

Tubocutaneous Fistula due to Endometriosis – A Differential Diagnosis in Cutaneous Fistulas with Cyclic Secretion

Fístula tubocutânea secundária à endometriose – diagnóstico diferenciado em fístulas cutâneas com secreção cíclica

Edinari Nunes de Sousa Lopes¹ Lia Cruz Vaz da Costa Damásio¹ Laio Santana Passos¹

¹Department of Gynecology, Universidade Federal do Piauí - UFPI, Teresina, Brazil

Address for correspondence Edinari Nunes de Sousa Lopes, MD, Departamento de Ginecologia, Universidade Federal do Piauí - UFPI, Av. Universitária – Ininga, Teresina, PI, Brazil 64049-550 (e-mail: edinarinunes@hotmail.com).

Rev Bras Ginecol Obstet 2017;39:31–34.

Abstract

The development of a tubocutaneous fistula due to endometriosis in a post-cesarean section surgical scar is a rare complication that generates significant morbidity in the affected women. Surgery is the treatment of choice in these cases. Hormonal therapies may lead to an improvement in symptoms, but do not eradicate such lesions. In this report, we present a 34-year-old patient with a cutaneous fistula in the left iliac fossa with cyclic secretion. Anamnesis, a physical examination, and supplementary tests led us to suggest endometriosis as the main diagnosis, which was confirmed after surgical intervention.

Keywords

- ▶ gynecology
- ▶ endometriosis
- ▶ fistula

Resumo

O desenvolvimento de fístula tubocutânea secundária à endometriose em cicatriz cirúrgica após cesariana é uma complicação rara, que gera importante morbidade às mulheres acometidas. A cirurgia é o tratamento de escolha nesses casos. Terapias hormonais podem conduzir a uma melhora dos sintomas, mas, de forma alguma, levam à erradicação de tais lesões. No presente relato, temos uma paciente de 34 anos de idade que apresentava uma fístula cutânea em fossa ilíaca esquerda com secreção cíclica. Anamnese, exame físico e exames complementares nos levaram a aventar como principal hipótese diagnóstica a endometriose, que foi confirmada após intervenção cirúrgica.

Palavras-chave

- ▶ ginecologia
- ▶ endometriose
- ▶ fístula

Introduction

Endometriosis is a benign disease defined by the presence of endometrial glands and stroma outside the uterus.¹ The average age at diagnosis is between 25 and 35 years.^{2,3} Endometriosis outside the pelvis is rare, and most cases occur in surgical scars after procedures involving the female genital tract.⁴ In this report, we present a rare case of tubocutaneous fistula due to endometriosis that developed after a cesarean section. The

fistula stretched from the left uterine tube to the left inguinal region along the anatomical path of the round ligament.

Case Report

A 34-year-old woman sought the surgery service of the University Hospital of Teresina in August 2015 due to a history of discharge from a cutaneous opening in the left iliac fossa that had varied in color from citric yellow to red

received
June 11, 2016
accepted
November 17, 2016

DOI <http://dx.doi.org/10.1055/s-0036-1597754>.
ISSN 0100-7203.

Copyright © 2017 by Thieme-Revinter Publicações Ltda, Rio de Janeiro, Brazil

License terms



and had exhibited cyclical behavior over the course of six years. She presented with the following obstetrical history: one miscarriage late in 1999; two vaginal deliveries at term, one in 2002 and another in 2003; and one cesarean section in 2008, which interrupted a pregnancy of between 35 and 36 weeks, due to history of anemia and severe thrombocytopenia associated with maternal-fetal Rh incompatibility.

Regarding the medical history related to the cesarean birth, the patient reported that six months after the cesarean section, she resumed regular menstrual cycles, which were associated with pain and redness in the left iliac fossa, abdominal distension, and fever. At the time, fluid collection in the left inguinal region was diagnosed, and drainage was performed with serosanguineous secretion. The patient showed a partial improvement in the symptoms, and, a few months later, a serous discharge in the collection area began, which was cyclical,

and it appeared eight days before menstruation and lasted until the end of the menstrual period. Thereafter, she was subjected to two more surgical procedures, including adhesiolysis in the fistula tract and drainage collection. The patient was in possession of the histopathological results of the latter approach (performed in 2013), which showed a foreign body-type chronic granulomatous inflammation and nonspecific lymphadenitis.

