CASE REPORT

Insidious perforation of the rectum by a fallopian tube: the need to keep 'an open mind' when dealing with deep infiltrating endometriosis (DIE)

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SUMMARY

Endometriosis is a benign chronic disease which can have different degrees of severity and can potentially affect any organ. Intestinal endometriosis occurs in 3%–37% of the cases, being more frequent in the rectosigmoid transition. Transmural involvement of intestinal endometriosis is extremely rare and is usually associated with recurrent abdominal pain. Due to the cyclical hormone influence, endometriosis implants may infiltrate the deeper layers of the intestinal wall and may lead to bowel obstruction or perforation. We present a case of transmural perforation of the rectum wall by an adjacent organ (left fallopian tube) that occurred insidiously in a patient with deep infiltrative endometriosis. A complete set of images is presented, regarding the preoperative, intraoperative and postoperative findings.

BACKGROUND

Endometriosis is a benign chronic disease that is defined by the presence of endometrial tissue outside the uterine cavity. Although its real prevalence is unknown, it is estimated that it affects about 10%–15% of women of reproductive age¹ and about 30%–50% of women with infertility diagnosis.² In addition to its strong association with infertility, its clinical relevance is based on the fact that endometriosis is the most frequent cause of pelvic pain in women of reproductive age and may also cause other severe symptoms like dysmenorrhea, deep dyspareunia, dyschezia and dysuria.

In terms of classification, deep infiltrative endometriosis (DIE) is considered when the disease infiltrates at a depth of more than 5 mm. Ectopic endometrial implants can occur nearly anywhere in the body, but the most frequent locations of the DIE are ovaries, uterosacral ligaments, peritoneum, rectovaginal septum, fallopian tubes, rectosigmoid colon and bladder.

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Usually, intestinal endometriosis typically takes the form of asymptomatic superficial implants but, occasionally, as a result of cyclical hormonal influences, these implants may proliferate and infiltrate the deeper layers of the intestinal wall. However, the involvement of the submucosa and mucosa (transmural involvement) is rare and is usually associated

with recurrent abdominal pain.⁵ When there is transmural involvement of the wall, obstruction or perforation of the bowel may occur.

Although the definitive diagnosis is histological, the association of clinical history and examination, complemented by imaging and endoscopic examinations, allows us to predict the existence of endometriosis with a high degree of reliability. For this reason, imaging evaluation of patients with suspected DIE is fundamental not only for the differential diagnosis with other diseases but also for planning the surgical approach.

Transvaginal ultrasound should be the first imaging test to be performed because it is a simple and affordable test. In addition, when performed by an experienced professional, it allows the diagnosis of DIE with great reliability. Hudelist *et al* evaluated the diagnostic efficacy of ultrasonography in the diagnosis of intestinal endometriosis with a sensitivity and a specificity of 91% and 98%, respectively.⁶ MRI is complementary to ultrasound and may be indicated in cases where DIE is suspected at specific sites (such as in the ureters, appendiceal, above the rectosigmoid junction) or in cases where the ultrasound is not completely enlightening.

The main goals of DIE treatment are symptoms improvement and optimisation of the fertility. Medical treatment should be the first therapeutic approach, being the surgery reserved for cases of poor response to medical treatment or when the woman intends to become pregnant.

DIE surgery is a highly complex procedure that must be performed by multidisciplinary teams with great experience in the treatment of this disease. Given the pathophysiology of the disease, surgeries can be highly demanding. Therefore, a complete preoperative study as well as the presence of a specialised team is mandatory to optimise the results. However, even when all these prerequisites are met, intraoperatively unexpected findings can happen and one should be prepared.

We present a case of a patient with DIE, whose preoperative examinations suggested the presence of a nodule of the rectovaginal septum that infiltrated the rectosigmoid transition and whose intraoperative findings revealed instead to be a transmural perforation of the rectum by the left fallopian tube. We also present the correlation between preoperative examinations and intraoperative findings.

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Figure 1 MRI T2-weighted sagittal image shows an exofit image originated in the left adnexal region of heterogeneous appearance with poorly defined limits and which seemed to creep into the lumen of the sigmoid colon. It also revealed a thickening of the rectovaginal fascia and the uterosacral ligaments.

CASE PRESENTATION

A 38-year-old woman, nulliparous, with medical history of primary infertility and chronic pelvic pain was referred to our hospital. She complained of chronic pelvic pain and dysmenorrhea which had started 3 years ago when she stopped the estroprogestative contraceptive pill to get pregnant. She also complained of deep dyspareunia, dyschezia and occasional rectal bleeding.

On gynaecological examination, bimanual evaluation revealed a tender, fixed and painful retrocervical nodule which seemed to involve the left uterosacral ligament and an elastic mass was palpable on the left adnexal area. No other positive findings were found on examination.

Given the clinical suspicion of DIE and the recognised benefits of hormonal contraceptives in this disease, an estroprogestative contraceptive pill was started to minimise patient symptoms.

INVESTIGATIONS

Considering the clinical hypothesis of DIE, additional diagnostic tests were performed:

MRI (figure 1):

DIFFERENTIAL DIAGNOSIS

In the context of a patient with DIE suspected of intestinal involvement, a colonoscopy was performed (figure 2).

TREATMENT

Given the severity of the symptoms, the desire for fertility and the presence of lesions compatible with DIE of the rectovaginal septum with probable intestinal infiltration, the patient was proposed for surgery (left ovary cystectomy and excision of the rectovaginal septum nodule). Since an intestinal lesion coexisted, the patient was prepared for an eventual intestinal resection.

For its well-known advantages, a laparoscopic surgery was proposed in partnership with a colorectal surgeon.

