
Isthmocele in a retroflexed uterus: a report of an unrecognized case

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Summary

The term “isthmocele” refers to a niche on the anterior wall of the uterine isthmus or of the cervical canal at the site of a previous cesarean delivery scar. Such anatomic defect can cause many gynecologic sequelae that only recently have been identified and described. Hysteroscopy is commonly considered the gold standard for the diagnosis and also for the treatment, at least in the case of defects of small size. The authors described the case of a 37-year-old woman who underwent a cesarean section (CS) seven years before, with a long lasting history of menstrual irregularities, and pelvic pain increasing during menstruation at the hypogastric level. Magnetic resonance imaging (MRI) showed an exceptionally large isthmocele on the anterior wall of a retroflexed uterus which was otherwise misinterpreted as the uterine cavity filled with menstrual blood during a previous hysteroscopy (HSC). Although exceptional, this case highlights the possibility that a large sized isthmocele in a retroflexed uterus could be misinterpreted as the uterine cavity filled by menstrual blood at HSC. In this case MRI definitely clarified the diagnosis.

Key words: Isthmocele; Magnetic resonance imaging; Retroflexed uterus; Hysteroscopy.

Introduction

According to the latest global estimates, approximately 15% of deliveries take place by cesarian section (CS), with about 20 million CS deliveries occurring each year worldwide [1]. Such proportion has steadily increased in almost all middle- and high-income countries over the last three decades. Latest estimates indicate that in 2009, 39% of all women in Italy delivered by CS [2], making it the European country with the highest rate of CS.

Nearly half of the women may incur a poor uterine scar healing after CS which can cause anatomic defects of the uterine cavity of various degree [3]. Such defect may lead to gynecologic sequelae that only recently have been identified and described. Besides the most feared complications of cesarean scar pregnancy and uterine rupture during subsequent pregnancy and labor, also abnormal bleeding, pelvic pain, and infertility, have all been described as possible consequences [4]. Such anatomic defects can have different shapes such as thin linear defect, focal saccular outpouching, unilateral or bilateral diverticula and fistula, and different locations such as the uterine body, lower uterine segment, uterine isthmus and the upper endocervical canal [5]. Particularly the term “isthmocele” refers to a niche on the anterior wall of the uterine isthmus or of the cervical canal at the site of a previous cesarean delivery scar.

The authors report the case of a woman with a large isthmocele found on magnetic resonance imaging (MRI)

which was otherwise undiagnosed during a previous hysteroscopy (HSC).

Case Report

In November 2013, a 37-year-old woman came to the Department of Gynecology and Obstetrics of the present Institute. She was complaining of menstrual irregularities and intense pelvic pain increasing during menstruation at the hypogastric level. These symptoms began about four years before.

The obstetric history of the patient revealed that she underwent dilatation and curettage under the diagnosis of missed abortion at seven weeks, in March 2006. In October 2007, the patient underwent CS for breech presentation. In August 2010 a HSC was performed due to an episode of vaginal hemorrhage. Hematometra was found and endometrial ablation was performed. In October 2012, a second episode of vaginal bleeding occurred, and once again hematometra was found at HSC. Also a thickening of the endometrium of the posterior wall of the uterine body was described. Endometrial ablation was again performed and a histopathologic diagnosis of disordered proliferative endometrium was made. Despite such intervention of endometrial curettage, the patient never experienced a relief of the symptoms.

In November 2013 a transvaginal-ultrasound (TVUS) was performed. The exam was partially hampered by the clinical condition of the patient and the intestinal bloating. It showed an inhomogeneous, mainly hypo-echoic, and ill-defined area, suggesting the presence of a partially organized fluid collection within the pelvis. Because of the results of previous HSC, the unclear findings on TVUS, and the refuse of the patient to undergo further endocavitary examinations due to the pelvic pain, she was

