# Ankh-Habashy Sign, A Novel Sonographic Marker for Isthmocle using 3D-TVUS-Surface Mode-An observational study and review of literature

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#### **Objectives**

The isthmocele or cesarean scar defect (CSD) is an evolving iatrogenic long term complication of cesarean section (CS). It represents an incomplete healing of the CS scar that leads to the presence of a wedge shaped anechoic area of myometrial discontinuity at the CS site with at least 2 mm depth. The aim of our study is to evaluate the shape of the uterus in the coronal plane in cases that has isthmocele using the surface mode of the three-dimensional transvaginal ultrasound (3D-TVUS). To our best knowledge this is the first study to describe the coronal sonomorphology of the CSD using 3D-TVUS.

# **Methods**

This retrospective observational study that enrolled 84 cases over a period of 2 years (from august 2017- august 2019) from a single tertiary center. All are in the reproductive age window: 18-42 years old. All had complained AUB in the form of postmenstrual spotting with or without chronic pelvic pain (CPP) and/or secondary infertility. All cases are scanned postmenstrual by the same sonographer using TVUS. Ishmocele is defined as a wedge shaped hypoechoic area of myometrial discontinuity at the CS site in the sagittal plane of the uterus with a depth of  $\geq$  2mm. We measured the residual myometrial thickness (RMT) and the adjacent myometrial thickness (AMT) in all cases. After obtaining an optimal 2D sagittal uterine view, the 3D box was set to include the whole uterus from the fundal serosa till the external OS. Our target was to produce a panoramic volumetric image of the uterus that include the cavity above, the cervical canal below and the isthmus in between.

#### Results

We noticed a lateral bulge or shadow at the level of the isthmus in 82.1% of cases of isthmocele. The lateral bulge produced by the isthmocele simulates the "ankh"; an ancient Egyptian hieroglyphic symbol of life. We call this sonomorphology the "Ankh-Habashy sign". The mean age of patients was 29.5 years. The mean number of previous CS was 2. The mean interval from the last CS was 3 years. All cases were complained of AUB in the form of postmenstrual spotting. 42.9% of cases had an associated

secondary infertility. 66.7% of cases had an associated chronic pelvic pain (CPP). 56% of the cases were RVF uterus, 26% were erect uterus and 19% were AVF uterus.

## Conclusion

Lateral bulge at the level of the isthmus in the coronal panoramic view of the uterus using 3D-TVUS-surface mode; "the Ankh-Habashy sign" is a reproducible sonographic sign that is present in most cases of isthmocele.

# INTRODUCTION

The isthmocele or cesarean scar defect (CSD) is an evolving iatrogenic long-term complication of cesarean section (CS). It represents an incomplete healing of the CS scar that leads to the presence of a wedge shaped anechoic area of myometrial discontinuity at the CS site with at least 2 mm depth. Isthmocele incidence is varied among literature according to the methods used for diagnosis; namely sonography, sonohystrography (SHG), hystrosalpingography (HSG) or hystoroscopy. CSD is present in at least fourth of those having previous CS (using TVUS) and in at least half of them (using SHG).(6-9) Although the incidence looks high, only about third of them are symptomatic. Symptomatic CSD cases have a variable spectrum of complains that are grouped under the "Cesarean Scar Syndrome – CSS".

The causes of the abnormal uterine bleeding (AUB) that happened in those with CSD are retention of menstrual blood in the niche, defective myometrial contractility at the scar or impaired menstrual drainage due to fibrosis below the niche.

Various risks had been proposed for the development of the isthmocele. Though isthmocele is not uncommon, it is commonly missed during routine gynecological sonographic scanning. In such cases index of suspicion should be raised by the history especially postmenstrual spotting and that directs the examiner to focus on the isthmus. The best time for diagnosis of CSD is postmenstrual using the transvavinal ultrasound (TVUS) focusing on the area of the isthmus; between the lower part of the corpus and the internal cervical os. CSD appears as a hypoechoic triangle with its base toward the cavity and the apex toward the anterior wall of the isthmus. Less common shapes are V-shape, U-shape, semicircle, droplets, rectangle or

circle. The aim of our study is to evaluate the shape of the uterus in the coronal plane in cases that has isthmocele using the surface mode of the three-dimensional transvaginal ultrasound (3D-TVUS). To our best knowledge this is the first study to describe the coronal sonomorphology of the CSD using 3D-TVUS.