During surgery, a frozen pelvis was observed. The right ovary was adherent to the ovarian fossa and the left ovary was not visible, being blocked by the presence of extensive adhesions that conditioned the pulling of the rectosigmoid transition to the left pelvic wall and homolateral adnexa (figure 3).

Figure 2 Colonoscopy confirmed the presence of a sessile luminal lesion with 3.5 cm, 12 cm from the anal margin. A biopsy was performed and histological results revealed an inflammatory polyp of the colon.

Adhesiolysis was initiated, and once the dissection of the left pararectal space had begun, it was found that there was no cleavage plane between the left fallopian tube and the rectum wall. A more thorough dissection found that the tube pierced the rectum, and the distal part of the fallopian tube was found in an endoluminal position.

After the fallopian tube was released, a hole in the anterior wall of the rectum was found, with approximately 1.5 cm, corresponding to the transmural pathway of the fallopian tube, which pierced the rectum (figure 4). It was verified that the rectal endoluminal formation described in MRI and found in the colonoscopy corresponded to the left fallopian tube (figure 2).

Left salpingectomy, discoid intestinal resection of the rectum and cystectomy were performed (figure 5). The histological diagnosis was compatible with tubal endometriosis and chronic salpingitis, and there were no suggestive aspects of endometriosis in the excised rectal segment.

OUTCOME AND FOLLOW-UP

The postoperative period was uneventful. Patient was discharged on the fifth postoperative day, and there were no postoperative complications. She was re-evaluated 4 weeks after surgery and was asymptomatic. Patient is now awaiting infertility treatment.

Figure 3 Left adnexa blocked by thick adhesions to the rectosigmoid transition.

Figure 4 Hole in the anterior wall of the rectum after removal of the distal portion of the tube (pavilion) that pierced the entire wall thickness up to the lumen of the rectum.

The surgery was performed 3 months ago and the MRI performed 2 months after surgery showed no abnormal findings (figure 6).

DISCUSSION

Although endometriosis is a relatively frequent disease, intestinal endometriosis is uncommon, affecting 5.3% to 12% of women with DIE.⁷ Chapron *et al* found in their study that, regarding intestinal DIE, the most frequent sites affected were the rectum and the rectosigmoidal junction (65.7%).⁴

The main symptom of colorectal endometriosis is pelvic pain that can radiate to the rectum and anus, although other symptoms such as rectal bleeding, dyschesia or diarrhoea may occur. Nevertheless, 35% of the patients are asymptomatic.⁸

The foci of deep endometriosis are constituted by associated endometrial, fibromuscular and adipose tissues which infiltrate adjacent organs, forming solid plaques.

This pathophysiological process usually occurs in a progressive way, being able to reach different levels of severity, depth and, consequently, different levels of anatomic distortion.

When surgery is considered, preoperative imaging is crucial for surgery planning with pelvic ultrasound being the first choice for pelvic imaging. Since the ultrasound limit is the rectosigmoid transition level, MRI may be important to complement the preoperative evaluation of patients suspected of having severe

Figure 5 Anterior discoid resection of the rectum.

Figure 6 MRI T2-weighted sagittal figure 2 months after surgery. No endoluminal lesions were found.

disease. In such cases, colonoscopy is also important because it allows the identification of lesions that affect the intestinal mucosa to obtain histological samples and to define the distance between the anal margin and the lesion.⁹

However, as demonstrated in this case, even when there is involvement of the intestinal mucosa, the samples obtained are often insufficient to predict a definitive histological diagnosis.¹⁰ The previous evaluation of the distance between the lesion and the anus is important in the definition of the surgical strategy, preparing the surgical team and the patient for the possible interventions and surgical complications.

Although rare, it is known that intestinal DIE can be associated with serious complications such as intestinal occlusion or intestinal perforation. Intestinal perforation due to endometriosis is very rare and has been reported in the literature, most frequently associated with pregnancy or women who have recently given birth.^{5 11} However, there have also been published some cases of bowel perforation in non-pregnant women although they are usually associated with acute abdominal symptoms.¹²

In the presented case, although there was a transmural perforation of the rectum wall, this happened insidiously, without the occurrence of signs or symptoms of acute abdomen. Besides, what is different from other cases published is that the perforation occurred by the fallopian tube and not by an endometriotic implant as usually occurs.

This clinical case represents well the inflammatory and adherent character of the endometriosis evidencing the degree of anatomical distortion with which the surgeons can confront.

In addition, our case highlights the importance of a complete preoperative study when dealing with severe DIE. In our case, despite the unexpected intraoperative finding of transmural perforation of the rectum by the fallopian tube, the presurgical imaging evaluation was essential to predefine the surgical steps of the surgery as well as to provide accurate information for the preoperative counselling of our patient, informing her about the possibility of bowel resection.

The presence of such an unexpected finding during the surgery corroborates the great complexity of endometriosis pathophysiology but also the importance of the multidisciplinary performance of these surgeries by teams experienced in this disease setting.

The interest of this case rests in this exceptional form of presentation of DIE, with insidious transmural perforation

of the bowel by the left fallopian tube and also in the set of images obtained before, during and after surgery which allowed the complete imagiological description of this rare case.

Learning points

- The differential diagnosis of deep infiltrative endometriosis with colon involvement is challenging and requires knowledge of other infiltrative pathologies, including malignant disease.
- The appropriate preoperative assessment using the adequate imaging tests may allow for an accurate preoperative diagnosis of the location and extent of the disease, which is extremely important for the correct surgical planning.
- Bowel perforation is rare and usually occurs with acute symptomatology although it may occur insidiously.
- The definitive diagnosis of the true severity of the disease is only possible intraoperatively and the presence of a highly differentiated team is mandatory for the best outcomes.

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