University of Novi Sad, Faculty of Medicine Novi Sad[1]
Clinical Center of Vojvodina, Novi Sad,
Clinic of Gynecology and Obstetrics[2]

Case report
Prikaz slučaja
UDK 618.5-089.888.61-06:618.14-007.253
https://doi.org/10.2298/MPNS2212374K

UTEROCUTANEOUS FISTULA AFTER CESAREAN SECTION − A RARE DIAGNOSIS NOT TO BE MISSED – A CASE REPORT

UTEROKUTANA FISTULA NAKON CARSKOG REZA, RARITET KOJI SE NE SME ZABORAVITI – PRIKAZ SLUČAJA

Anita KRSMAN[1, 2], Branislava BATURAN[1, 2], Sanja BULATOVIĆ[1], Đorđe PETROVIĆ[1, 2], Ksenija ANTIĆ TRIFUNOVIĆ[2] and Dragan STAJIĆ[1, 2]

Summary

Introduction. Uterocutaneous fistula is a rare complication of cesarean section which is challenging to diagnose and treat. The aim of this paper is to present a case of uterocutaneous fistula in order to contribute to the literature and help in the therapy and diagnosis of this rare complication. **Case Report.** A 29-year-old patient was referred to our clinic two months after her second cesarean section. The immediate postpartum course was complicated by endometritis treated with antibiotic therapy. At the time of admission, she was afebrile, without complaints other than a 2 cm long wound dehiscence on the anterior abdominal wall. The uterocutaneous fistula was confirmed by injecting methylene blue through the dehiscence on the anterior abdominal wall, which then spread into the vagina through the cervix. After laboratory tests, ultrasound and clinical examination, the patient underwent surgery. A total excision of the fistula was performed by laparotomy. Histopathological findings confirmed the diagnosis of uterocutaneous fistula. The postoperative recovery was uneventful. At the follow-up examination, three months after surgery, the patient had no complaints; the menstrual cycles were normal, as well as the transvaginal ultrasound findings. **Conclusion.** Uterocutaneous fistula is a rare complication following cesarean section. Timely identification of the fistula, its complete resection, and adequate antibiotic therapy in case of infection are necessary.
Key words: Cutaneous Fistula; Uterine Diseases; Cesarean Section; Postoperative Complications; Treatment Outcome

Sažetak

Uvod. Uterokutana fistula je retka, ali terapijski i dijagnostički vrlo zahtevna komplikacija nakon carskog reza. Cilj rada bio je prikaz slučaja uterokutane fistule i dati doprinos literaturi i pomoći u terapiji i dijagnostikovanju ove retke komplikacije. **Prikaz slučaja.** Pacijentkinja stara 29 godina javlja se na Kliniku dva meseca nakon drugog carskog reza. Neposredni postpartalni tok bio je komplikovan endometritisom, koji je tretiran antibiotskom terapijom. U momentu prijema pacijentkinja je afebrilna, bez tegoba, osim dehiscencije u predelu incizije na prednjem trbušnom zidu u dužini od 2 cm. Uterokutana fistula je potvrđena ubrizgavanjem metilenskog plavila, kroz dehiscenciju prednjeg trbušnog zida, koje se potom kroz grlić izlilo u vaginu. Nakon laboratorijske, ultrazvučne i kliničke obrade, pacijentkinja je operisana. Tokom laparotomije, načinjeno je odstranjenje fistule u celosti. Patohistološki nalaz potvrdio je dijagnozu uterokutane fistule. Postoperativni tok protekao je uredno. Na kontrolnom pregledu, tri meseca nakon operacije, pacijentkinja je bez tegoba, urednih menstrualnih ciklusa i urednog ultrazvučnog nalaza. **Zaključak.** Uterokutana fistula je retka komplikacija nakon carskog reza. Neophodna je pravovremena identifikacija fistule, kompletna resekcija fistule kao i adekvatna antibiotska terapija u sličaju postojanja infekcije.
Ključne reči: kožna fistula; bolesti uterusa; carski rez; postoperativne komplikacije; ishod lečenja