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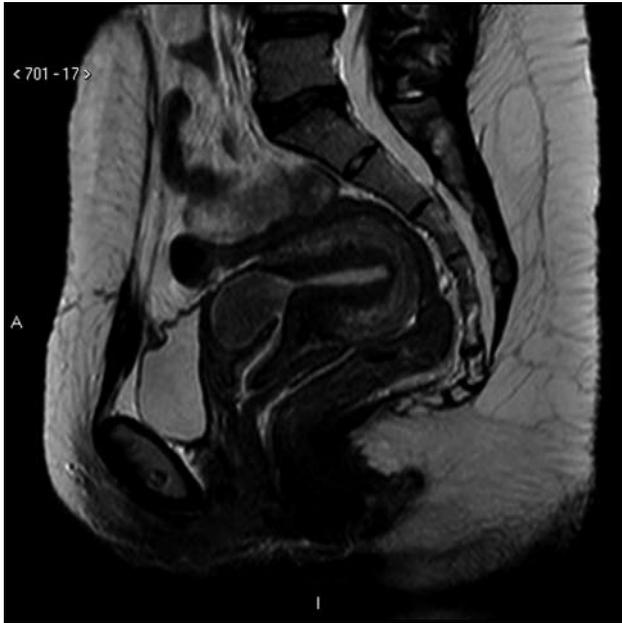


Figure 1. — Sagittal T2-weighted image clearly showing a retroflexed uterus. In the anterior wall of the uterine isthmus, at the site of a previous cesarean delivery scar, there is a pouch (35 mm deep). In this site the uterine wall is represented only by a very subtle layer of myometrium with a thin layer of endometrium lining the inner wall of the pouch. The sacular pouch appeared in continuity with the endometrial line by a neck of 15 mm.

referred to the authors' Department of Diagnostic Imaging for a pelvic MRI.

MRI was performed on 1.5 T scanner using a surface coil (phased array pelvic coil). MRI was performed during the third day of the menstrual cycle. The authors' imaging protocol consisted of oblique-axial (perpendicular to the major axis of the uterus) T1-weighted imaging (TR 487 ms/TE 10 ms, four-mm section thickness, 0.4-mm gap, 484 x 420 matrix, 41 cm FOV, and two signal averages), T1-weighted spectral fat saturation inversion recovery (SPIR) obtained in oblique-axial (TR 912 ms, TE 15 ms, four-mm section thickness, 0.3-mm gap, 484 x 420 matrix, 41 cm FOV, and two signal averages) and sagittal planes (TR 908 ms, TE 15 ms, four-mm section thickness, 0.3-mm gap, 484 x 420 matrix, 28 cm FOV, and two signal averages), and T2 weighted imaging obtained in oblique-axial (TR 3000 ms, TE 90 ms, four-mm section thickness, 0.4-mm gap, 41 cm FOV, and two signal averages) and sagittal plane (TR 3239 ms, TE 90 ms, 3.5 mm section thickness, 0.4-mm gap, 24 cm FOV, and four signal averages).

Sagittal images (Figures 1 and 3) clearly show a retroflexed uterus. A pouch within the anterior wall of the uterine isthmus at the site of a previous cesarean delivery scar, 35 mm deep, is clearly observed on sagittal and transverse views of T2-weighted images (Figure 1 and 2) where the uterine wall is represented by a very thin layer of residual tissue, consisting only of a very subtle layer of myometrium and a thin layer of endometrium lining the inner wall of the pouch, and continuing with the endometrium bordering the uterine cavity. On sagittal T2-weighted images, it is also evident the scar of the CS. By using T1-weighted images with saturation of fatty tissue, hypersignals were detected in the uterine cavity and in the niche suggesting the retention of menstrual blood in the hystmocele (Figure 3).



Figure 2. — Oblique-axial T2-weighted image showing a hyperintense niche that extends forward in the opposite direction to the uterine cavity (being the uterus retroflexed).

