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Case Report

Caesarean scar ectopic pregnancy management with simultaneous isthmocele repair

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ABSTRACT

Caesarean scar ectopic pregnancy (CSEP) is a very rare form of ectopic pregnancy. If not diagnosed and treated accordingly it can lead to life-threatening complications. Hitherto there is no standardised treatment established because of the rarity and diversity of the disease. We present the case of a 35-year-old third gravida primi para with caesarean scar pregnancy in 7th week of gestation. Since the patient declined a Methotrexate treatment, we performed hysteroscopic and laparoscopic resection of the ectopic pregnancy in combination with repair of the isthmocele. Because human chorionic gonadotrophin (hCG)-level showed no proper decline three weeks after the initial surgery a second look hysteroscopy and laparoscopy were performed with laparoscopic injection of MTX around the uterine suture, resulting in a full decline of hCG-level. The ultrasound performed before discharge shows a well-adapted uterine scar. A pregnancy can be aspired six-month post-surgery.

Keywords: Caesarean scar ectopic pregnancy, Caesarean scar defect, Isthmocele

INTRODUCTION

Caesarean scar ectopic pregnancy (CSEP) is the rarest of the ectopic pregnancies and thus an uncommon condition.[1] Nevertheless, since the initial description in 1978 by Larsen et al, one can find an increasing number of case reports, case series and reviews on the subject with currently almost 700 entries in the search engine Pubmed. This may be due to the fact that a CSEP can lead to serious complications such as hemorrhage or uterine rupture with consecutive hysterectomy, or even the death of the patient, especially if not recognized in time or treated incorrectly.[3,4] On the other hand, the frequency of cesarean sections has been increasing worldwide over the last decades, making complications such as CSEP more common and thus more relevant, and the desire for a structured algorithm for diagnosis and treatment more pressing.[5]

There are numerous therapeutic approaches.[4] These range from expectant management with reported full-term births, over drug-based local or systemic therapy (methotrexate (MTX), mifegyne, or local injection of potassium chloride), interventional therapies (high-intensity focused ultrasound (HiFu) or uterine artery embolization), vaginal procedures (dilation and curettage, suction curettage, hysteroscopic resection), laparoscopic or laparotomic resection and repair of the uterine defect to hysterectomy.[3,5-17] Often a combination of these therapeutic approaches is chosen. The chances of therapeutic success vary widely and depend on many factors such as: type of CSEP (type I/endogenous type: with growth towards the uterine cavity or type II/ exogenous type: with growth towards the abdominal cavity), gestational age, hCG-level, sonographic pattern of vascularization and vitality of pregnancy.[18,19]

CASE REPORT

We report the case of a 35-year-old third gravida, primipara with one miscarriage with curettage in 2017 and one full-term delivery via caesarean section also in 2017.

The patient is referred to us in the 7th week of gestation with inadequate hCG-levels and mild vaginal bleeding. Via ultrasound, a 3cm wide amniotic-cavity with amniotic sac can be shown, located in the thinned caesarean scar (Figure 1). The hCG-level at first consultation is 12000 mIU/ml.

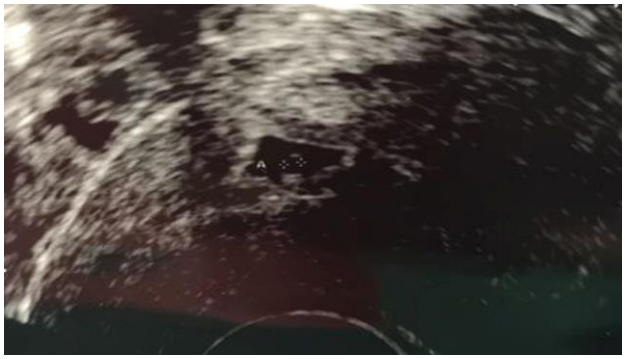

Figure 1: Pre-surgery ultrasound showing amniotic sac and isthmocele.

