

Endometriotic Uterocutaneous Fistula after Caesarean Section- Successful Diagnosis with Fistulogram and Complete Tract Resection and Medical Treatment: A Case Report

*F. D. H. Olalere, T. O. Kuye, T. A. Ottun, A. A. Adewunmi, Y.A. Oshodi, F. M. Akinlusi, J. O. Agbara, A. F. Tijani

Abstract— Extrapelvic endometriosis is a rare condition and most cases occur after gynecological or obstetrics procedure involving the uterus. Uterocutaneous fistula is a rarely reported clinical condition after uterine procedures. In this case report, endometriotic uterocutaneous fistula with lower uterine synechia following complicated caesarean section was diagnosed simply with fistulogram in a 21-year-old Nigerian woman. She was managed by fistula tract excision, uterine wall repair, adhesiolysis with rail-roading, combined with medical treatment using gonadotropin-releasing hormone agonist to prevent recurrent fistula and preserve fertility.

Index Terms— Caesarean section, Endometriotic uterocutaneous fistula, fistulogram, Uterine synechia.

I. INTRODUCTION

Endometriosis is defined as the presence of functional endometrial tissue outside the uterine cavity [1]. It is found on the ovary, posterior broad ligament, anterior and posterior cul-de-sacs, and uterosacral ligament; it is found less commonly on the fallopian tubes, bladder, colon, small bowel, and ureters. It has however been in the lungs, pericardium, caesarean scar and fistula [2-5]. An endometriotic uterocutaneous fistula (abnormal communication between uterus and skin) and extra pelvis endometriosis is a rare condition complicating uterine surgery [5, 6], and there are only a few reports of this, in the existing literature. The etiologies of uterocutaneous fistula are mostly iatrogenic, due to post-operative injuries, infection and inflammation [5]. Previous reports following caesarean section are associated with red degeneration of intramural fibroids, B-lynch sutures, multiple surgeries, insertion of drains or multi-associations [7-12]. It has also been reported following criminal abortion [6]. Because of the rarity of uterocutaneous fistula, there is no available algorithm for diagnosis and management [13]. Endometriotic infiltration of the abdominal wall scar following various obstetrical and gynecological procedures have been reported [2, 3], though rare and diagnosis is frequently made after excision and histopathology of the

lesion [3]. However, endometriosis of an uterocutaneous fistula tract scar is extremely rare. We report a case of endometriotic post caesarean uterocutaneous fistula with uterine synechia that was successfully treated with excision of the fistulous tract from the skin, repair of the fistulous uterus, adhesiolysis and adjunct medical therapy with GnRH analogue.

II. CASE REPORT

A 21-year-old Nigerian (black African) married woman, para 1 presented on the 1st of March 2019, which was two years after caesarean section in a private hospital with four months history of painful bloody discharge from a previous Pfannenstiel scar. She had emergency lower segment caesarean section 2 years prior to presentation due to prolonged obstructed labor and intrapartum eclampsia with the delivery of a live female neonate with severe birth asphyxia with birth weight of 3.4 kg. The child was transferred to a tertiary hospital, where she was managed for 4 weeks before discharged. One week after, the caesarean section was complicated with abdominal wound breakdown and she was managed conservatively for 5 weeks on admission and was discharged home for daily wound dressing on outpatient basis for another 6 months. Two years afterward, she noticed a swelling on the right side of the healed Pfannenstiel scar, which subsequently breakdown and started discharging blood. This marked the onset of her first menstruation after her last delivery, bleeding was copious, cyclical associated severe dysmenorrhea and spotting per vaginam. She presented at the private hospital where the first surgery was done after onset of symptoms, where she was given medical treatment with antibiotics and analgesics. However, persistence of her symptoms prompted a second consultation at a general hospital, where she was sent for the investigation and referral to our hospital on the 4th February 2019. In the past, she had one complicated caesarean delivery as already described otherwise there was no history of other prior abdomino-pelvic surgery. She attained menarche at 15 years and menstruates for 3-5 days in a regular 30-day cycle. She had no prior history of primary dysmenorrhea, dyspareunia, dysuria, or abnormal vaginal discharge. She was human immunodeficiency virus negative, had a single partner and had no history of sexually transmittable infections. She had no prior history or diagnosis of endometriosis. On physical examination, she was afebrile, not pale, had stable vital signs and a body mass index of 30.8 kg/m²s (weight 63 kg, height 1.43m). A physical examination of her abdomen

