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CLINICAL CASE

Intestinal endometriosis as a cause of intestinal stenosis. A case report

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Abstract

Endometriosis affecting the terminal ileum is a rare condition, appearing in 1-7% of women with endometriosis, with secondary intestinal occlusion estimated in 7-23%. Diagnosis before surgery is difficult and requires a high clinical suspicion; it should be a differential diagnosis in all women with abdominal pain. The clinical case is a 26-year-old female presented with abdominal pain and a lack of bowel movements, with poor response to medical management. A computed tomography scan shows signs suggestive of intestinal obstruction, leading to surgical treatment. A diagnostic laparoscopy was initially planned; however, due to the unavailability of the necessary equipment, a laparotomy was performed. Two areas of stenosis were found in the terminal ileum, managed with a right hemicolectomy and ileocolic anastomosis. Pathology reports revealed endometriotic implants in the areas of obstruction. In conclusion, while there is not established consensus regarding surgical management, a minimally invasive approach is recommended when feasible. Diagnosis can be confirmed only on histopathological examination of the surgically resected specimen. The decrease in abdominal pain, gastrointestinal discomfort, and the improvement in the quality of life are evident following surgical treatment.

Keywords: *Ileal endometriosis. Right hemicolectomy. Occlusion. Intestinal resection. Endometriosis. Stenosis.*

Introduction

Endometriosis is defined as the presence of endometrium in an abnormal or ectopic location. Histologically, it is the presence of endometrial-like tissue or glands outside the uterine cavity. It is a hormone-dependent gynecological disorder that is most often seen in reproductively active women[1,2].

It is a common cause of pain and infertility, but it also negatively affects quality of life, intimate relationships, participation in daily activities, social activity, productivity, and income[2].

In their study, Fuldeore et al. report that the prevalence of diagnosed endometriosis was estimated at 6.1% (2,922 out of 48,020 women surveyed); 52.7% of women were between 18 and 29 years old when they were diagnosed with endometriosis. The majority (86.2%) of women experience symptoms before diagnosis[3]. In Mexico, the incidence of endometriosis in women with primary and secondary infertility diagnoses has been studied, reaching 34.5% in a sample of 197 patients; however, the number of fertile patients with endometriosis is unknown[2].

The main theories about the pathogenesis of endometriosis seek to explain the appearance of endometrial tissue outside the uterus. Among them are: the theory of retrograde menstruation, proposed by Sampson in 1927, suggests that during menstruation, part of the

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flow moves through the fallopian tubes into the peritoneal cavity, carrying with it endometrial cells that adhere and proliferate in ectopic locations. Although it explains the presence in the peritoneal cavity, it fails to justify other locations of the disease. Theory of coelomic metaplasia: states that peritoneal mesothelium cells can transform into endometrial tissue under specific hormonal or inflammatory stimuli. This could explain the presence of endometriosis in more atypical sites, such as the diaphragm, where the phenomenon of retrograde menstruation would not have a direct role. Theory of lymphatic and blood dissemination: It proposes that endometrial cells migrate through the lymphatic system and blood circulation, which would justify the appearance of endometriosis in distant organs such as the lungs, brain, or even in the skin. Immunological theory: Suggests that a dysfunction of the immune system prevents the recognition and elimination of endometrial cells outside the uterus, allowing them to implant and grow. This would help to understand why some women with retrograde menstruation develop the disease, whereas others do not. Theory of genetic predisposition and epigenetics: Recent studies indicate that there are genetic and environmental factors that predispose some women to develop endometriosis, altering the expression of certain genes and favoring the formation of lesions. Together, these theories indicate that the pathogenesis of endometriosis is complex and multifactorial, combining immunological, genetic, hormonal, and anatomical characteristics to different degrees depending on each case[1,4-6].

Endometriosis affecting the gastrointestinal tract (infiltrating deep endometriosis) has been reported in between 3% and 37% of menstruating women. It is seen in the rectum-sigmoid, cecum, small intestine, and appendix in decreasing order of frequency. Involvement of the ileum is quite rare, and obstruction of the small intestine is very rare. Only a few cases have been reported so far, and all required emergency surgery[5,6].

We present the case of a patient with intestinal endometriosis who presented with intestinal pseudocclusion data, requiring emergency surgery. The diagnosis of endometriosis in the ileum was made by histopathology in the resected specimen.

Case presentation

This is a 26-year-old female patient, with no medical or surgical history before admission, date of last period 2 weeks before, regular cycles of 28 × 4 days, active sex life with barrier contraceptive method, dysmenorrhea of more than 5 years of evolution, attended in the emergency department for the presence of abdominal pain of 2 weeks of evolution, intermittent type, located in the right iliac fossa as well as in the hypogastrium, mild-to-moderate intensity, without irradiation to other areas and without apparent attenuation or aggravating factors, as the only accompanying symptom he mentioned absence of bowel movements 6 days before admission but with the presence of flatus, she self-medicated with oral analgesic having partial improvement, two similar previous conditions which were self-limiting.

