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Case Report

Successful Management of an Extremely Large Uterine Scar Defect

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ABSTRACT

Background: Cesarean section is one of the most common surgical procedures in young women. Cesarean scar defects may occur postoperatively and may cause symptoms such as chronic pain, postmenstrual bleeding as well as secondary infertility.

Case presentation: We report the case of a 35-year-old woman who was referred to our center for the management of a large cesarean scar defect. She had history of two cesarean sections and presented with abnormal uterine bleeding, chronic pain and dyspareunia. Hysteroscopic examination showed the presence of a little isthmocele on the anterior uterine wall. Hysteroscopic resection with electrocoagulation was performed. The patient remained symptomatic and, unfortunately, five years later, magnetic resonance imaging (MRI) revealed a large anterior isthmocele (50x80 mm). A complete repair of the large defect was achieved by laparotomy.Postoperative MRI as well as hysteroscopic examination showed no signs of residual isthmocele.

Conclusion: There are three main surgical options for isthmocele repair: vaginal approach, hysteroscopic resection or laparoscopic resection and repair of the uterine wall. While there is no clear consensus about general management of isthmocele, there are very few cases the medical literature of large isthmoceles, such as the one we describe. This case is interesting since no case of worsening of isthmocele following hysteroscopic treatment has been described so far.

Background

Cesarean section is one of the most frequent surgical procedures in women and its incidence is constantly rising in most developed countries [1]. In France, the rate of cesarean has been around 20% over the past decade [2]. One of the potential consequences of cesarean section is a defect of the myometrium at the site of the uterine scar, called a niche, isthmocele, uteroperitoneal fistula or diverticulum. This defect is a thinning or indentation of the myometrium at the anterior uterine wall. Most common symptoms include abnormal uterine bleeding, chronic pain and infertility. Treatment depends on the size of the defect, the symptoms and signs and the patient's desire for future pregnancies.

Case presentation

A 35-year-old woman was referred to our center for the management of a large cesarean scar defect. Her medical history showed a laparoscopic ovarian cystectomy for an endometrioma. She also had a history of two cesarean sections. Since the second cesarean section, she had presented with intermenstrual bleeding and chronic pelvic pain. Hysteroscopic examination showed a small cesarean scar defect on the anterior wall of the uterine isthmus. Operative hysteroscopy was performed with electrocoagulation of the defect. Afterwards, the patient reported persistent abnormal uterine bleeding, chronic pelvic pain and dyspareunia. Clinical examination showed a broad, flexible, anterior, round lesion. Magnetic resonance imaging (MRI) five years after the initial isthmocele treatment revealed cyst-like blood retention in a broad anterior isthmocele of 50x80 mm, in contact with the internal orifice of the cervix (Figure 1A). MRI confirmed the presence of posterior deep endometriosis suspected on clinical examination, which had found a large lesion behind the cervix invading the uterosacral ligaments, the torus and the rectosigmoid junction.

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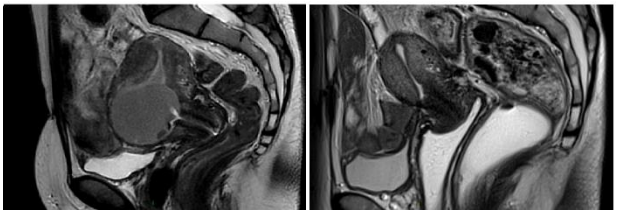
Since the patient wished to have more children, a conservative treatment was considered. Hysteroscopic examination revealed a wide pseudo-cavity totally occupied by thick reddish matter. Laparoscopy showed severe anterior bloating of the isthmic region corresponding to the cesarean scar defect previously described by MRI, as well as multiple pelvic adhesions. Adhesiolysis was attempted but failed due to the large uterine volume and the many posterior adhesions. Therefore, it was decided to use laparotomy to perform uterine adhesiolysis and scar defect resection, thus achieving complete uterus repair. After defect removal, the healthy myometrial edges were sutured with six cross-stitches of polyglactin. Postoperatively, the patient was given six-month GnRH analog treatment to block the menstrual cycle. We advised the patient to avoid attempting pregnancy for at least six months.



a **b**

Figure 1: (a) Before surgery **(b)** After surgery.