Introduction

A fistula is an abnormal communication between two epithelial surfaces that is most commonly found between the urinary and genital tract [1]. Pathological communication between the skin and the uterus (uterocutaneous fistula) is extremely rare, but it may be a complication in gynecology and obstetrics [2]. Approximately 120 cases have been reported over the past 200 years, 25 cases in the past 50 years, and 15 cases in the past 20 years [3]. We have not found any reported cases in the Serbian medical literature. This case report is bridging the gap and serves as a reminder that, although rare, uterocutaneous fistula is a possible complication after cesarean section that requires timely diagnosis and treatment.

Case Report

A 29-year-old patient was referred to the Department of Gynecology and Obstetrics two months after her second cesarean section. She was admitted to the Clinical Center due to partial dehiscence of the abdominal wound. An elective cesarean section was performed at 39 weeks of gestation because the

Corresponding Author: Dr Anita Krsman, Medicinski fakultet Novi Sad, KCV - Klinika za ginekologiju i akušerstvo, 21000 Novi Sad, Branimira Ćosića 37, E-mail: anita.krsman@mf.uns.ac.rs

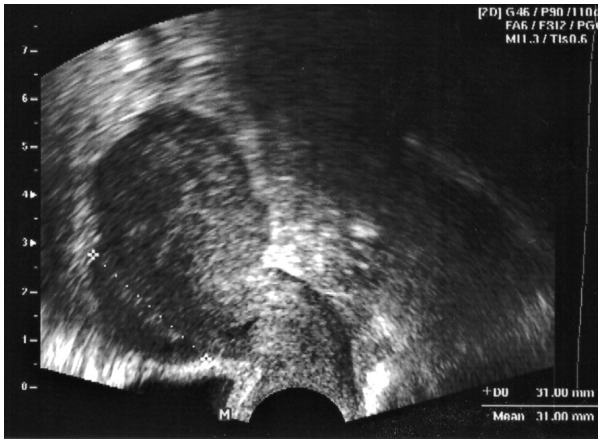

Figure 1. Transvaginal ultrasound before surgery shows an echogenic formation in the area of the uterine incision
Slika 1. *Transvaginalni ultrazvučni pregled pre operativnog zahvata pokazuje ehogenu promenu u predelu reza na uterusu*

previous pregnancy ended with a cesarean section as well.

The postoperative course was complicated by the development of endometritis. During the postpartum period, she received a parenteral combination antibiotic therapy (cefuroxime and metronidazole) which led to an improvement, so the patient was discharged on the thirteenth day after the cesarean section. Staphylococcus aureus was isolated from the wound swab, and oral antibiotic therapy was administered according to the antibiogram (ciprofloxacin).

At the control examination, after the toilet of the wound on the anterior abdominal wall with an antiseptic solution, the patient noticed a discharge of the solution through the vagina. The examination of the anterior abdominal wall revealed a Cohen incision with partial 2 cm long wound dehiscence in the central part with initial granulations. On speculum examination, the cervix and vagina were normal. Since uterocutaneous fistula was suspected, methylene blue solution was injected through the area of dehiscence of the incision on the abdominal wall, and it was seen coming through the cervix into the vagina. The ultrasound examination showed an echogenic formation, 31 x 24 mm in size, in the area of the uterine incision, which was connected with the skin **(Figure 1)**.

On admission, the patient was afebrile, with normal vital parameters, peristalsis and no difficulty urinating. Laboratory test results were normal with negative markers of infection (white blood cell count: 5.8 x 10^9/l, C-reactive protein: 3.9 mg/L). After clinical, ultrasound and anesthesia examination, the patient underwent surgery. Methylene blue solution was injected immediately before the surgery through the scar to determine the direction of the fistula. After opening the anterior abdominal wall by Pfannenstiel laparotomy, uterine adherence to the anterior abdominal wall was noticed. Within