*Dr. Mrs. F. D. H. Olalere

Dr. Mrs. T. O. Kuye, Dr. Mrs. T. A. Ottun, Dr. A. A. Adewunmi, Dr. J. O. AGBARA, Dr. Y. A. Oshodi, Dr. F. M. Akinlusi, Dr. A. F. Tijani
Obstetrics and Gynecology Department, Lagos State University College of Medicine, (LASUCOM), Ikeja, Lagos, Nigeria
Lagos State University Teaching Hospital, Ikeja, (LASUTH) Lagos, Nigeria.

(Fig.1) showed a localized tenderness at the abdominal scar, healed pfannenstiel scar with fistulous skin puckering at about 4cm to the right lateral edge, oozing blood and the speculum vaginal examination was grossly normal. Full blood count (PCV was 46%, WBC was $7.4 \times 10^9/L$, platelets 239,000), urinalysis, hepatitis B surface antigen and Hepatitis C antibodies were normal. Ultrasound was suggestive of uterocutaneous fistula. Fistulogram done on the 25th March 2019. It's revealed a connection between the uterus and the skin and also highlighted the endometrial cavity. She was counselled on the findings and options of management and placed on combined oral contraceptive pills. She however, declined surgery and defaulted for 5months. After which she presented on the 19th of July 2019 and consented for surgery. She was co-managed with the general surgeon for the risk of bowel involvement. She had an exploratory laparotomy with fistula tract excision, lower uterine adhesiolysis and rail roading done on the 30th October 2019 with intra-operative findings of a dense, thickened fistulous adhesion band from the skin, rectus sheath, peritoneum and the anterior wall of uterus. Adhesion to the greater omentum with loops of small bowel, both fallopian tubes were buried into the fistulous band and the distal third of lower uterine wall was adhered to the bladder, anterior abdominal wall and normal ovaries (Figure 3). Incidental findings of the uterine synechia distal to the fistulous tract, for which blunt and sharp adhesiolysis and rail roading was done, silicon foley catheter inflated with 3ml of fluid was left in-situ as a stent to create a passage-way for the menstrum and avert re-accumulation and repeat synechiae and uterocutaneous fistula. The estimated blood loss was 500 millilitres. The immediate postoperative conditions were satisfactory. She was placed on combined oral contraceptive pills once daily for three months and had intrauterine catheter removed after ten days at the outpatient clinic. Histology of the specimen removed revealed an unremarkable epidermis with densed collagenous dermis and numerous endometrial glands with adjacent endometrial stroma tissues which lacks atypia, features in keeping with endometriosis. She was commenced on GHRH analogue, SC Zoladex 3.65mg for 6 months. At follow-up, the patient was satisfied with the repair and healing because she has had menstruation twice without the menstrual discharge at the well healed abdominal scar.

III. DISCUSSION

Uterocutaneous fistula is an abnormal communication between the epithelial lining of the skin and the uterus. Uterocutaneous fistula is a rare condition, which had been documented following infection, inflammation, postpartum sepsis, invasive endometriosis and uterine procedures⁵. Endometriotic fistula in a patient with no previous endometriosis prior uterine procedures is extremely rare in literature, the first reported case was reported in Greece⁵ about 17years ago in a woman with four previous caesarean section and two first trimester termination of pregnancy and use of drain in the last caesarean section. She was managed as case of scar endometriosis which is found in 0.4% of caesarean section, however the intraoperative findings of endometriotic uterocutaneous fistula was found⁵. Endometriosis is the presence of functional endothelial glandular and stroma tissue outside uterine cavity^{1,8}. There

