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Bi-salpingo Colonic Fistula Report of Both Fallopian Tubes Fistulizing with Sigmoid Diverticulum with Literature Review

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Abstract. Fistulous communication between the colon and female adnexal structure rarely occurs. The rarity of the condition reflects that only few cases have been reported in the literature, however, the bisalpingocolonic fistulae have never been reported. A case study of a 28-years-old woman with primary infertility of four years duration diagnosed with bilateral salpingocolonic fistulae by hysterosalpingography (HSG), which was performed as part of the routine work-up of infertility showed; contrast filed both tubes till fimbrial ends then joined diverticulum in sigmoid colon with opacification down to the rectum. Laparoscopic salpingectomy and closure of fistula were done. The patient was referred to the assisted conception unit and underwent two *in vitro* fertilization. An embryo transfer resulted on the second attempt in pregnancy and a normal healthy full term baby. In conclusions, Salpingocolonic fistula is a very rare disease and closure of the fistula is the treatment of choice with the need to IVF assisted conception.

Keywords: Tubo colonic fistula or colosalpingeal fistula, Diverticular disease, Infertility

Introduction

Fistulous communication between the colon and female adnexal structure rarely occurs. The rarity of the condition is reflected in the fact that only

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few cases have been reported in the literature, but the bisalpingocolonic fistulae have never been reported. Fistulae have been described between the fallopian tube and rectum, sigmoid, appendix, caecum and ileum, patient with this previous fistula may be asymptomatic^[1-6].

Diverticular disease is the most common colonic disorder in the Western world, with 50% of people over the age of 80 years having evidence of the diverticulosis. Fistulae are present in 20% of those who undergo surgery for complications of the condition. The great majority of fistulae being from the sigmoid to the bladder or vagina^[1-4,7].

Because of the close proximity of the female reproductive organs with other pelvic organs, fistulation can occur between the ureter, small or large bowels resulting in vesicovaginal, vesicouterine, vesicoenteric, ureterovaginal, ureteroenteric and enterovaginal fistulas^[8-10].

There is a wide variety of complaints in a woman being investigated for infertility suspected with fistulization. This ranges from the asymptomatic to the history of cyclic hematuria, and discharge of urine, feces, foul smelling secretions or air through unfamiliar orifices with perineal dermatitis^[11-13]. The actual demonstration of the fistulous tract and identification of its underlying cause is quite tedious and difficult. Hysterosalpingogram, a common radiological examination is routinely used in the investigation of infertility and sub-fertility in women^[14]. This report presents, bi-salpingocolonic fistula caused primary infertility on a patient treated by laparoscopic salpingectomy, and closure of the colonic fistula led to successful assisted IVF conception on second attempts.

Case Report

A 28-year-old female presented to King Abdulaziz University Hospital infertility clinic with primary infertility for four years. Patient had history of regular period with no dysmenorrhea, dyspareunia, unusual vaginal discharge, and history of weight gain, galactorrhea or any bowel disease. She had a past history of laparoscopy since 2 years for ovarian cystectomy. Physical examination revealed a healthy female with no relevant abnormal general or local genital physical findings apart of the scar in umbilicus from previous laparoscopy. Hormonal profile revealed, prolactin level 98 mIU/L, TSH 6.37 mIU/L, FSH 3.27, LH 2.84 mIU/L. The husband was clinically normal and all investigations for male factors of infertility were within normal. Pelvis ultrasound was

done on the third menstrual day; the uterus was AVF measuring $7.0 \times 3.0 \times 4.5$ cm and it is heterogenous in echotexture, with no fibroid, ovarian mass, or free fluid in pouch of Douglas. The endometrium measures 0.40 cm, while both ovaries appeared normal, the right ovary measured $2.4 \times 1.9 \times 3.3$ cm while left ovary measured $2.6 \times 1.9 \times 3$ cm. Both ovaries had few tiny follicles.

The patient was started on L-thyroxin to correct hypothyroidism, and three cycles of ovulation induction with Clomid, and human menopausal gonadotrophin were conducted with no pregnancy. Hysterosalpingogram (HSG) was done under normal sterile procedure using non-ionic contrast (iohexol 300 mg). Following initial injection of about 8 mls of contrast, normal endometrium outline with no filling defect, contrast filed both tubes till fimbrial ends, then joined small diverticulum in sigmoid opacifing down to the rectum (Fig. 1). There is no evidence of free intraperitoneal spilling. The conclusion was bilateral tubo colonic fistula. Barium enema was done with a single contrast, appropriate spots



Fig. 1. Hysterosalpingography showing bi-salpingocolonic fistula.

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film obtained, no fistula could be detected and normal flow of contrast through the rectum sigmoid, descending colon, transverse and ascending colon, with normal mucosal pattern without any narrowing, stricture or filling defect. The patient and her husband were counseled and were opted for fistula resection. This was carried out successfully by laparoscopic salpingectomy and closure of colonic fistula performed under general anesthesia in lithotomy position with small subumbilical incision, Veress needle was inserted, and gas is used to inflate. A 10 mm trocar was inserted, scoping showed moderate adhesion while uterus looked normal. Both tubes inspected, pulled together and seen adhered to the sigmoid colon, and multiple small spots of endometriosis in the uterus and pouch of Douglas. Subsequently, 4 more 5 mm trocar was inserted in the right and left iliac fossa, and right to the umbilical line subumbilical for manipulation. Adhesiolysis, performed between tubes and colon then dissected away from the colon and a salpingectomy was performed. The patient was referred to the assisted conception unit and underwent two in vitro fertilization, plus embryo transfer cycles when second trail was successful, thus, she had normal healthy full term baby.