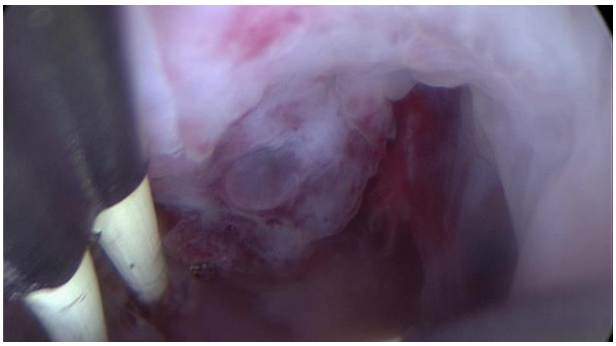

Figure 2: Hysteroscopy showing the amniotic sac penetrating the uterine cavity.

After laying out the possible therapeutic options, the patient decides for a surgical approach. Despite detailed consultation, a treatment with methotrexate is initially rejected. Since the patient still has the desire to have a child, uterine defect repair is agreed upon in the same procedure. The surgery is scheduled as combined hysteroscopy and laparoscopy.

During surgery the ectopic pregnancy can be visualized both hysteroscopically and laparoscopically (Figure 2). After laparoscopic exposure and adhesiolysis of the bladder, a partial loop-resection is performed hysteroscopically via bipolar resectoscope, followed by laparoscopic excision of the remaining tissue using a monopolar needle (Figures 3-5). Finally, the uterine defect is adapted with continuous suture with barbed filament (V-lock) (Figure 6). The blood loss during the procedure is negligible. The hCG-level two days after surgery is 2959 mIU/ml and the patient is discharged three days after surgery in good health. The ultrasound that was performed before discharge shows a well-adapted uterine scar with no evidence of an isthmocele or amniotic cavity (Figure 7).

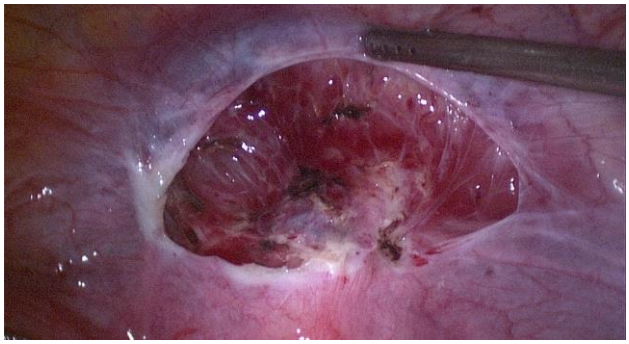

Figure 3: Laparoscopy showing the scar-pregnancy as livide discoloration in the isthmocele after adhesiolysis of the bladder.

Figure 4: Hysteroscopy after removal of the amniotic sac via bipolar loop.

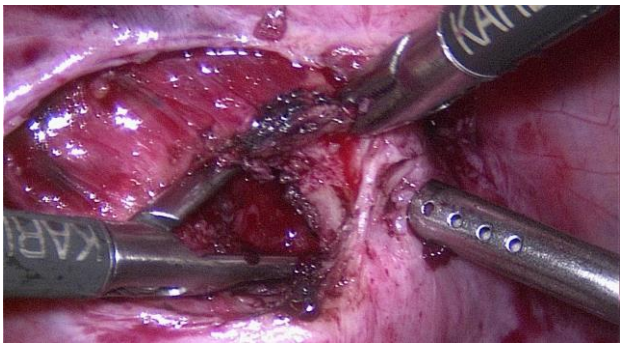

Figure 5: Laparoscopy after excision of the isthmocele area.

Figure 6: Laparoscopy after suture of the uterine defect.