are different sites of endometriosis, each sites with different theories explaining the probable cause, Rare sites of endometriosis include the brain, lung, umbilicus, caesarean scar, uterocutaneous fistula^{2-5,16}. It's a disease of many theories, retrograde flow of menstrum in patient with an outflow tract obstruction account for the pelvic spread¹, however in this case presented, there was lower uterine synechia, and possibly tubal blockage, prior history of prolonged obstructed labour which may be a nidus for sepsis, she had post- operative sepsis with protracted wound healing, all of these might have compromised the integrity of the uterine scar and predispose her to uterine wound scar dehiscence. She might have developed a sinus present in the caesarean scar site, following wound sepsis. Since there were dense adhesions between the uterus and abdominal wall, the sinus on the wound in the abdominal wall got communicated with the uterine wound dehiscence site in the uterus leading to the formation of endometriotic uterocutaneous fistula. Paucity of data on this condition had negative impact on the availability of standard operating practice in diagnosis and treatment, different physician deploy different options of management that's available and affordable for the betterment of patient's care, Several diagnostic methods have been applied, trans abdominal ultrasound is the baseline used to detect any structural uterine abnormality, while the fistulogram that provides the highlight of the fistula is the first choice¹⁷, hystero-salpingo sonography with contrast material via cervical opening, hysterosalpingography (HSG), contrast enhanced Computed tomography, MRI and hysteroscopy used to visualize the uterine neck of the fistula had been reported¹⁸. Most uterocutaneous fistulas originate from some type of infective process that disrupts the continuity of tissues involved. Once a fistula is diagnosed the basic principle in treatment is obliteration of opening of fistulous tract¹⁸. Uterocutaneous fistulas can be managed medically, but permanent cure remained surgical. Hysterectomy was initially employed to treat uterocutaneous fistula, however conservative surgical fistula tract excision alone or Gonadotrophin releasing hormone analogue alone¹⁹ or with surgery and successful treatment outcome had been reported.

Conservative surgical treatment associated with medical therapy can be an efficient procedure in women who desire subsequent pregnancy. This case report highlights the rare possibility of the endometriotic uterocutaneous fistula occurring in a woman following caesarean section complicated with post-operative sepsis. Making obstetric care available, affordable and presence of skilled birth attendance with appropriate surgical skills and good post-operative care in a parturient at risk are necessary to avert the on towards outcome that is distressing to the patient. Conflict of interest: No potential conflict of interest relevant to this article was reported.

IV. FIGURES

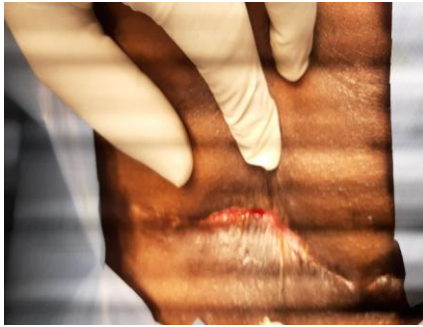


Fig. 1: image of the anterior abdominal wall of the fistula prior surgical excision. Fistulous tract covered with blood.



Fig. 2 : Fistulogram showing a (scout film above), b (left lower), & c (right lower);, the left hydrosalpinx and right tube not delineated ,filling defects noted around the uterine fundus and the left upper fourth of the uterus and lower uterine segment.

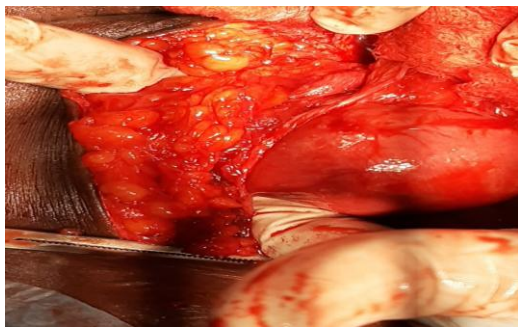


Fig. 3: Intraoperative findings with gloved fingers inserted into the two ends of the fistulous tract after dissection

V. ACKNOWLEDGMENT

We appreciate the assistance rendered by all doctors in Reproductive, endocrine and fertility units of the department of Obstetrics and Gynecology, Lagos State University Teaching Hospital, LASUTH. Professor Hajara Umar and Professor Adedosu for going through the drafted manuscript. Dr. Fatimah Rabi, Dr. T. Odetayo and Dr. Ogunyemi, for active participation in the management of the patient.

REFERENCES

[1] V.L. Holt, B. Trabert, K. Upson. "Endometriosis" in Women's health (second edition),2013, pp. 271-84. Available online 2012, December,7. <https://doi.org/10.1016/B978-0-12-384978-6.00018-2>

[2] Kafkasli, R. R. Franklin RR, D. Sauls. "Endometriosis in the uterine wall cesarean section scar." *Gynecol Obstet Invest.*, vol. 42, (1996), pp. 211-3.

