

Isthmocele: An important sequelae of caesarean section - report of three cases and mini review

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CASE STUDY

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ABSTRACT

An isthmocele appears as a fluid-filled pouch-like defect in the anterior uterine wall at the site of a prior caesarean section, and ranges in prevalence from 19 per cent to 84 per cent, a direct relation to the increase in caesarean sections performed worldwide. It is the result of incomplete healing of the isthmic myometrium after a low transverse uterine incision performed for caesarean section. Although mostly asymptomatic, it may cause menstrual abnormalities, chronic pelvic pain, and secondary infertility. Scar tissue dehiscence, scar pregnancy, and abnormally adherent placenta are some of the obstetric complications associated with this defect. Diagnosis of the defects can be made with transvaginal ultrasound (TVUS), saline infused sonohysterogram (SIS), hysterosalpingogram, hysteroscopy, and magnetic resonance imaging (MRI). Surgical treatment

of an isthmocele is still a controversial issue but it should be offered to symptomatic women or asymptomatic patient who desires future pregnancy. When surgery is the treatment choice, laparoscopy guided by hysteroscopy, or hysteroscopy alone are the best options depending on the isthmocele's characteristics and surgeon expertise.

We would like to present a mini-review of the topic with contribution of three cases.

Key Words

Isthmocele, fluid-filled pouch, caesarean section

Implications for Practice:

1. What is known about this subject?

Isthmocele is a late complication of caesarean section.

2. What new information is offered in this case study?

It is very important to be diagnosed because it can lead to sterility.

3. What are the implications for research, policy, or practice?

All symptomatic cases with isthmocele have to be treated by laparoscopy or hysteroscopy and the same applies to all asymptomatic patients with desire for pregnancy.

Background

The incidence of birth after caesarean section (CS) increases on a global scale.^{1,2} This is due to decrease of the rate of operative vaginal deliveries, vaginal delivery of twin gestation, breech deliveries, and vaginal birth after caesarean section.³ On the other hand the World Health Organization accepts, that only incidence of caesarean

section of 10-15 per cent in the population results in decrease of maternal and foetal mortality, and this effect is not observed above these values.⁴ Some authors report of incidence of caesarean section of above 50 per cent, which results in increase of complications from it, without presence of benefit for the mother and foetus.^{5,6} These complications are of early (haemorrhage that required hysterectomy or massive blood transfusion; uterine rupture; cardiac arrest; acute renal failure; major infection; wound disruption was threefold increased for caesarean delivery as compared with vaginal delivery), and of late occurrence (infertility, pelvic adhesions, and pelvic pain that has been previously described).⁷

Among these complications is the caesarean scar defect, which is connected with various obstetrical and gynaecological problems - uterine rupture and ectopic caesarean scar pregnancy, spotting, dysmenorrhea, dyspareunia, or chronic pelvic pain.⁸⁻¹⁴ This defect increases the risk from complications with some gynaecological procedures as intrauterine device placement, evacuation and embryo transfer.^{14,15} The caesarean scar defect is called isthmocele - this term was used for the first time by H. Morris in the year 1995.¹⁶ It represents not completely well healed myometrium at the site of caesarean incision. The range of its incidence is from 24 per cent to 56 per cent.¹² Risk factors for its emergence are history of multiple complicated deliveries (CDs), advanced stage of labour and uterine retroflexion.^{17,18}

We represent three cases of isthmocele, which were treated at our Clinic in the period from January 1, 2018 to December 31, 2018.

Case details

First clinical case

This is a 22-year-old patient with no concomitant diseases and previous operations, apart from a caesarean section 14 months ago. There were no previous vaginal deliveries and no abortions were reported. Menstruations have become excessive, painful, and more prolonged for 8 months. Echogenic zone with sizes of 1×3cm was found from the ultrasound examination over anterior uterine wall in the area of surgical scar (Figure 1).

After performing of standard preoperative preparation, hysteroscopy was performed, which found a pseudo-cavity in the scar tissue. The latter cavity was resected. Patient was discharged on the same day. The menstruations normalized two months after that and still are normal twelve months later.

Second clinical case

This is a 28-year-old patient with no concomitant diseases and no previous surgical operations apart from a caesarean section 21 months ago. There was one previous normal vaginal delivery, and no abortions were reported.

Due to excessive and painful menstruations, ultrasound examination was performed, which found a very pronounced echogenic formation over the anterior uterine wall with sizes of 2×3cm. After performing of standard preoperative preparation, hysteroscopy was performed, which found a defect in the scar tissue. The latter defect was resected. Patient was discharged on the same day. The menstruations normalized two months after that and are still normal ten months later.

Third clinical case

This is a 20-year-old patient with no concomitant diseases and no previous operations apart from a caesarean section eight months ago. There were no previous vaginal deliveries and no abortions were reported. There has been slight genital bleeding for four months. A very pronounced echogenic formation over anterior uterine wall with sizes of 1×2cm was found after ultrasound examination performed. After performing of standard preoperative preparation, hysteroscopy was implemented, which found a defect in the scar tissue. The latter defect was resected. Patient was discharged on the same day. The above-mentioned bleeding with spotting disappeared, and the menstruations were normal for a period of eight months from then until now.