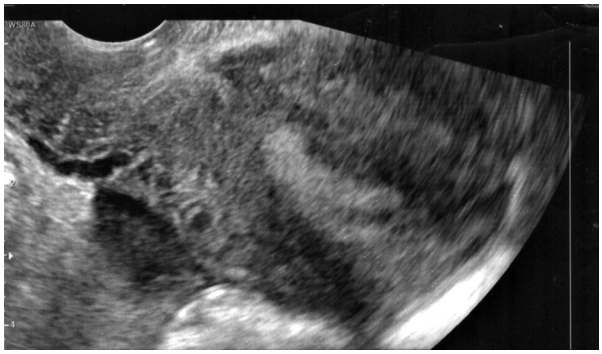

Figure 2. Transvaginal ultrasound three months after surgery shows a normal finding of the uterus without pathological changes
Slika 2. *Transvaginalni ultrazvučni pregled tri meseca nakon operacije pokazuje uredan nalaz, bez patoloških promena*

the connection, there was a 5 mm wide fistula. The entire fistula channel connecting the skin to the uterus with the surrounding tissue was excised, and the sample was sent for histopathological analysis. Uterine opening and debridement were performed, so individual sutures were placed on the layers of the uterus. Adhesiolysis of the omentum from the anterior abdominal wall adhesion was performed. After the drains were placed in the pouch of Douglas and anterior uterine space, 48 h after the surgery the drains were removed.

Postoperative antibiotics and analgesia were administered. Pathological examination showed inflammatory necrosis of the uterine muscle with a fistula tract in the muscular wall of the uterus with hemorrhage and fibrin deposition. The patient was discharged on the seventh postoperative day. She had a menstruation 4 weeks after surgery, and she had no complaints. There was no sinus on the abdomen discharging blood. The follow-up ultrasound examination 3 months after surgery was normal **(Figure 2).** She was advised to use contraception.

Discussion

Our patient presented with a fistula following cesarean section, but there are other recognized risk factors for uterocutaneous fistula. The most common are septic abortion, postoperative sepsis, pelvic abscesses, intrauterine device-related actinomycosis, abdominal drainage, operative delivery, endometriosis and true intra-abdominal pregnancy because of incomplete placenta removal, congenital anomalies [4–10]. Studies have shown that the onset of symptoms varies from 2 months to even 6 years after the last surgery [6]. Our patient was referred to the hospital barely 2 months after the cesarean section. The dominant symptom of the uterocutaneous fistula is a retrograde effusion of menstrual blood simultaneously with normal menstruation [9]. In our patient, this symptom was not found, probably due to lactation amenorrhea. The diagnosis of

uterocutaneous fistula is made by fistulography [11] and magnetic resonance imaging [6]. In case of a small opening in the skin and suspicion of uterocutaneous fistula, hysterosalpingography with methylene blue injection through the cervix may be helpful [12, 13]. There is no standard treatment for uterocutaneous fistula, because it is a rare clinical condition (less than 15 cases reported in the last 20 years worldwide) [14]. Some authors used a minimally invasive surgery (laparoscopy) to excise the fistula tract [15]. Some papers report a novel successful treatment of uterocutaneous fistula with gonadotropin-releasing hormone agonists [16, 17]. In our case, a fistulectomy was performed to preserve the patient's fertility.

Conclusion

Certain studies have shown that congenital anomalies and earlier curettage of the uterus can be the reasons for both uterine rupture and uterocutaneous fistula. However, cesarean section remains the most significant risk factor for the occurrence of this rare complication. An uterocutaneous fistula should be suspected in any patient who presents with cyclic bloody discharge from a surgical scar. Given the increased incidence of cesarean section, obstetricians must be aware of the potential complications of surgical delivery. Uterocutaneous fistula is a rare complication of the cesarean section that should be considered and recognized on time so that the patient can receive adequate treatment.

References

1. Eleje GU, Udigwe GO, Okeke MP, Nwokoro JM, Onyejiaku LC, Ezugwu CJ, et al. Post cesarean uterocutaneous fistula with successful repair and successful outcome: a case report. J Pregnancy Neonatal Med. 2018;2(2):27-30.