[3] Goel P, Sood SS, Dalal A, Romilla. Cesarean scar endometriosis: Report of two cases. *Indian J Med Sci* 2005; 59:495-8.

[4] [A.K. Lahiri, K. Sharma, N. Busiri](#), "Endometriosis of the uterine cesarean section scar: a case report", *Indian Journal of Radiology and Imaging*,vol.18(1), Feb.,2008pp.66-8.DOI: [10.4103/0971-3026.37111](https://doi.org/10.4103/0971-3026.37111)

[5] K. Dragoumis, T. Mikos, M. Zafrakas, E. Assimakopoulos, P. Stamatopoulos, J. Bontis."Endometriotic uterocutaneous fistula after cesarean section: A case report,". *Gynecol Obstet Invest.*, vol.57(2), 2004, pp. 90-2. Epub 2003 Dec 9. PubMed PMID:14671417.

[6] S.K. Gupta, V.K. Shukla, D.N. Varma and S.K. Roy. "Uterocutaneous fistula" *Postgrad Med J.*, vol. 69. (1993) pp. 822- 3

[7] Shukla D, Pandey S, Pandey LK, Shukla VK. "Repair of uterocutaneous fistula". *Obstet Gynecol* 2006; 108:732-3.

[8] G. Eldem, B. Turkbey, S. Balas, E. Akpınar. MDCT diagnosis of uterocutaneous fistula. *Eur J Radiol.*, vol. 67, (2008) pp.129-30

[9] M. Thakur, S.S. Rathore, A. Jindal, K. Mahajan. "Uterocutaneous fistula following B-Lynch suture for primary postpartum haemorrhage." *BMJ Case Rep.* 2018 5; pii: bcr-2017-223518.

[10] Lim PS, Shafiee MN, Ahmad S, et al. Utero-cutaneous fistula after caesarean section secondary to red degeneration of intramural fibroid. *Sex Reprod Healthc.*, vol. 3, (2012), pp. 95-6.

[11] V.S. Vellanki, S. Gogineni, J.S. Kanakamedala. "Case report of uterocutaneous fistula". *J Women's Health Care.*, vol. 4. (2015), 231.

[12] G. Maddah, A. S. Fattahi, A. Rahnema S. T. Jamshidi. "Uterocutaneous Fistula Following Cesarean Section: Successful Management of a Case." *Iran J Med Sci.*, vol. 41. (2016); pp. 157-60.

[13] Thubert T, Denoiseux C, Faivre E, A. Naveau, C. Trichot, X. Deffieux. "Combined conservative surgical and medical treatment of a uterocutaneous fistula". *J Minim Invasive Gynecol.*, vol. 19 (2012) pp. 244-7

[14] L. Taff, S. Jones. "Cesarean scar endometriosis: a report of two cases". *J. Reprod. Med.*, vol. 47, (2002) pp. 450-2.

[15] P.V. Bagade, M. M. Guirguis. "Menstruating from the umbilicus as a rare case of primary endometriosis: a case report," *Journal of medical case reports.*, vol. 3, pp.9326. 10 Dec. 2009, doi:10.1186/1752-1947-3-9326.

[16] P. V. Bagade, M. M Guirguis. "Menstruating from the umbilicus as umbilicus as a rare case of primary umbilical endometriosis: a case report." *Journal of medical case reports* vol. 3, pp. 9326. 10 Dec. 2009, doi:10.1186/1752-1947-3-9326

[17] K. J. Min, S. Lee, S. Lee, J. H. Hong, J. Y. Song, J. K. Lee et.al Uterocutaneous fistula after pelvicoscopic 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.*, vol.61(5), (2018) Sep 641-64. Available: <https://doi.org/10.5468/ogs.2018.61.5.641>

[18] A. Jindal, H. Chaudhary, M. Thakar.Tubercular uterocutaneous fistula after caesarean section: a case report," *Women's health.*, vol. 17.2018, pp. 3-4. Available:<https://doi.org/10.1016/j.crwh.2018.02.001>

[19] Seyhan, B. Ata, B. Sidal, B. Urman. "Medical treatment of uterocutaneous fistula with gonadotropin-releasing hormone agonist administration." *Obstet Gynecol.*, vol. 111(2 Pt 2), pp. 526-8. doi: 10.1097/01.AOG.0000281670.94265.5c.