Discussion

Perforation of an inflamed diverticulum can lead to abscess or fistula formation between the colon and the adjacent organs, most commonly resulting in colovesical and colovaginal communication^[15]. A direct extension of diverticulitis into the adnexa is rare, despite of the proximity of the left ovary to the sigmoid colon. Only few case reports are found describing salpingocolonic fistulas complicating diverticulitis, but none of them reports bi-salpingocolic fistula^[15-17]. As found in other studies, complications of diverticulitis are difficult to diagnose on the basis of clinical and laboratory signs^[18]. None of salpingocolonic fistulae cases had specific gynecologic symptoms of this complication. Their physical signs and symptoms could be explained by diverticulitis alone^[19]. Moreover, the clinical findings in our patient, with and those without salpingocolonic fistula were similar. Patient with salpingocolonic fistula may be asymptomatic as appeared in our case and reported cases, Sanjeeb *et al.*^[1] and Petignat *et al.*^[6]. Other possible causes of this rare type of fistula include Crohn's disease^[3-5]. While the rectum and bladder are some of the more common sites of fistula formation in Crohn's disease, the fallopian tube is extremely rare^[3]. Michelassi et al.^[3]

reviewed 18 years of fistulizing Crohn's disease at the University of Chicago and found two cases of entero-salpingeal fistulas out of 290 fistulas. There is no feature of Crohn's disease in this patient presented; however, the lifetime risk of developing fistulas for patients with Crohn's disease has been reported to be 20-40%^[3]. The diagnosis of salpingocolonic fistula in most cases is usually incidentally during HSG, for whatever reason, commonly infertility. This was the finding in the case presented, but in bi-salpingocolonic fistula, which never reported before. Others, however, found ultrasonography a valuable non-invasive alternative modality for diagnosis of pelvic fistula. As it, visualize vesicouterine fistula, permitting correct diagnosis and obviating the need for further invasive procedure^[20,21]. Occasionally, salpingocolonic fistulas or salpingoenteric fistula may be diagnosed intraoperatively, especially those resulting from Crohn's disease and complicating diverticulitis ^[3,4,22].

Fistulae to the fallopian tubes are rare, so their management is not very well described. But it is important to note that the contribution of the bi-salpingocolonic fistula to the infertility in our patient is very well defined. As both tubes joined with sigmoid colon as such opted for only fistula resection, and assisted conception as in our patient when IVF succeeded in conception within second attempts, resulting in a full term healthy baby after laparoscopic salpingectomy and closure of the colonic fistula. Most studies reporting salpingocolonic fistula advocate fistula closure with salpingectomy to treat this rare disease, and to prevent the ectopic pregnancy subsequently^[23-26]. In fistulae resulting from Crohn's disease and complicated diverticulitis, en-bloc fistula resection and salpingectomy are also recommended^[3-5]. However, the type of resection will be tailored to the peculiarity of the patient.

Conclusion

Salpingocolonic fistulae from any cause are rare and may be a silent disease that can cause infertility or can be an associated factor. The use of hysterosalpingography as complimentary modality for investigation of infertility is once again, strengthened. Laparoscopic salpingectomy as a treatment to salpingocolonic fistula is a feasible treatment option available for this rare presentation, as in this case report. The availability and accessibility to assisted reproductive technology will help this type of patient achieve successful pregnancy.

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تقرير حالة ناسورتنائي من كل من قناتي فالوب مع رتج القولون السيني مع مراجعة الأدب

نوال سالم السيناني

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المستخلص. الناسور بين القولون والملحقات الأنثوية (الأنبوبية) نادرة الحدوث. ندرة هذه الحالات تنعكس بقلة الحالات المنشورة في الأدب الطبى. على كل حال لم تسجل أي حالة لنواسير قولونيية أنبوبية في الجهتين. هناك حالة لسيدة عمرها ٢٨ سنة مع عقم أولى ل ٤ سنوات شخص لها ناسور قولوني أنبوبي بالجهتين عن طريق التصوير الرحمى الأنبوبي الظليل الذي أجري لها بشكل روتيني لاستقصاء أسباب العقم، أظهر أن المادة الظليلة ملأت الأنبوبين حتى النهايات الأهدوبية، ثم اندمجت مع الرتج في القولون السيني مع وصول المادة إلى المستقيم. تم استئصال الأنابيب واغلاق النواسير باستخدام المنظار. حولت المريضة إلى وحدة مساعدة الحمل وتمت محاولتين لتلقيح مجهري. وتم نقل جنين في المحاولة الثانية وأدت إلى ولادة طبيعية لطفل بتمام الحمل. الخاتمة: الناسور القولوني الأنبوبي نادر جدًا، وقد يتسبب في عقم دون ظهور أعراض، ويجب استبعاد وجوده بعد استبعاد العوامل الأخرى للعقم. إن إغلاق الناسور بين القولون والجهاز التناسلي هو العلاج الأنسب وبعدها يحال المريض لوحدة المساعدة في الحمل.