Based on this clinical background, the patient was referred to the hospital's gynecology team with a diagnosis of tubocutaneous fistula due to endometriosis. A gynecological examination revealed the presence of a pfannenstiell scar and a longitudinal scar on the left iliac fossa, and a speculum examination showed no communication of the vaginal walls with the fistula. The patient underwent additional tests. An abdominal ultrasound examination showed: a hypoechoic tract with a diameter of 0.5 cm, located in the left inguinal region

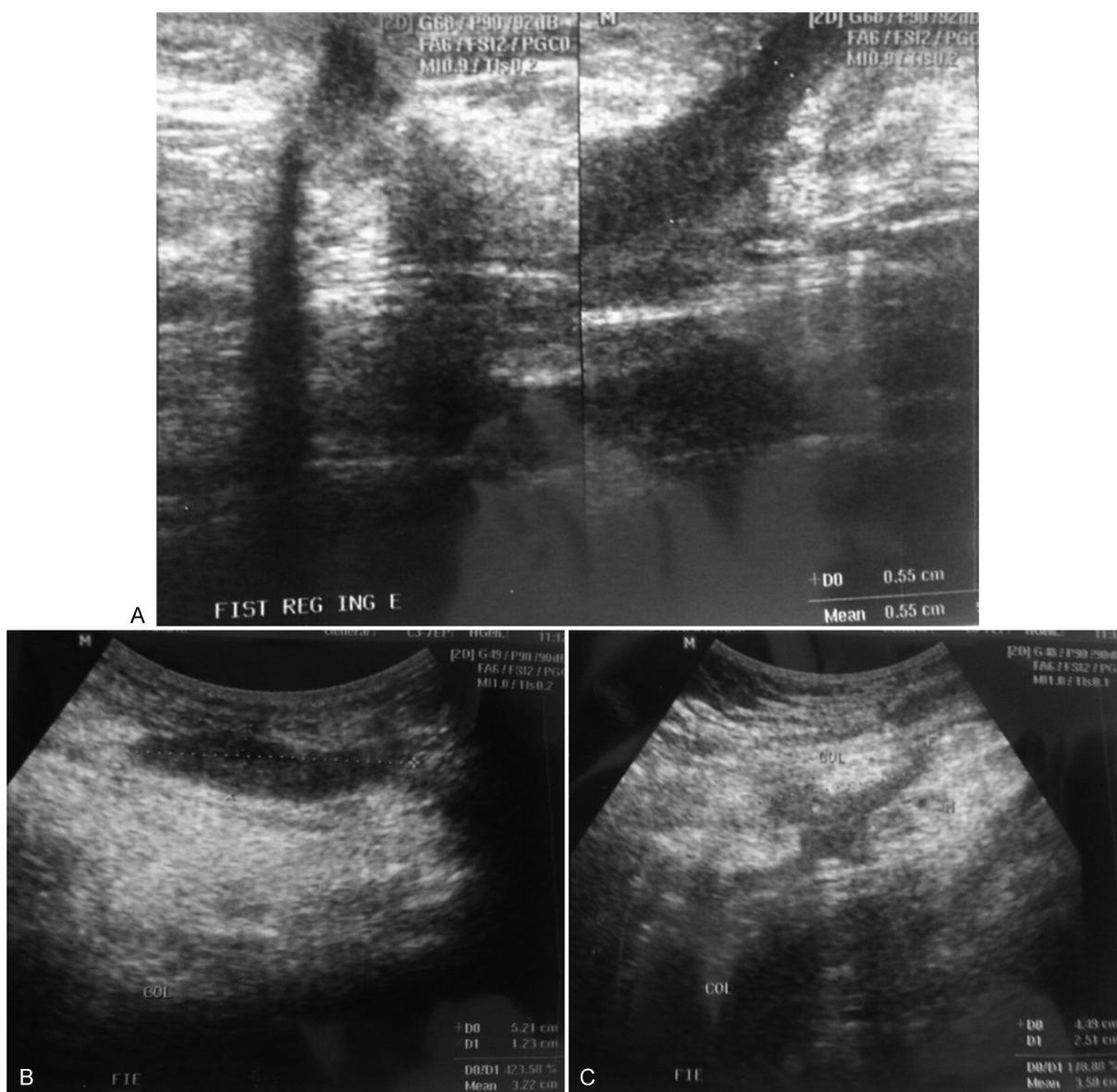


Fig. 1 Abdominal ultrasound showing a hypoechoic tract located in the left inguinal region (panel A); a hypoechoic area located in the subcutaneous tissue, suggesting fluid collection, and showing communication with the external environment through the aforementioned tract (panel B); and a second hypoechoic area, showing communication with the aforementioned lesion, located close to the abdominal and internal oblique rectal muscles, suggestive of fluid collection (panel C).

(►**Fig. 1A**); a hypoechoic area, with partially defined limits, located in the subcutaneous tissue, measuring 5.2×1.2 cm, suggesting fluid collection, and showing communication with the external environment through the aforementioned tract (►**Fig. 1B**); and a second hypoechoic area, with partially defined limits, showing communication with the aforementioned lesion, located close to the abdominal and internal oblique rectal muscles, measuring 4.5×2.5 cm, suggestive of fluid collection (►**Fig. 1C**). Magnetic resonance imaging (MRI) of the pelvis showed laminar pelvic fluid collection on the posterior aspect of the rectus abdominis muscle on the left, next to the pubis, measuring $\sim 6.2 \times 0.9$ cm, extending to the ipsilateral iliac fossa. In this area, a fistulous tract could be observed that pierced the musculature, forming another subcutaneous laminar fluid collection area measuring $\sim 5.3 \times 1.0$ cm and draining at its lower portion toward the cutaneous fistula (►**Fig. 2**). Fistulography revealed a contrast-filled cavitation in the subcutaneous area of the inguinal region, extending laterally to the left iliac fossa for ~ 8 to 10 cm, without communication with the viscera or deep planes (►**Fig. 3**).

