Three cases of successful operative treatment for cesarean scar syndrome

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Abstract
Cesarean scar syndrome (CSS) is caused by a defect in a prior cesarean section scar. Patients with CSS have gynecologic symptoms such as post-menstrual brown discharge, pelvic pain, and secondary infertility. We report three cases of CSS that received successful surgical treatment. Two cases showed post-menstrual brown discharge and one case showed a history of secondary infertility for four years. Transvaginal ultrasonography and magnetic resonance imaging indicated the cesarean scar defect and blood pooling. In all cases, we performed surgical treatment. We could not identify the cesarean scar defect by laparoscopy only. In two cases, we performed combined hysteroscopic and laparoscopic study, and in one case, we cut the myometrium vertically to identify the defect. Immediately after surgery, the post-menstrual brown discharge disappeared, and the case with secondary infertility achieved pregnancy six months after surgery. In conclusion, using hysteroscope or cutting the myometrium vertically was useful for identifying the defect.

Key words: cesarean scar syndrome, laparoscopic surgery, hysteroscopic surgery

Introduction
Cesarean section rate is increasing globally. Although cesarean deliveries can be necessary for saving fetuses as well as mothers, there are increasing long-term obstetrical risks for subsequent deliveries such as placenta previa, placenta accreta, uterine rupture, and cesarean scar pregnancy.

Previous cesarean section not only increases obstetrical risks of subsequent deliveries but also risks of gynecologic problems. Some women with cesarean scar defects have gynecologic symptoms such as menstrual abnormalities, post-menstrual brown discharge, pelvic pain, and secondary infertility. This condition was first reported by Hayden Morris in 1995 who coined the term "cesarean scar syndrome (CSS)."

While some reports have shown that surgical intervention is effective for symptom relief, a standardized treatment protocol has yet to be established and few reports have described in...
detail how to treat CSS surgically. Therefore, we report three cases of successful surgical treatment of CSS to help clarify which techniques are important for a standardized protocol.

In all three cases, written informed consent was obtained. The methods of the surgery were approved by the institutional Review Board at SHIN-YURIGAOKA General Hospital (No.20200929-2-3).

**Case History**

**Case 1**
The first case is of a 42-year-old Japanese woman, gravida 1, para 1, who underwent cesarean section in 2016 and had two weeks of post-menstrual brown discharge after delivery.

**Case 2**
The second case is of a 36-year-old Japanese woman, gravida 2, para 2, who underwent cesarean section in 2009 and 2017 and had prolonged menstruation for 20 days after her second cesarean section.

**Case 3**
The third case is of a 39-year-old Japanese woman, gravida 1, para 1, who underwent cesarean section in 2011 and, after the cesarean section, experienced secondary infertility for four years.

**Diagnosis**
In all three cases, transvaginal ultrasonography (TV-US) and magnetic resonance imaging (MRI) were performed. TV-US and MRI indicated the presence of a cesarean scar defect and blood pooling in the defect (Fig.1). The residual myometrial thickness measured by MRI was 2.0 mm, 3.0 mm, and 7.2 mm for case 1, case 2, and case 3, respectively. In case 3, bilateral ovarian endometriosis was also found by TV-US and MRI.

**Treatment**
The patients of case 1 and case 2 did not want to retain the ability to conceive. We presented hormone treatment, including low dose estrogen-progestin, levonorgestrel, or dienogest, surgical repair, and hysterectomy as treatment options for their symptoms, and both women selected surgical repair. Conversely, the patient of case 3 wished to conceive. We therefore performed routine infertility test such as salpingography by ultrasound, blood test for hormone level and male infertility test. All tests were normal except MRI which showed a cesarean scar defect and blood pooling in the defect. We suspected that CSS might be a cause of her secondary infertility. We presented infertility treatment (timed intercourse, intrauterine insemination and artificial reproductive technology) and surgical repair as her treatment options, and she selected surgical repair. In all cases, a board-certified member of the Japanese Society of Gynecologic and Obstetric Endoscopy performed surgeries, with a different surgeon for each case.

