Successful pregnancy and vaginal delivery after laparoscopic excision of a congenital uterine cervical diverticulum: A case report

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Abstract

Uterine cervical diverticulum is a very rare malformation. Affected patients are reported to have infertility issues and problems during the perinatal period. A 32-year-old nulliparous woman visited another obstetrics and gynecology hospital because of infertility. A cyst branching out from the uterine cervix was discovered. Subsequently, she conceived via assisted reproductive technology, but the uterine cyst was left untreated. Eventually, the pregnancy was terminated due to an enlarged uterine cyst and several birth defects. She was referred to our hospital where she was diagnosed with a uterine cervical diverticulum. We excised the diverticulum via a laparoscopic approach. Afterward, she became pregnant and delivered a baby vaginally at 37 weeks. To our knowledge, this is the first report of successful delivery after laparoscopic diverticulum excision. We recommend cervical diverticulum excision before pregnancy because of the potential adverse events associated with cervical diverticulum during pregnancy.

Key words: congenital cervical diverticulum, laparoscopic surgery, pregnancy, infertility, uterine diverticulum excision.

Introduction

Uterine cervical diverticulum is a very rare malformation.\(^1\)–\(^5\) Some studies have reported that patients diagnosed with a uterine cervical diverticulum presented with lower abdominal pain, abnormal uterine bleeding\(^1\)–\(^4\) and infertility.\(^1\) The optimal treatment for uterine cervical diverticulum in a patient with infertility has been under debate. Herein, we describe a patient with a congenital uterine cervical diverticulum who had a successful delivery after laparoscopic excision of the diverticulum. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Case

A 32-year-old woman had experienced dysmenorrhea as a subjective symptom since she was 28 years old. When she was 30 years old, she visited the obstetrics and gynecology department of another hospital with a history of primary infertility. A cyst, measuring 4 cm and arising from the uterine cervix, was detected via transvaginal ultrasound examination, but no diagnosis was made. The surgery was not chosen because the cyst was assessed to be unrelated to her infertility. She was offered assisted reproductive technology because of her unexplained infertility. Intracytoplasmic sperm injection was required owing to fertilization failure in the initial conventional in vitro fertilization attempt.
After conceiving, the cyst gradually enlarged, and she complained of lower abdominal pain and atypical genital bleeding. At 17 weeks of gestation, the atypical genital bleeding was stopped; however, the diverticulum enlarged to 10 cm. Moreover, the fetus was diagnosed with VATER association: the presence of anomalies in the vertebrae, anus, trachea, esophagus and renal (kidneys). At 21 weeks of pregnancy, she underwent an induced abortion. Subsequently, she was referred to our hospital to treat the uterine cervical cyst and infertility.

In our hospital, bimanual palpation revealed a mobile cyst measuring 5 cm, which had become shrunken after the abortion, located at the right side of the uterine cervix. Magnetic resonance imaging (MRI) revealed that the cyst, which was connected to the uterine cervix (Fig. 1a, b), measured 5.5 × 5.0 × 4.5 cm in size. Hysterosalpingography (HSG) revealed that the diverticulum branched from the cervical canal of the uterus (Fig. 1c). Computed tomography revealed that the right ureter was passed dorsal to the diverticulum. The right uterine artery was located along the posterior aspect of the diverticulum, reaching the cervix and diverticulum. Because the enlarged diverticulum was assessed for genital bleeding and tenderness during the first pregnancy, we decided to excise it before the next pregnancy.