Given that the patient had no cutaneous opening discharge at that time, she was discharged from the hospital with guidance to return at the first sign of secretion drainage, which occurred 10 days afterwards (►**Fig. 4**). She was readmitted and underwent surgery, which was performed over two sessions by gynecology, general surgery, and urology specialists on September 21, 2015. Initially, a surgical hysteroscopy was performed, in which a normal uterine cavity was observed with tubal ostia, and no presence of lesions was detected. However, an outlet of solution was observed, which was used for distension during the procedure, via the cutaneous opening of the fistula. Methylene blue was not visible after introduction into the fistula hole. Later, a laparotomy was performed with supra-aponeurotic resection and excision of the entire wall and fistulous tract, which exhibited the discharge of a chocolatey secretion. At the opening of the aponeurosis, a new extraperitoneal



Fig. 2 Magnetic resonance imaging of the pelvis showing laminar pelvic fluid collection on the posterior aspect of the rectus abdominis muscle on the left, extending to the ipsilateral iliac fossa. In this area, a fistulous tract could be observed that pierced the musculature, forming another subcutaneous laminar fluid collection area and draining at its lower portion toward the cutaneous fistula.

collection area could be seen closely adhering to the left horn. A left salpingectomy was performed with the excision of the entire wall of the collection area. The histopathology results confirmed the diagnosis of endometriosis.

The patient was readmitted on the 14th postoperative day for drainage of the purulent secretion via the surgical wound. She was given antibiotic therapy guided by the secretion culture; she recovered well, and was discharged. Seven weeks after the procedure, she was readmitted at an outpatient basis and exhibited complete healing of the wound without secretion drainage or pain complaints; the patient was in amenorrhea due to the continued use of desogestrel.



Fig. 3 Fistulography showing a contrast-filled cavitation in the subcutaneous area of the inguinal region, extending laterally to the left iliac fossa for ~ 8 to 10 cm, without communication with the viscera or deep planes.



Fig. 4 Physical examination showing secretion drainage of the cutaneous opening in the left iliac fossa.

Discussion

Extrapelvic endometriosis may be associated with a wide variety of cyclic symptoms reflecting the affected organs.¹ Physical findings, when present, are related to the location and extent of the disease,⁵ and greater diagnostic sensitivity is present when the patient is investigated during menstruation.⁶ The present case showed a clinical background highly suggestive of endometriosis due to the cyclical character of the presence of fistula debit and the characteristics of the drained content, which varied in color from citric yellow to bright red.