On arrival at the emergency department, she had vital signs within normal parameters, physical examination with flat abdomen, slight distension, peristalsis present, decreased in frequency, pain on deep palpation generalized without evidence of acute abdomen, rest without alterations. As part of the diagnostic approach, laboratory tests were requested with only an alteration in leukocytosis of 14.08 ($\times 10^9$/L) at the expense of neutrophilia 79.5%. An abdominal X-ray in two positions and an abdominal ultrasound without reporting alterations.

As none of the studies were inconclusive and pain had decreased, strict medical surveillance was decided with bowel rest, defecatory surveillance, and analgesics. 24 h later, she again presented abdominal pain, absence of bowel movements, no fever, nausea or vomiting, demotion in leukocytosis and neutrophilia, for which a tomography of the abdomen and pelvis contrasted with the following findings: thickening of the ileal wall, maximum caliber of 44 mm, discrete striation of fat at the level of the terminal ileum at its junction with the cecum in close relation to the apparent right annex, rest unaltered.

It was decided to perform diagnostic laparoscopy; however, the equipment was not available, so laparotomy was performed, showing 12 areas of stenosis at the level of the terminal ileum, the first at 8 cm from the ileocecal junction, with an occlusion of 100% of its lumen when it rotated on its own axis, the second at 15 cm from the ileocecal junction with occlusion of approximately 60% of its lumen (Fig. 1). A right hemicolectomy was performed with mechanical ileotransverse anastomosis. The histopathological report of the specimen sent was: endometriosis in the muscle wall and serous wall of the ileum (transmural involvement), as well as secondary extrinsic compression in two areas, surgical edges without histological evidence of lesion (Fig. 2).

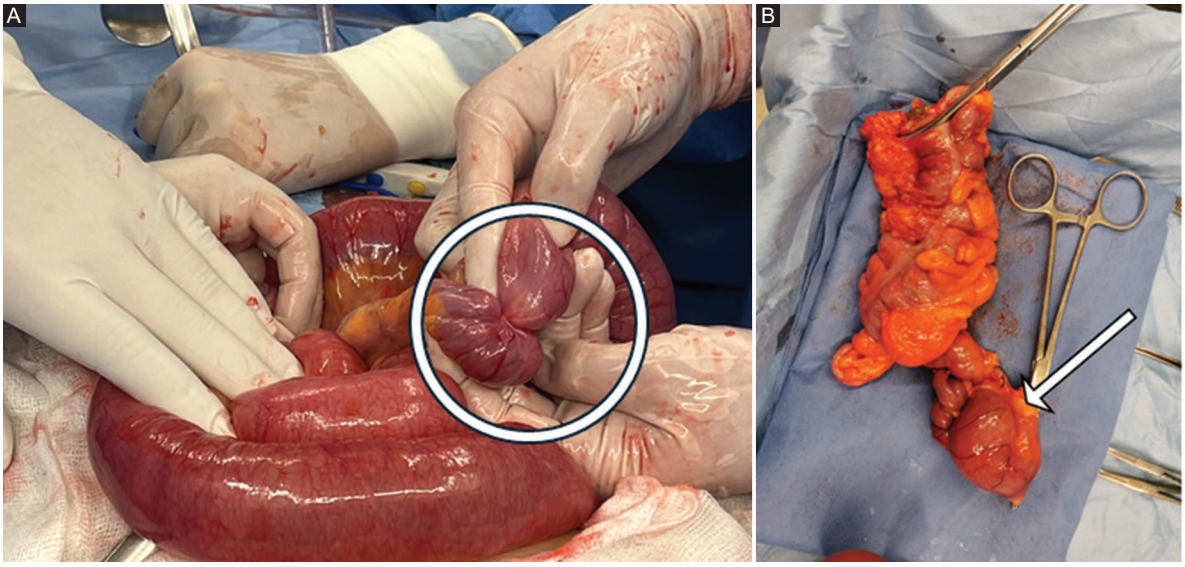

Figure 1. A: the area of stenosis in the terminal ileum (white circle), found as a finding during laparotomy, is appreciated. **B:** specimen resulting from right hemicolectomy with a stenosis area with torsion on its own axis (white arrow).