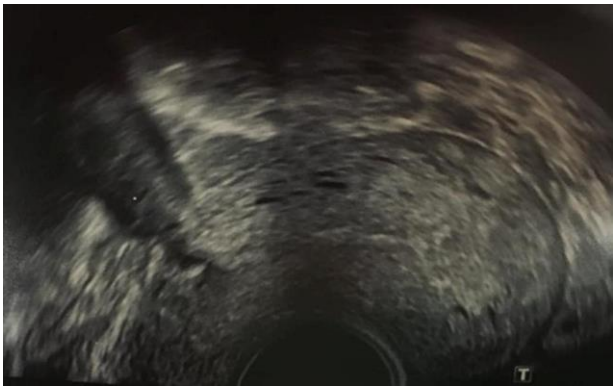

Figure 7: Post-surgery ultrasound.

Histologically, pregnancy material can be found in both the hysteroscopically and laparoscopically obtained tissue. In addition, adenomyosis is detected in the resected tissue obtained laparoscopically. The hCG-level one-week post-surgery shows a further decrease to 816 mIU/ml.

The patient is referred to the gynecologist in private practice for further monitoring. During follow-up, the gynecologist detects an increase in hCG-levels and refers the patient back to us. After an initial period of expectant management under observation of hCG-levels an increase up to 1975 mIU/ml can be shown without any specific ultrasound findings. In agreement with the patient, a second look hysteroscopy, laparoscopy and local injection of methotrexate around the uterine scar is scheduled almost three weeks after the first surgery, since the patient still does not want systemic methotrexate administration. The hysteroscopy shows a well-adapted uterine scar, but some loosened villous tissue in the scar area, which is obtained via curettage (Figure 8). Laparoscopically, the suture is also perfectly adapted, and there is no evidence of pregnancy remnants. Methotrexate 50 mg/m^2 body surface is injected around the suture (Figure 9). Post-surgery the patient does not report any side effects caused by the Methotrexate application nor does she have surgery related complaints. Over the course of the following three weeks, the hCG-level decline under the limit of detection (Figure 10). In the tissue obtained via curettage further pregnancy tissue can be verified.

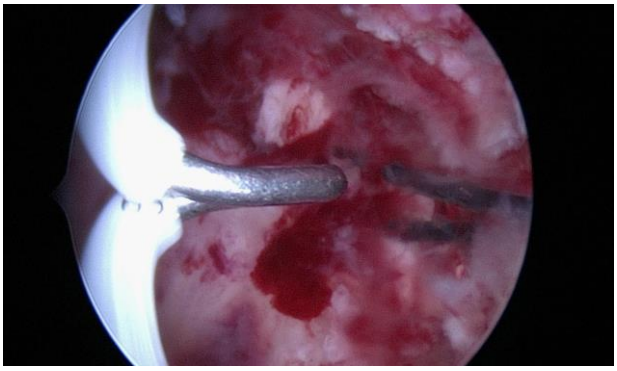

Figure 8: Second-look hysteroscopy showing the suture.

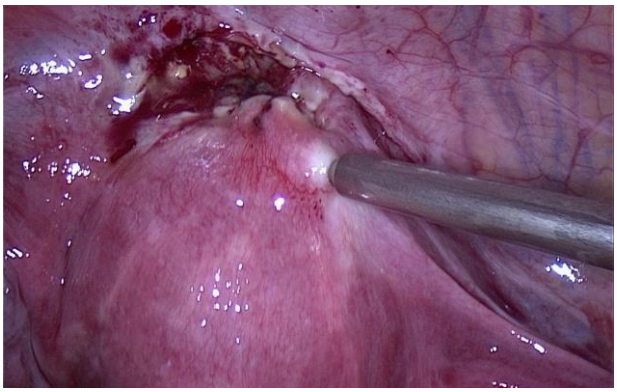

Figure 9: Second-look laparoscopy with injection of MTX around the suture.

