomyces*, *E coli*, *Klebsiella pneumoniae*, *Bacteroides fragilis*, *Candida albicans*, *Streptococcus* sp, and *Staphylococcus* sp have been found in patients with an IUD [3,4]. Our theory is that the infection in our patient may be related to the accumulation of blood because of the PCDS diverticulum and presence of an IUD.

Umezaki et al [1] reported one case with a true uterine diverticulum resembling a degenerated leiomyoma. Three known cases of uterine diverticula resembling a pelvic tumor

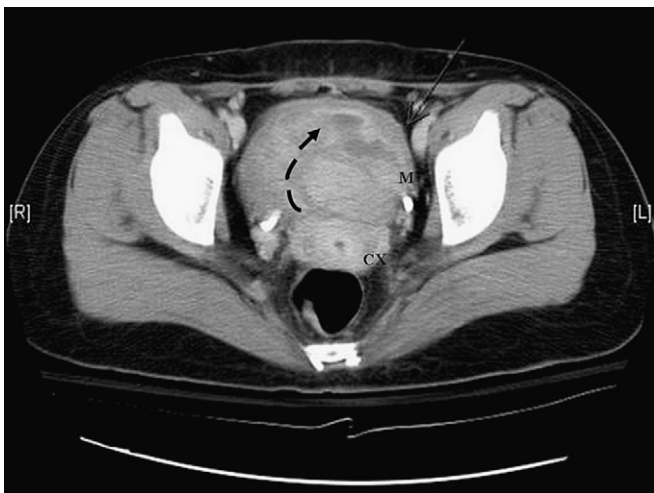


Fig. 2. The computed tomography showed a 5 × 6 × 9 cm exophytic tumor (M) in lower anterior aspect of lower uterine corpus with central fluid accumulation and connection to uterine cavity (arrow) just above the bladder.

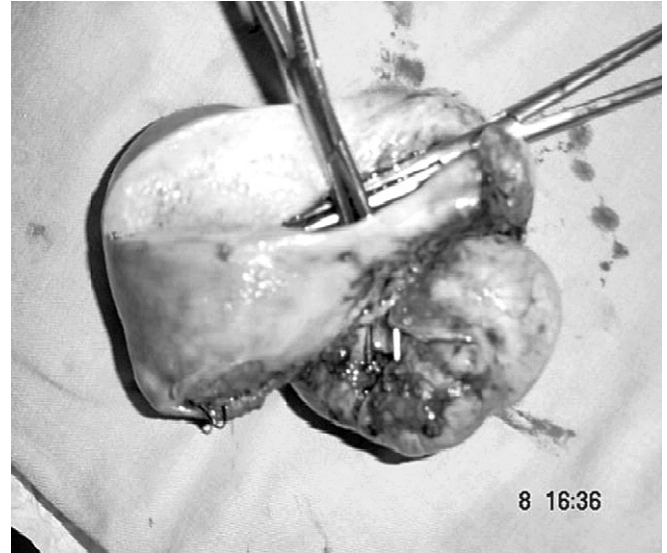


Fig. 3. The diverticulum was connected to the uterus and contained purulent discharge.

have been reported, and all were true diverticulum. A PCDS diverticulum presenting as a pelvic tumor resembling a degenerated leiomyoma is very rare (Table 1). In patients with PCDS diverticulum, most present with bleeding disturbances (84% with postmenstrual spotting) and infertility [7]. Histological study of PCDS obtained during surgery showed 50% with fibrosis, 21% with endometrial tissue, and 8% with inflammatory infiltration [7]. In our case, a fibrotic mass was discovered 3 years before the patient's admission and progressed with fluid accumulation and repeated inflammatory process. It has been suggested that transvaginal sonography is the first choice for screening of PCDS diverticulum. Therefore, we believe that follow-up sonography is necessary in patients with PCDS diverticulum, whether or not symptoms are present.

In summary, we reported an extremely rare case of sepsis secondary to PCDS diverticulum complicated with abscess

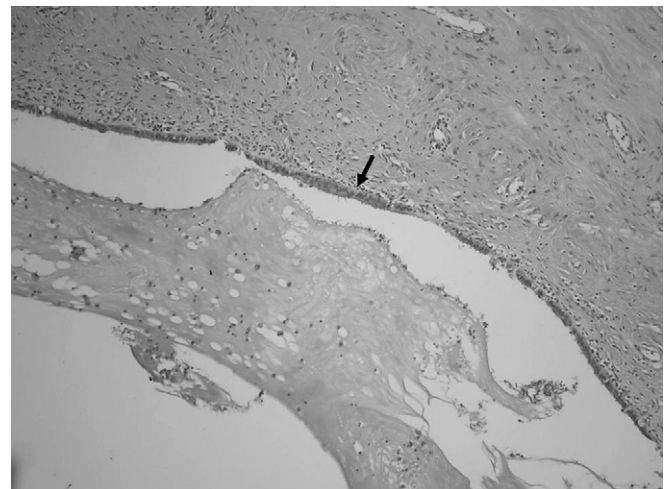


Fig. 4. The connection of the uterus and diverticulum was lined by epithelial cells resembling endocervical epithelium (arrow) (hematoxylin and eosin, 100×).

Table 1  
Comparison of reported cases of uterine diverticulum resembling a pelvic tumor

Characteristics	Bennett [6]	Engel and Rushovich [5]	Umezaki et al [1]	Present case
Age (yr)	24	34	41	41
Parity	0	0	0	2
Symptoms	Abdominal pain	Abdominal pain	Abdominal fullness Vaginal discharge Fever	Abdominal fullness Vaginal discharge Fever
Site	Right lateral	Right posterior	Midline anterior	Midline anterior
Size (cm)	2.5 × 2 × 1.5	7.5 × 4.5 × 3.5	18 × 17 × 16	9 × 6 × 5
Treatment	Diverticulectomy + RSO	TAH	TAH	TAH
Final diagnosis	True uterine diverticulum	True uterine diverticulum	True uterine diverticulum with infection	PCDS diverticulum with infection

PCDS = previous cesarean delivery scar; RSO = right salpingo-oophorectomy; TAH = total abdominal hysterectomy.

formation and resembling a uterine tumor. PCDS diverticulum should be included in the differential diagnosis of a pelvic mass.

## References

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