Discussion

The isthmocele is a specific complication, which occurs as a result of caesarean section. With the increase of incidence of CS, the frequency of isthmocele also increases. It varies in a wide range according to different authors - from 24 per cent to 56 per cent.¹²

This complication has various clinical manifestations, while the most frequent among them are postmenstrual spotting, chronic pelvic pain, and secondary infertility. The most frequent symptom of patients with caesarean scar defect (CSD) is postmenstrual abnormal uterine bleeding, which is often described as leaking of dark blood. There was postmenstrual bleeding in 33.6 per cent of women with isthmocele, while there is a direct connection between the size of defect and the quantity and duration of bleeding. It is accepted that the reason for that is the retention of blood in the defect and disturbed contractile ability of the impaired myometrium.¹² Other symptoms are dysmenorrhea (with

incidence of 53 per cent), chronic pelvic pain (39.6 per cent), and dyspareunia (18.3 per cent).¹⁸ They are explained with chronic inflammation, presence of small polyps and/or lymphocytic infiltration present in the scar. The secondary infertility is also connected with CSD, while it is explained with disturbance of the passage of spermatozoa and the difficult implantation of embryo.

The diagnosis of isthmocele is made based on previous medical history of caesarean section, clinical presentation, and ultrasound and/or hysteroscopy. Currently, there is lack of consensus on the definition of caesarean scar defect.¹⁹

The ultrasound examination is usually the first and most frequently used method for making of the diagnosis. The isthmocele is defined by the presence of an anechoic area at the site of a previous caesarean section; it usually has triangular shape with the vertex toward the isthmus. Another criterion for the diagnosis is the presence of fluid within the incision site.¹⁴ The most suitable moment for performing of the examination is the late proliferative phase, in which the cervical mucus may fill up the defect. The use of 3D ultrasound helps making of the diagnosis. Currently, contrast enhanced ultrasonography is considered to be the gold standard in isthmocele diagnostics.¹¹

Another method is the hysterosalpingography, but in this case this is an occasional finding. The presence of anatomic defects as diverticula or thin linear defects at the lower uterine cavity is a common finding in patients with a previous caesarean section, and this defect can be found in about 60 per cent of patients with the use of hysterosalpingography.²⁰

The diagnostic hysteroscopy allows direct visualization of the defect, which presents as a pseudo-cavity.

Magnetic resonance (MRI) may also be used as a diagnostic option. The MRI display a linear low-signal niche, sometimes filled with some fluid collection.¹⁹

Some classifications are used for determination of the grade of isthmocele. One of them is the classification of Gubbini et al.,²¹ in which the surface of the defect is measured using its thickness and width. The defect is determined as grade 1 when it is less or equal to 15mm³, grade 2 - with a surface between 16 and 25mm³ and grade 3 - when larger than 25mm³. In their initial report, they found that more than 55 per cent of cases were of grade 1. The second classification is the one of Ofili-Yebovi et al.,¹⁸ which is based on the measurement of the endometrial thinning at the caesarean

defect; the authors defined the degree of thickness by the ratio between the myometrial thickness at the level of the defect and the thickness of the adjacent myometrium, and defined a severe defect as a ratio >50 per cent and dehiscence as a ratio equal or more than 80 per cent.

Other authors have defined CSD as severe when the remaining myometrium is less than 2.2mm visualized by vaginal ultrasound or 2.5mm in women who undergo sonohysterogram.¹⁷

With respect to the therapy of this condition, there is no unanimous opinion what it should be. Different operative approaches are offered, but one must be familiar with the fact that they are used only in symptomatic patients, and thereafter it is only proper to be taken oral contraceptives for several months for reduction of menstrual bleeding.

The use of contraceptives without other medications is controversial. Some authors consider that it does not reduce bleeding, while others hold the contrary opinion.²² There is no consistent study about the use of hormonal intrauterine device to treat this condition.¹⁹

At the present moment, different surgical methods are used in the practice for correction of isthmocele. Most frequently this is resectoscopic hysteroscopic surgery. It was used for the first time by Fernandez et al., who resected the fibrotic tissue of the inferior part of the scar and thus improved the drainage of menstrual blood.²³ Unfortunately, this approach bears the risk of perforation of uterus and injury of urinary bladder, and that is why some authors recommend abstaining from its use with myometrium, localized above the defect, of less than 2mm.²⁴

Another operative option is the laparoscopic correction – it represents excision of the defect and its recovery as far as non-affected tissues, while this approach increases the thickness of myometrium and this occurs under visual control reducing the risk of complications.²⁵

The vaginal approach is used most rarely – just like the laparoscopic approach, it is a repairing operation, which recovers the defect and increases the thickness of the uterine wall. It represents opening of the cervico-vesical space and the dissection of the bladder, the scar is opened, and the fibrotic tissue is excised. The opened scar is then closed with 2 layers of suture.²⁶

As far as our patients are concerned, in two of the cases there was dysmenorrhea with pronounced excessive

menstruation, and in another one - postmenstrual uterine bleeding. In the three cases the diagnosis was made based on ultrasound examination, and confirmed by hysteroscopy. The treatment included hysteroscopic resection of the defect. The results were excellent while the normalization of menstruations occurred within two months.

Conclusion

With the increase of incidence of caesarean section, the frequency of complications connected with it increased as well. Every clinician must be familiar with them and take them into consideration when a decision is made for the type of parturition. The isthmocele is a condition, which may be completely harmless, but also may result in serious consequences. That is why the specialists in obstetrics and gynaecology must not only be able to diagnose it, but to adequately treat it.

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Figure 1: Aechogenic zone with sizes of 1×3cm over anterior uterine wall in the area of surgical scar



PEER REVIEW

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CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

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PATIENT CONSENT

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1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.