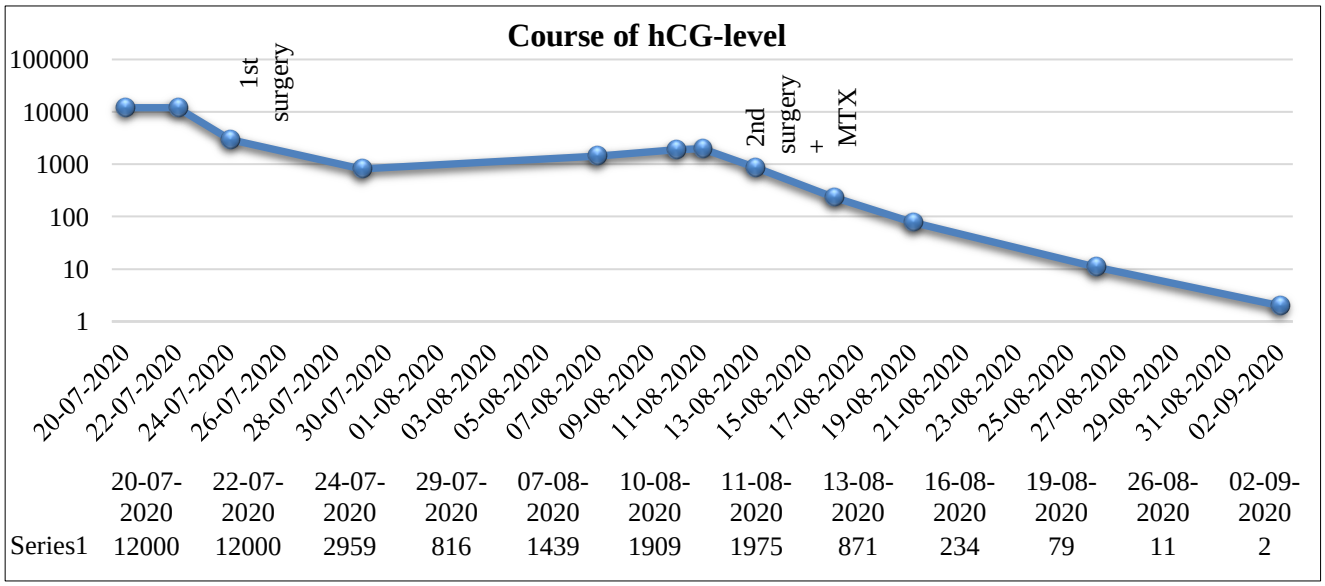

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Figure 10: Course of hCG-level over time with indication of the interventions.

DISCUSSION

CSEP is an inhomogeneous and potentially life-threatening disease. Therefore, the decision on the appropriate therapy must be made promptly, but in consideration of the given circumstances and the needs of the patient. Two reviews show that drug therapy only, whether local or systemic, causes a re-intervention rate of 25-38%, and systemic methotrexate only in particular can lead to serious complications such as hemorrhage and hysterectomy in 12% of cases.[21,22] Especially in cases with high hCG-level and large findings, drug therapy only seems to be less effective and rather dangerous.[22] In contrast, Petersen et al report a 97% success rate for primary laparoscopic therapy without serious complications. Hysteroscopy only has a success rate of only 83% and leads to serious complications in 3%. The combination of hysteroscopy and laparoscopy as chosen by us provides a safe assessment of the localization of the pregnancy (type I or II) and the option to surgical remove the findings from the uterine cavity or the abdomen. In our case the findings were wall-penetrating (Figures 2 and 3) whereas sonographically estimated rather as type I (Figure 1). Despite macroscopic complete resection during the first surgery (Figures 4 and 5) intrauterine residuals were found during the second surgery.

In this case, the initial combination with local methotrexate administration might have destroyed microscopic residuals and prevented a second intervention. Since an isthmocele can lead to secondary infertility and pregnancy complications such as recurrent ESCP, disturbance of placentation, and uterine rupture in addition to pain and bleeding disorders, repair of the isthmocele during laparoscopy in the infertile patient is reasonable.[23] Simultaneous isthmocele repair is also described in numerous case reports with different surgical techniques, but there is no follow-up of patients in terms of fulfillment of the childbearing desire.[24-27]

However, results of publications that report isthmocele repair outside of CSEP suggest, that the repair can successfully treat secondary infertility and reduce pregnancy complications. In a paper by Donnez et al, 44% of treated women with the desire to have a child became pregnant after isthmocele repair and had full term deliveries of healthy children via caesarean section.