In case 1 and case 3, we performed hysteroscopy-assisted laparoscopic surgeries, and in case 2, we performed only a laparoscopic surgery. In all three cases, we cut the peritoneum of the vesicouterine pouch using monopolar scissors to separate the bladder from the anterior wall of the uterus. We could not identify the defect of the cesarean scar solely from the laparoscopy. Thus, in case 1 and case 3, we identified the defect hysteroscopically (Fig. 2). We then turned off the endoscopic light and identified the defect where the hysteroscopic
light had been seen (Fig. 3). After the complete resection of the defect’s fibrotic tissue using monopolar scissors, we repaired the myometrium via interrupted suture using absorbable monofilament (0 MONOCRYL®) (Fig4). In case 2, we cut the myometrium at the
center of the vesicouterine pouch vertically to identify the defect (Fig. 5, 6-a). The length of the vertical incision was about 1 cm. The defect was identified as the thinnest part of the myometrium. Then, we resected the wall of the defect and repaired the myometrium via interrupted suture using absorbable monofilament (0 monocryl®)(Fig. 6-b).

After the surgery, the residual myometrial thicknesses measured by MRI or TV-US were 5.7 mm, 9.6 mm, and 10.9 mm for case 1, case 2, and case 3, respectively (Fig. 7). In case 1 and case 2, the symptoms, such as post-menstrual brown discharge and prolonged menstruation, disappeared after surgery. Case 3 underwent oocyte pick-up during the contraceptive period after surgery and achieved pregnancy by in vitro fertilization (IVF) six months after surgery.

Discussion

We encountered three cases of women with CSS who received successful surgical treatment. We performed two different methods to identify the defect of a previous cesarean scar: using a hysteroscope or cutting the myometrium vertically.

Previous reports show that surgical treatment is effective for CSS symptom relief as well as for treating infertility due to CSS4-8. These reports also show that there are various operative methods to obtain CSS symptom relief, such as hysteroscopic surgery, laparoscopic surgery, vaginal surgery, and robotic surgery8,10. However, a standardized protocol of the surgical treatment for CSS has not been established yet and few reports have shown in detail how to treat CSS surgically. According to some previous reports, hysteroscopic surgery should be utilized only when the residual myometrial thickness is sufficiently large (for example, more than 2.5 mm or 3.5 mm) because of the risk of uterine rupture associated with hysteroscopic surgery7,8.

In the three cases we reported, we performed laparoscopic surgery because, in two cases, the residual myometrial thickness was insufficient (2.0 mm and 3.0 mm), and, in the other case, we needed to resect ovarian endometriosis during the same procedure. It was difficult to detect the location of the cesarean section scar by only observing laparoscopically3. Thus, we performed two different methods in order to identify the defect. In one case, we cut the myometrium vertically to identify the defect. One previous report showed the method of cutting the myometrium vertically to identify the defect during the laparotomy11. This is the first report to show this method laparoscopically. In this method, we only needed two surgeons for the laparoscopic surgery because hysteroscopy was not required. However, we do not know whether the vertical suture give an influence on future pregnancies. Thus, we recommend this method only for patients who do not wish to conceive. In the other two cases, we performed hysteroscopy in order to detect the defect of the cesarean scar. Observing the defect hysteroscopically and detecting the defect with the guidance of a hysteroscope was useful to distinguish the location and the size of the cesarean section scar3. Although hysteroscopy-assisted laparoscopic surgery needed an additional surgeon who was proficient with hysteroscopy3, hysteroscopy-assisted laparoscopic surgery was safer and less invasive. In addition, we can observe endometrial cavity by hysteroscopy, so using
hysteroscopy is good for patients with infertility.

Some previous studies have shown that operative treatment is effective for treating secondary infertility caused by CSS\textsuperscript{4,7}. Pooling of old blood in the cesarean scar defect, which produces cytokines, may obstruct embryo implantation, leading to secondary infertility\textsuperscript{4,7}. According to previous reports, the pregnancy rate of women with CSS and secondary infertility after surgery was about 65\%\textsuperscript{4,7}. Among our reported cases, only one patient wished to conceive again and achieved pregnancy six months after surgery via IVF.

In conclusion, we encountered three cases of women with CSS who received successful surgical treatment. The defect of the uterine wall could not be identified solely laparoscopically; thus, we performed two different methods to identify the defect. Using a hysteroscope or cutting the myometrium vertically are useful for identifying the defect safely. Therefore, these procedures should be considered when creating a standardized protocol for treating CSS surgically.

The summary of this report was presented on the 57th annual meeting of Japan Society of Gynecologic and Obstetric Endoscopy and Minimally Invasive Therapy.

Conflict of Interest

The authors have no conflicts of interest relevant to this article.

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