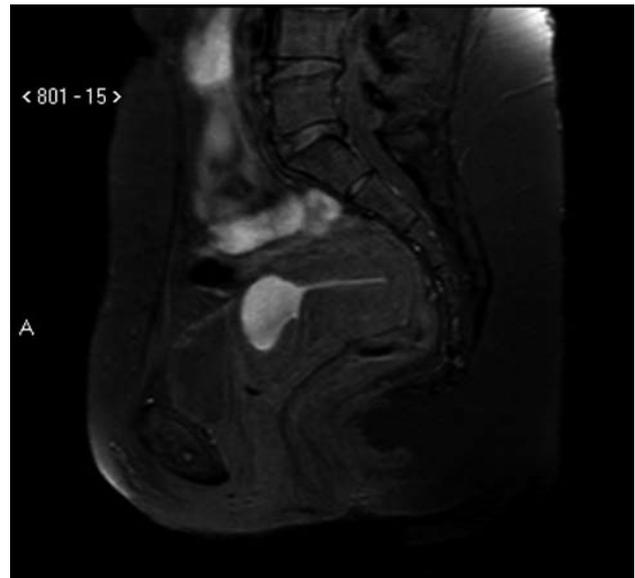


Figure 3. — By using T1-weighted images with saturation of fatty tissue, the niche appears inhomogeneously hyperintense (the signal in the declive portion is slightly inhomogeneous and slightly less hyperintense). This feature is suggestive of menstrual blood retention in the niche. A thin hyperintense line is also evident in the uterine cavity suggestive of menstrual blood.

Discussion

As the incidence of CS is on the increase worldwide, the complications associated with them are becoming more common. Therefore the effect of postcesarean complications on women's health is a focus of increasing attention.

Here the authors described the features on MRIs of a very large isthmocele (35 mm deep) in a woman with a history of cesarean delivery seven years before, who complained of pelvic pain increasing during menstruation and intermenstrual bleeding. This case is particular not only for the impressive dimension of the niche, indeed in a series of 44 symptomatic women described by Monteagudo *et al.*, the largest niche was 11.5 mm deep [6], but also because MRI allowed the diagnosis of the isthmocele which was otherwise unrecognized at HSC. It is widely accepted that HSC is the “gold standard” for cesarean scar defect (CSD) assessment, in fact it enables confirming diagnosis and assessing treatment [7, 8]. In this case the isthmocele was unrecognized at HSC. This was probably due to the retroflexed position of the uterus and the large size of the CSD. Indeed, due to its location within the anterior wall of the uterus, the pouch of the isthmocele was misinterpreted as the uterine cavity.

A recent systematic review by Roberge S *et al.* showed that hystero-graphy, sonohysterography (SHG), or TVUS can all detect uterine scar defects, with SHG having the higher detection power [3]. With ultrasound, the thickness of the residual myometrium, the thickness of myometrium bordering the scar, and the depth of the filling defect in the scar can all be depicted [9]. However in this case not even the TVUS allowed to clarify the diagnosis, perhaps because of some conditions that hampered the examination, such as the presence of pelvic pain and the intestinal bloating.

Finally MRI made clear the diagnosis of hystmocele. MRI also depicted the presence of functioning endometrial tissue within the walls of the isthmocele, the hypertrophic fibrotic scar which hampered the outflow of the blood at the ostium of the niche, and the accumulation of blood within the pouch. Moreover MRI, allowing a panoramic view, provided a clear depiction of the relationship between the niche and surrounding organs.

A widely accepted approach for the treatment of CSD has not been developed. Surgical techniques for repair of CSD include laparoscopic excision, resectoscopic treatment, vaginal revision, and endometrial ablation. The treatment should be individualized according to the symptoms, the sac size, and the patient’s desire for future fertility. Hysteroscopic surgical procedures are aimed at resecting the fibrotic tissue that sometimes hangs below the scar, thereby improving menstrual drainage and avoiding blood accumulation. Furthermore HSC enables the fulguration of superficial dilated blood vessels or endometrium-like glands inside the diverticulum to avoid the *in situ* production of fluid or blood. The procedure had to be performed with special care to prevent perforation of the isthmus [10]. Such risk is expected to be particularly high when the defects is deep with only a thin layer of residual myometrium, particularly when fulguration procedures are needed to destroy endometrial tissue within

the wall of the pouch. In such cases surgical procedures may be preferable. MRI can be especially useful when surgery must be planned, because the correct topography of the lesion is clearly depicted together with possible associated changes in the surrounding tissue and structures.

In a recent review Tower *et al.* observed that CSD are an under-recognized cause of abnormal uterine bleeding and other gynecologic complications [4], and this case confirms such remark.

Although exceptional, this case highlights the possibility that a large sized isthmocele in a retroflexed uterus could be misinterpreted as the uterine cavity filled by menstrual blood at HSC. In this case, MRI definitely clarified the diagnosis.

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