Our isthmocele repair showed ideal adaptation both via ultrasound and in the second intervention three weeks after primary surgery. We recommend, that a pregnancy should not be attempted until six-months post-surgery for optimal wound healing results.

CONCLUSION

Combined hysteroscopic and laparoscopic surgery, especially for type II CSEP, seems to be a safe and effective treatment as shown in several publications. We suggest, that additional local injection of MTX in the caesarean scar during laparoscopy should be considered to reduce the risk for residuals even further, while, in the reported case-causing no additional side effects.

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REFERENCES

1. Parker VL, Srinivas M. Non-tubal ectopic pregnancy. Arch Gynecol Obstet. 2016;294(1):19-27.
2. Larsen JV, Solomon MH. Pregnancy in a uterine scar sacculus--an unusual cause of postabortal haemorrhage. A case report. S Afr Med J. 1978;53(4):142-3.
3. Calì G, Timor-Tritsch IE, Palacios-Jaraquemada J, Monteaugudo A, Buca D, Forlani F, et al. Outcome of Cesarean scar pregnancy managed expectantly: systematic review and meta-analysis. Ultrasound Obstet Gynecol. 2018;51(2):169-75.
4. Glenn TL, Bembry J, Findley AD, Yaklic JL, Bhagavath B, Gagneux P, et al. Cesarean Scar Ectopic Pregnancy: Current Management Strategies. Obstet Gynecol Surv. 2018;73(5):293-302.
5. Liu L, Ross WT, Chu AL, Deimling TA. An updated guide to the diagnosis and management of cesarean scar pregnancies. Curr Opin Obstet Gynecol. 2020;32(4):255-62.
6. Fabrega-Foster K, Hamper U. Cesarean Delivery Scar Pregnancy: A Case Report and Brief Literature Review. Ultrasound Q. 2015;31(3):195-7.
7. Bodur S, Özdamar Ö, Kılıç S, Gün I. The efficacy of the systemic methotrexate treatment in Cesarean scar ectopic pregnancy: A quantitative review of English literature. J Obstet Gynaecol. 2015;35(3):290-6.
8. Kelekçi S, Aydoğmuş S, Aydoğmuş H, Eriş S, Demirel E, Şen Selim H. Ineffectual Medical Treatment of Cesarean Scar Ectopic Pregnancy With Systemic Methotrexate: A Report of Two Cases. J Investig Med High Impact Case Rep. 2014;2(1):2324709614528903.
9. Timor-Tritsch IE, Monteagudo A, Santos R, Tsymbal T, Pineda G, Arslan AA. The diagnosis, treatment, and follow-up of cesarean scar pregnancy. Am J Obstet Gynecol. 2012;207(1).
10. Kalampokas E, Boutas I, Panoulis K, Kalampokas T. Novel Medical Therapy of Cesarean Scar Pregnancy With a Viable Embryo Combining Multidose Methotrexate and Mifepristone: A Case Report. Medicine (Baltimore). 2015;94(41):e1697.
11. Xiao J, Zhang S, Wang F, Wang Y, Shi Z, Zhou X, et al. Cesarean scar pregnancy: noninvasive and effective treatment with high-intensity focused ultrasound. Am J Obstet Gynecol. 2014;211(4):356.
12. Le Gall J, Fichez A, Lamblin G, Philip CA, Huissoud C. Cesarean scar ectopic pregnancies: combined modality therapies with uterine artery embolization before surgical procedure. Gynecol Obstet Fertil. 2015;43(3):191-9.