Six months after surgery, MRI showed complete repair of the scar defect, with a normal anterior uterine wall (Figure 1B). Postoperative hysteroscopic findings were also normal. Moreover, the patient reported total resolution of symptoms postoperatively, even after stopping GnRH analog treatment. So far, the patient has not attempted a new pregnancy. Isthmocele is a frequent complication that may occur after cesarean section. Its incidence has been rising in proportion to the number of cesarean deliveries. The reported prevalence varies between 24% and 84% in a random population of women with at least one cesarean delivery, depending on diagnostic criteria and method of detection [3]. In the medical literature, common risk factors for the development of cesarean scar defect include cesarean section with cervical dilatation ≥ 5 cm or labor duration ≥ 5 h, low (cervical) hysterotomy, single-layer uterine wall closure, use of locking sutures, closure of hysterotomy with endometrial-sparing technique, multiple cesarean deliveries, uterine retroflexion as well as patient-related factors that may impact wound healing, such as diabetes and smoking [3, 4].

Isthmocele is not always symptomatic, but the main symptoms and signs include chronic pelvic pain, abnormal bleeding, dyspareunia, dysmenorrhea as well as secondary infertility. In the case of pregnancy, there is higher risk of uterine rupture, ectopic pregnancy or abnormal placentation [3, 5]. Symptom severity seems to be correlated with the size and depth of the scar defect [6]. First-line diagnosis is made by transvaginal ultrasonography or saline-infusion sonohysterography. MRI, 3-D ultrasonography, and hysteroscopy are useful imaging modalities that can additionally aid in the diagnosis. Recently, European experts published their consensus guidelines to help standardize the definition of isthmocele. The following were proposed: measurement of the residual myometrial thickness on a sagittal view; measurement of the width of the niche on a transverse plane; the niche's definition as an indentation of at least 2.0 mm, classified as simple, simple with one branch, or complex (with more than one branch); necessity of measuring the distance between the vesicovaginal fold and top of the niche; measurement of the adjacent myometrial thickness [7].

The management of cesarean scar defects is not yet clearly defined. The technique of choice depends on the patient's symptoms and desire for future fertility. If there is no such desire, hysterectomy can be performed. If the patient wishes to maintain the possibility of a pregnancy, hysteroscopic resection, laparoscopic resection and repair or a vaginal approach can be performed in order to reduce symptoms arising from the defect [8-10]. In the current case, we describe a large isthmocele, the size of which dramatically increased after hysteroscopic resection. There is, to our knowledge, no such case described in the medical literature, since repair of an isthmocele after operative hysteroscopy is usually complete and successful [11-14].

A large isthmocele is defined by a residual myometrial thickness less than 50% or ≤ 2.2 mm when evaluated by transvaginal sonography and ≤ 2.5 mm when evaluated by sonohysterography or by a defect size greater than 2 cm. A total defect is defined as no remaining myometrium over the defect [3]. There are very few studies in the medical literature that include management of large isthmoceles. The current case is an example of successful management of an extremely large isthmocele. Laparotomy is not often described as an option for isthmocele treatment, but in difficult cases with lots of adherences and extremely large defects it appears to be a reasonable and effective solution.

Conclusion

Cesarean scar defects are becoming a frequent complication as the number of cesarean deliveries constantly rises. We report a very rare case of an extremely large uterine scar defect, which dramatically increased after hysteroscopic treatment. Management required the removal of the defect combined with a solid uterine suture by laparotomy.

Capsule

Successful resection by laparotomy of an extremely large isthmocele that appeared after hysteroscopic treatment.

Conflicts of interest

None.

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