## **METHODS**

This is a retrospective observational study that enrolled 84 cases over a period of 2 years (from august 2017- august 2019) from a single tertiary center. All are in the reproductive age window: 18-42 years old. All had complained AUB in the form of postmenstrual spotting with or without chronic pelvic pain (CPP) and/or secondary infertility. AUB was defined as uterine bleeding not related to pregnancy nor menses. CPP was defined as pain in the pelvic region those last at least 6 months. Secondary infertility was defined as failure of conception for 12 months or more with regular intercourse without the use of contraception in those who already get pregnant before. All cases were scanned postmenstrually by the same sonographer using "Samsung, H-60, 5-9 MHz TVUS probe". Ishmocele is defined as a wedge-shaped hypoechoic area of myometrial discontinuity at the CS site in the sagittal plane of the uterus with a depth of  $\geq$  2mm. We measured the residual myometrial thickness (RMT) and the adjacent myometrial thickness (AMT) in all cases. RMT is the thickness of the overlying myometrium overlying the niche, i.e. the thinnest myometrial thickness from the apex of the niche till the serosa. AMT is the thickness of the normal myometrium adjacent and cranial to the defect (figure 3). After obtaining an optimal 2D sagittal uterine view, the 3D box was set to include the whole uterus from the fundal serosa till the external os then the green line manipulated till it was near the endometrial interface then acquisition started using the surface mode with volume angle set at 800 and the sweep quality set at high. If the coronal view of the uterus was suboptimal because the endometrial interface and the cervical canal were not aligned well, we took another volume with green line manipulation and/or suprapubic pressure to align them to generate a better view. Our target was to produce a panoramic volumetric image of the uterus that include the cavity above, the cervical canal below and the isthmus in between. Postacquisition manipulation using magic cut had been used to produce a better spatial perception. Our data were analyzed using IBM SPSS software package version 20.0. (Armonk, NY: IBM Corp). Qualitative data were described using number and percent. Quantitative data were described using mean, standard deviation and median (minimum and maximum). Significance of the obtained results was judged at the 5% level. Chi-square test for categorical variables to compare between groups. Fisher's Exact or Monte Carlo correction for chi-square when more than 20% of the cells have expected count less than 5. Student t-test for normally distributed quantitative variables to compare between two groups. Mann Whitney test for not normally distributed quantitative variables, to compare between two groups.

# RESULTS

We noticed a lateral bulge or shadow at the level of the isthmus in 82.1% of cases of isthmocele. We used the 3D magic cut to remove artifacts around the cavity, the isthmus and the cervix in order to produce a panoramic coronal view of the whole uterus. The lateral bulge produced by isthmocele simulates the "ankh"; an ancient Egyptian hieroglyphic symbol of life (We call this sonomorphology the "Ankh-Habashy sign". 72% of cases where the sign was present it was apparent after the first 3D acquisition, and the remaining (29%) needed more than one acquisition to align the endometrial interface with the cervical canal.

The mean age of patients was 29.5 years (range 18-42years). The mean number of previous CS was 2. The mean interval from the last CS was 3 years (range 1-7 years). All cases were complained of AUB in the form of postmenstrual spotting. 42.9% of cases had associated secondary infertility. 66.7% of cases had an associated chronic pelvic pain (CPP). 56% of the cases were retroverted retroflexed uterus (RVF), 26% were erect uterus and 19% were anteverted anteflexed uterus (AVF). The previous variables are presented. 63.1% of our cases were had a small isthmocele (i.e. RMT ≥50% of AMT) and 36.9% of them were had a large isthmocele (i.e. RMT < 50% of AMT). All cases who had a large isthmocele displayed the Ankh-Habashy sign and 71.7% of those who has a small isthmocele showed the sign. Age of the patients and uterine direction were not statistically significant factors for thedetection of Ankh-Habashy sign; p values of them were 0.294 and 1.0 respectively. Number of previous CS and the interval from the last CS were statistically significant factors for Ankh-Habashy sign detection; p values of them were 0.010 and 0.002 respectively.