2. Olalere FDH, Kuye TO, Ottun TA, Adewunmi AA, Oshodi YA, Akinlusi FM, et al. Endometriotic uterocutaneous fistula after cesarean section- successful diagnosis with fistulogram and complete tract resection and medical treatment: a case report. World Journal of Innovative Research. 2020;8(4):4-6.

3. Vellanki VS, Goginemi S, Jahnavi Kanakamedala S. Case report of utero-cutaneous fistula. J Womens Health Care. 2015;4(2):231.

4. Osman SA, Al-Badr AH, Malabarey OT, Dawood AM, AlMosaieed BN, Rizk DEE. Causes and management of urogenital fistulas. A retrospective cohort study from a tertiary referral center in Saudi Arabia. Saudi Med J. 2018;39(4):373-8.

5. Aggarwal R, Indiran V, Maduraimuthu P. Different etiologies of an unusual disease: colouterine fistula - report of two cases. Indian J Radiol Imaging. 2018;28(1):37-40.

6. Anderson KB, Søgaard-Andersen E, Aleksyniene R, Frandsen AP. Spontaneous utero-cutaneous fistula between a benign uterine leiomyoma and abdominal skin: a case report. Case Rep Womens Health. 2020;29:e00282.

7. Nwogu C, Ugwu A, Soibi-Harry A, Nwokocha S. Uterocutaneous fistula postabdominal myomectomy: successful repair – case report and review of literature. Nigerian Journal of Experimental and Clinical Biosciences. 2021;9(3):199.

8. Mahto S, Ghimire R, Kunwar S, Saha R. Successful outcome of uterocutaneous fistula: a case report. JNMA J Nepal Med Assoc. 2021;59(241):913-5.

9. Offiong RA, Adewole ND, Zakari MM, Okochi DO. Uterocutaneous fistula: a rare clinical entity. New Nigerian Journal of Clinical Research. 2018;7(11):35-7.

10. Antić-Trifunović K, Krsman A, Šuvaković Z, Stajić G, Ilić Đ, Dickov I. Rupture of the unscarred uterus during induced termination of pregnancy in the second trimester: a case report. Med Pregl. 2021;74(9-10):324-6.

11. Min KJ, Lee J, Lee S, Lee S, Hong JH, Song JY, et al. Uterocutaneous fistula after pelviscopic myomectomy - successful diagnosis with hystero-salpingo contrast sonography and complete tract resection and medical treatment for fertility preservation in young woman: a case report. Obstet Gynecol Sci. 2018;61(5):41-4.

12. Sonmezer M, Sahincioglu O, Cetinkaya E, Yazici F. Uterocutaneous fistula after surgical treatment of an incomplete abortion: methylene blue test to verify the diagnosis. Arch Gynecol Obstet. 2009;279(2):225-7.

13. Shah N, Changede P, More V. Laparoscopic management of post-cesarean section uterocutaneous fistula. J Obstet Gynaecol India. 2019;69(4):380-2.

14. Ruiz Arteaga JD, Valdez Murillo AN, Hernandez Trejo MC. Utero-cutaneous fistula: a case report and literature review. Ginecol Obstet Mex. 2012;80(2):95-8.

15. Loue V, Koffi A, Adjoby R, N'Guessan K, Alla C, Gbary E, et al. Postmyomectomy uterocutaneous fistula. J Gynecol Surg. 2013;29(1):36-8.

16. Seyhan A, Ata B, Sidal B, Urman B. Medical treatment of uterocutaneous fistula with gonadotropin-releasing hormone agonist administration. Obstet Gynecol. 2008;111(2 Pt 2):526-8.

18. Lawal IK, Suleiman AK, Ketare N, Obiokonkwo CA. Scar endometriosis as a complication of surgically treated uterocutaneous fistula. Trop J Obstet Gynaecol. 2020;37(1):213-5.

Rad je primljen 20. VIII 2022.
Recenziran 6. IV 2023.
Prihvaćen za štampu 8. IV 2023.
BIBLID.0025-8105:(2022):LXXV:11-12:374-376.