We performed laparoscopic diverticulum excision using the pneumoperitoneum system under general anesthesia. We inserted a ureteral stent into the right ureter to identify its location during surgery. The surgical procedure was performed using the diamond port configuration with a uterine manipulator. First, we opened the right anterior broad ligament of the uterus, after which we could look at the diverticulum (Fig. 2a, b). We injected diluted vasopressin locally into the tunica muscularis tracheae of the diverticulum to reduce blood loss. We incised the connection between the diverticulum and uterine cervix conically, from the side of the diverticulum, so that the myometrium was preserved for closing and reinforcing the cervical defect (Fig. 2c). Subsequently, we repaired the incised part by overlapping the excess myometrium (Fig. 2d) and closed the opening of the broad ligament of the uterus via suturing. The postoperative course of the patient was uneventful, and she was discharged 8 days after surgery. The pathological diagnosis revealed that the epithelium of the cyst was of the endometrium with tubal epithelial metaplasia, with a smooth muscle layer below it and a serosa in the outermost layer (Fig. 3). Its image was consistent with that of a uterine diverticulum.

Two months postoperatively, we performed HSG (Fig. S1), which revealed no leaks from the sutured area. The patient was allowed to conceive 3 months after the surgery. She became pregnant via frozen embryo transfer, and at 37 weeks, she delivered a female baby weighing 2440 g via vacuum extraction. After delivery, she did not experience recurrence of the uterine diverticulum, and her menstruation was normal.

**Discussion**

To our knowledge, this is the first reported case of a successful term delivery following the laparoscopic excision of a congenital uterine diverticulum. Some previous reports have described diverticula, but most of the diverticula were secondary diverticula, which were diagnosed after uterine rupture during the third stage of labor, and some other cases were secondary...
A congenital uterine cervical diverticulum is a very rare malformation and is difficult to diagnose. The pathogenesis was considered to be a partial duplication of the Mullerian duct or an extension of the uterine wall, which has been weakened by local fusion failure of the Mullerian duct. The uterine diverticulum is an isolated accessory cavitated mass that usually communicates and protrudes from the corpus uteri. The uterine diverticulum is lined with endometrial epithelium with glands and stroma. Other diseases that resemble uterine cervical diverticulum include malformation of the Mullerian duct such as accessory and cavitated uterine masses, unicornuate uterus with a non-communicating rudimentary horn, uterine myoma and ovarian cyst, etc. In previous reports, the diagnosis of a cervical diverticulum was eventually confirmed via hysteroscopy and laparoscopy; however, a combination of HSG and MRI would have been sufficient to reach an accurate diagnosis. In this case, similar to previous reports, MRI and HSG were useful for the diagnosis. Schickele stated that a diverticulum must be covered with peritoneum, contain smooth muscle in its wall, contain endometrial glands, be connected to the uterine cavity by a tract and be lined by decidua. In our case, the three-layer structure comprised the intima, basal layer and muscle layer were maintained, and there was no contradiction as a diverticulum in the pathology. Preoperative HSG of the uterus showed that the diverticulum was connected to the cervical canal of the uterus and the diverticulum. Because she had no history of uterine surgery, this case could be diagnosed as a congenital uterine diverticulum.

To our knowledge, there were 22 reported cases of congenital uterine diverticulum in the literature. The mean age of the patients was 35.4 years.
(19–54 years), and the location of the diverticulum was the cervix in 13 cases (59%) and the corpus uteri in 5 cases (22.7%) in the reported cases. Patient symptoms were atypical bleeding (8 cases; 36.3%), abdominal pain (6 cases; 27.2%), infertility (4 cases; 18.2%), dysmenorrhea (2 cases; 9%), prolonged menstruation (1 case; 4.5%) and a feeling of abdominal fullness (2 cases; 9%). Four cases were diagnosed during pregnancy. The results of those 4 cases were as follows: 2 cases (9%) were preterm birth, 1 case (4.5%) was an intrauterine fetal death, and the other was an ectopic pregnancy (4.5%). Hysterectomy was performed in 8 cases (36.3%); diverticulum resection was performed in 6 cases (27.2%); and transcervical fenestration was performed in 1 case (4.5%). There was no reported pregnancy after diverticulum resection.