Development of endometrioma in the surgical scar after cesarean section is a rare complication, with a reported frequency of no more than 0.4%.⁷ The possible mechanisms involved in the formation of female genital tract fistulas include previous pelvic surgery, the use of drains, surgical wound dehiscence, and invasive endometriosis.⁸⁻¹⁰ In the present case, the possible triggering factors included the occurrence of a previous cesarean section and the use of drains in the left iliac fossa collection area. The endometrial tissue may have been implanted in the surgical scar, causing the erosion of the underlying tissue through a cyclical inflammatory process.

Magnetic resonance imaging is superior to transvaginal ultrasound for detecting the peritoneal implants of endometriosis and collections, but it still identifies only 30–40% of the lesions observed during surgery.¹¹ The patient in question was submitted to the cited additional tests, which diagnosed the presence of two liquid collection areas, but did not show the connection of these collection areas with the ipsilateral fallopian tube.

Surgery is the treatment of choice in cases of endometriotic fistula. Hormone therapy may lead to an improvement in symptoms, but does not eradicate such lesions.^{12,13} To avoid recurrence and the emergence of additional complications, the patient underwent complete surgical excision of the lesion. Because the patient was young, and in order to avoid chronic problems resulting from estrogen deprivation, we decided to preserve the ovaries.

In summary, we present an extremely rare case of an endometriotic tubocutaneous fistula, the description and literature review of which provide greater awareness of this clinical entity, offering useful information for the correct diagnosis and treatment.

References

- 1 Fritz MA, Speroff L. *Endocrinologia, ginecologia e infertilidade*. Rio de Janeiro: Revinter; 2015
- 2 Kuohung W, Jones GL, Vitonis AF, et al. Characteristics of patients with endometriosis in the United States and the United Kingdom. *Fertil Steril* 2002;78(4):767–772
- 3 Hediger ML, Hartnett HJ, Louis GM. Association of endometriosis with body size and figure. *Fertil Steril* 2005;84(5):1366–1374
- 4 Taff L, Jones S. Cesarean scar endometriosis. A report of two cases. *J Reprod Med* 2002;47(1):50–52
- 5 Vercellini P, Trespidi L, De Giorgi O, Cortesi I, Parazzini F, Crosignani PG. Endometriosis and pelvic pain: relation to disease stage and localization. *Fertil Steril* 1996;65(2):299–304
- 6 Koninckx PR, Meuleman C, Oosterlynck D, Cornillie FJ. Diagnosis of deep endometriosis by clinical examination during menstruation and plasma CA-125 concentration. *Fertil Steril* 1996;65(2):280–287
- 7 Roncoroni L, Costi R, Violi V, Nunziata R. Endometriosis on laparotomy scar. A three-case report. *Arch Gynecol Obstet* 2001;265(3):165–167
- 8 Doderio D, Corticelli A, Caporale E, Cardamone C. Endometriosis arises from implant of endometriotic cells outside the uterus: a report of active vesicouterine centrifugal fistula. *Clin Exp Obstet Gynecol* 2001;28(2):97–99
- 9 Floberg J, Bäckdahl M, Silferswärd C, Thomassen PA. Postpartum perforation of the colon due to endometriosis. *Acta Obstet Gynecol Scand* 1984;63(2):183–184
- 10 Gupta SK, Shukla VK, Varma DN, Roy SK. Uterocutaneous fistula. *Postgrad Med J* 1993;69(816):822–823
- 11 Stratton P, Winkel C, Premkumar A, et al. Diagnostic accuracy of laparoscopy, magnetic resonance imaging, and histopathologic examination for the detection of endometriosis. *Fertil Steril* 2003;79(5):1078–1085
- 12 Purvis RS, Tyring SK. Cutaneous and subcutaneous endometriosis. Surgical and hormonal therapy. *J Dermatol Surg Oncol* 1994;20(10):693–695
- 13 Rivlin ME, Das SK, Patel RB, Meeks GR. Leuprolide acetate in the management of cesarean scar endometriosis. *Obstet Gynecol* 1995;85(5 Pt 2):838–839