# DISCUSSION

Though CSD was first described by Morris et al. (based on pathological findings) in 1995, the increasing body of literature focusing on this iatrogenic uterine pathology in the last two decades was paralleled by a similar increasing CS rates worldwide during this period. CSD had been described by different synonyms in literature; e.g. CS niche, dehiscent (or deficient) CS, trans-mural hernia, CS diverticulum, CS pouch or CS sacculation.

The earliest study for the CS scar by the TVUS was by Chen et al. in 1990 when they reported a possible presence of a wedge shaped defect at the site of the CS scar. Monteguado et al. in 2001 was first who described the anechoic loss of myometrial continuity at the CS site as a "niche". Later on, in 2003; Fabres et al. had described this wedge defect as a triangular anechoic area with an apex that point anteriorely. Gubbini et al. on 2008 was first who described the CSD as an "isthmocele". Since when, several studies reported different shapes of the isthmocle by the TVUS; e.g. semicircle, droplets, rectangle or circle. The exact prevalence of the isthmocle is unknown; it had been proposed to be present in 24-70% of those who had previous CS when assessed by TVUS and in 56-84% when assessed by SHG. The cause of the higher CSD prevalence (and also a larger defect size) when using SHG as compared to the TVUS is the increasing intrauterine pressure in SHG that will enhance the CSD detection.

Multiple risk factors had been implicated for CSD development. Ofili-Yebovi et al considered two of them as the principle risk factors; namely multiple previous CS and RVF uterus. Other risk factors are suggested by Bij de Vaate et al for potential predisposing factors for ishtmocele development but their cause-effect relationships were still inconclusive. They are CS in active labor, CS done when the cervix is dilated 5 cm or more and single layer closure. Only about third of cases that have an isthmocle are symptomatic. Symptomatic CSD cases have a variable spectrum of complains that are grouped under the "cesarean scar syndrome- CSS", this term was first described by Morris et al. in 1996. CSS encompasses gynecological and/ or obstetrical abnormalities. Gynecological symptoms include AUB (mainly in the form of postmenstrual spotting), pain and/ or secondary infertility. AUB is the commonest complain of these cases. Pregnancy-related complications of CSD include cesarean scar pregnancy (CSP), placental attachment disorders (PAD) or rupture uterus. Most studies that targeted the niche sonography were focused on its diagnosis (using 2D-TVUS or SHG) or its dimensions. Few studies focused on its 3D-TVUS assessment. Ludwin et al. evaluated the uterine niche using 3D saline contrast sonohystrography (3D-SCSH) and Sonographybased Automated Volume Count software (SonoHystroAVC) in order to calculate the actual niche volume and incorporated it in

a scoring system to improve the reliability of sonomorphology of uterine scar defects. Although the coronal views generated by them using these software produced the Ankh-Habashy sign, they did not described it. In comparison to our study, we thought that their evaluation had 5 limitations. Four limitations are related to the use of the saline contrast which are; increase the scan time, more painful, fear of interruption of early pregnancy and the need of an assistant to inject the contrast. Last limitation is the need of AVC software that not present in all 3D-TVUS transducers. Moreover their proposed cut-offs were still arbitrary or clinically irrelevant.

To our best knowledge our study is the 1st one to describe the characteristic shape of the cesarean scar defects in the coronal plane using the 3D-TVUS-surface mode. We think that this sign is simple, reproducible and easily implemented in clinical practice. The main limitation of our study was that we did not compare Ankh-Habashy sign with sonohystrography (2D or 3D) to reach a reliable detection rate.

#### **CONCLUSION**

Lateral bulge at the level of the isthmus in the coronal panoramic view of the uterus using 3D-TVUS-surface mode; "the Ankh-Habashy sign" is a reproducible sonographic sign that is present in most cases of isthmocele. It was present in 82.1% of our cases. All cases with large isthmocele showed the sign. The higher the number of previous CS and the longer the interval from the last CS were significant predictors for sign detection. Further studies are needed to compare this sign with the isthmocele detection using 2D and 3D saline infusion sonography (SIS).