In the present case, the major clinical symptoms were dysmenorrhea and infertility. Some studies have reported that the presence of a uterine diverticulum can cause infertility.\textsuperscript{1,6} Although the reason for the infertility is unclear, Seoud et al. speculated that the abnormal cervical condition might have created a hostile cervical environment that impaired sperm function and transportation into the uterine cavity.\textsuperscript{1} They considered three mechanisms as possibilities. First, motile spermatozoa deposited at the external cervical os may enter the large collecting diverticulum, which acts as a reservoir and causes a dilutional factor to the number of progressive spermatozoa.\textsuperscript{1} Second, the presence of old blood in the diverticulum may contribute to various toxins, which could adversely alter sperm motility and function.\textsuperscript{1} Third, such a cervical anatomic defect could affect normal endocervical canal glandular function, hence, interfering with normal sperm storage, capacitation and transport.\textsuperscript{1} Since the present case has already known to be fertilization failure and has been stepped up to intracytoplasmic sperm injection, the infertility factor of the diverticulum and the effect of surgical treatment could not be substantially verified. However, to date, the treatment of a cervical diverticulum in a patient with infertility has not established. Some reports have described that cervical diverticulum in pregnancy can result in preterm birth, intrauterine fetal death and ectopic pregnancy.\textsuperscript{1,3,6} Moreover, in our case, the uterine cervical diverticulum increased in size during the first pregnancy. The previous literature\textsuperscript{1} also shows that diverticula grow during pregnancy, and we speculate that it is because intrauterine pressure increases with pregnancy. The mechanism by which the diverticulum causes complications during pregnancy is thought to be due to the fluid stored in the diverticulum causing infection. Indeed, her first pregnancy course was poor owing to several untoward symptoms, recurrent genital bleeding and lower abdominal pain. The absence of term delivery in the past literature suggests that

\textbf{Figure 3} The histopathological image of the cyst. Hematoxylin and eosin staining of the cyst (a. x100, b. x400). The pathology showed a three-layered structure. That is, the inner layer was the endometrium, the lower layer was the smooth muscle and the outermost layer was the serosa (a). An endometrial gland with tubal epithelial metaplasia was found (b).
the present case might have had adverse prenatal events, miscarriage, intrauterine fetal death and preterm delivery, even without induced abortion.

Given the risk during the perinatal period in which a uterine diverticulum is present, we recommend surgical excision of a cervical diverticulum before pregnancy, based on the findings in this case. According to past reports, the perinatal prognosis of the uterine diverticulum is poor, and active management is considered important if the patient desires to bear children. There is no clear surgical indication based on the size of the diverticulum, but the risk of growth during pregnancy should be informed to the patient even if the diverticulum is very small. In the resection of diverticulum, it is necessary to completely remove functional endometrium and cervical gland; however, it is desirable to preserve the muscle layer as much as possible to avoid perforation, stenosis, occlusion and excessive deformation of cervical canal, pathological condition like cesarean scar syndrome, perinatal uterine rupture, if leiomyoma or adenomyosis is not present. Because, in our case, we preserved the muscle layer, we could convince the patient to attempt a vaginal delivery. However, if the diverticulum is allocated in the upper part of the uterine body, an elective cesarean section might have been chosen similar to that of cesarean scar syndrome, perinatal uterine rupture, if leiomyoma or adenomyosis is not present. Because, in our case, we preserved the muscle layer, we could convince the patient to attempt a vaginal delivery. However, if the diverticulum is allocated in the upper part of the uterine body, an elective cesarean section might have been chosen similar to that of the post myomectomy patient for the uterine fibroid.

This case revealed that the patient with congenital uterine diverticulum, the rare form of Mullerian anomaly, can safely achieve pregnancy and childbirth when appropriate diagnosis and management are provided. It is important to determine the uterine site for the resection considering postoperative recurrence, fertility and perinatal prognosis and to study muscle layer sutures. The findings of this report need to be verified by confirmatory studies that will be accumulated in the future.

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Disclosure

None declared.

Supporting information

Additional Supporting Information may be found in the online version of this article at the publisher’s web-site:

Figure S1 Postoperative findings. Postoperative hysterosalpingography of the uterus (arrow: diverticulum resection site)