13. Arslan M, Pata O, Dilek TU, Aktas A, Aban M, Dilek S. Treatment of viable cesarean scar ectopic pregnancy with suction curettage. Int J Gynaecol Obstet. 2005;89(2):163-6.
14. Jurkovic D, Knez J, Appiah A, Farahani L, Mavrelos D, Ross JA. Surgical treatment of Cesarean scar ectopic pregnancy: efficacy and safety of ultrasound-guided suction curettage. Ultrasound Obstet Gynecol. 2016;47(4):511-7.
15. Pan Y, Liu MB. The value of hysteroscopic management of cesarean scar pregnancy: a report of 44 cases. Taiwan J Obstet Gynecol. 2017;56(2):139-42.
16. Timor-Tritsch IE, Monteagudo A, Bennett TA, Foley C, Ramos J, Kaelin Agten A. A new minimally invasive treatment for cesarean scar pregnancy and cervical pregnancy [published correction appears in Am J Obstet Gynecol. 2020;222(6):618.
17. Chen ZY, Zhao L, Nusilati A, Zhao D, Zhou M, Zhang XM. Clinical comparative analysis of surgical resection of the pregnancy by hysterotomy and hysterectomy for cesarean scar pregnancy. Zhonghua Fu Chan Ke Za Zhi. 2017;52(2):98-102.
18. Vial Y, Petignat P, Hohlfeld P. Pregnancy in a cesarean scar. Ultrasound Obstet Gynecol. 2000;16(6):592-3.
19. Tahaoglu AE, Dogan Y, Bakir MS, Baglı I, Peker N, Cavus Y, et al. A single centre's experience of Cesarean scar pregnancy and proposal of a management algorithm. J Obstet Gynaecol. 2019;39(2):259-64.
20. Birch Petersen K, Hoffmann E, Rifbjerg Larsen C, Svarre Nielsen H. Cesarean scar pregnancy: a systematic review of treatment studies. Fertil Steril. 2016;105(4):958-67.
21. Maheux-Lacroix S, Li F, Bujold E, Nesbitt-Hawes E, Deans R, Abbott J. Cesarean Scar Pregnancies: A Systematic Review of Treatment Options. J Minim Invasive Gynecol. 2017;24(6):915-25.
22. Dior UP, Palma-Dias R, Reidy KL, Cheng C, Healey M. Cesarean Scar Pregnancies: Incidence and Factors Associated with Conversion to Surgery from Medical Management. J Minim Invasive Gynecol. 2019;26(5):919-27.
23. Futyma K, Gałczyński K, Romanek K, Filipczak A, Rechberger T. When and how should we treat cesarean scar defect - isthmocoele? Ginekol Pol. 2016;87(9):664-8.
24. Mahgoub S, Gabriele V, Faller E, Langer B, Wattiez A, Lecointre L, et al. Cesarean Scar Ectopic Pregnancy: Laparoscopic Resection and Total Scar Dehiscence Repair. J Minim Invasive Gynecol. 2018;25(2):297-8.
25. Kiyak H, Wetherilt LS, Seckin KD, Polat I, Kadirogullari P, Karacan T. Laparoscopic Excision of a Scar Pregnancy and Isthmocele Repair. J Minim Invasive Gynecol. 2018;25(4):582.
26. Nyangoh Timoh K, Enderle I, Leveque J, Lavoue V. Robotic-assisted laparoscopy using hysteroscopy treatment of a residual cesarean scar pregnancy and isthmocele. Gynecol Obstet Fertil Senol. 2020;48(5):460-1.
27. Yoon R, Sasaki K, Miller CE. Laparoscopic Excision of Cesarean Scar Pregnancy with Scar Revision. J Minim Invasive Gynecol. 2020;S1553-4.
28. Donnez O, Donnez J, Orellana R, Dolmans MM. Gynecological and obstetrical outcomes after laparoscopic repair of a cesarean scar defect in a series of 38 women. Fertil Steril. 2017;107(1):289-96.

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