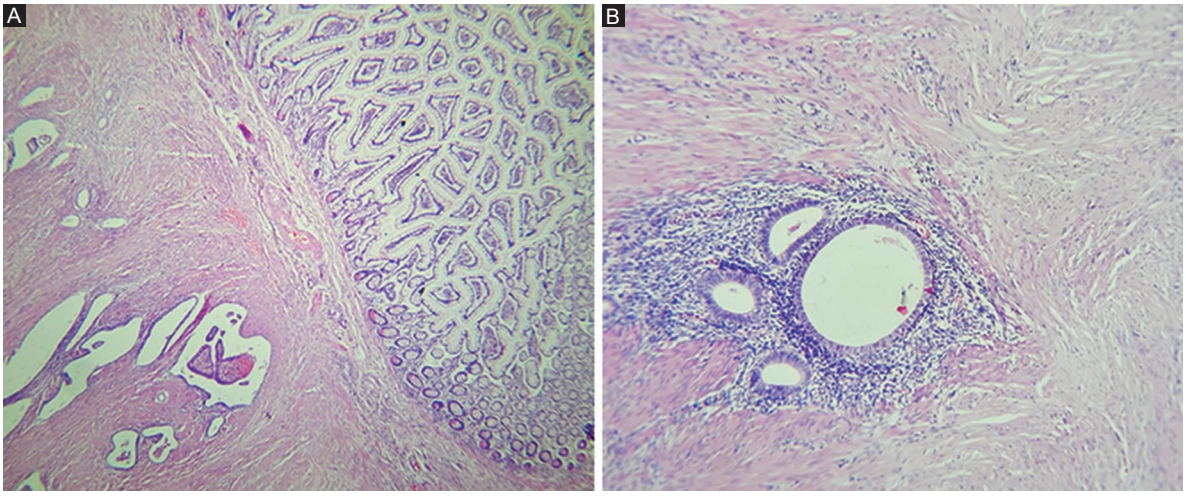

Figure 2. A: photomicrograph shows ileum mucosa. Below in the muscular mucosa, note the presence of glands and stroma with endometrial phenotype. **B:** photomicrograph shows glands and endometrial stroma without atypia, immersed in smooth muscle of the intestinal muscularis propria with transmural involvement.

The patient evolved favorably, tolerating a liquid diet at 24 h postoperatively and a normal diet at 36 h. The patient was discharged on the 6th day and summoned to an outpatient clinic for the removal of stitches, finding a wound in adequate condition. 2 months later, with total remission of symptoms, she was discharged from the General Surgery Service and sent to the Gynecology Service due to histopathological diagnosis of endometriosis.

Discussion

Countless hypotheses and theories have been proposed for the pathogenesis of endometriosis. From the implantation theory, which is currently the most widely accepted, to the stem cell theory. However, it is currently still a question to be resolved[1,2,5,6]. Endometriosis can be asymptomatic, but it also presents a wide variety of clinical manifestations, including pain, dyspareunia,

intermittent bleeding, and infertility. The symptoms that have the greatest impact on the quality of life of patients are those associated with pain, such as dysmenorrhea, profound dyspareunia, cyclic pelvic pain, dysuria, and dyschezia, which occur cyclically according to the menstrual cycle. In addition, intestinal endometriosis can cause irritative functional symptoms, such as diarrhea, intestinal cramps, hematochezia, and mucus expulsion, due to the cyclic release of inflammatory mediators, as well as obstructive mechanical symptoms, such as constipation and abdominal distension, caused by enlarged nodules, intestinal angulation and stenosis, and retraction of fibrotic tissue. Some specific symptoms, such as cyclic dysphasia and tenesmus, are typical of rectal endometriosis[1,2,5-7].

To date, no individual classification system adequately classifies endometriosis. The American Society for Reproductive Medicine's revised criteria for staging endometriosis (revised American Society for Reproductive Medicine) are the most widely used and are useful for clinicians to explain the severity of endometriosis in simple terms to patients. The ENZIAN classification describes in detail infiltrating deep endometriosis (PID) involving retroperitoneal and/or abdominal structures. In addition, the ENZIAN classification is probably most useful when determined using imaging modalities and can be used for surgical planning; however, it has little acceptance worldwide[1,8,9].

Previously, diagnostic imaging modalities were not very successful, but new advances in the field of imaging show promising results for detecting intestinal lesions. In a report, Gillen et al. show that multislice tomography combined with oral contrast located 94.8% of intestinal endometriotic nodules[10]. Magnetic resonance imaging has a high sensitivity for detecting endometriosis, but it has difficulty distinguishing it from other diseases, as well as being expensive[10-12]. Ultrasound, on the other hand, is an inexpensive and efficient means of examining and diagnosing intestinal endometriosis. However, diagnostic accuracy depends significantly on the sonographer's experience[1,13].

Due to the low frequency and non-specific symptoms, multiple differential diagnoses should be considered, such as tuberculous enteritis, yersinia enterocolitis, carcinoid tumors, lymphomas, Behçet's disease, and amoebomas, among others. The final diagnosis is based on histopathology and the presence of endometrial epithelial and stromal cells at ectopic sites[2,5,6,11-13].

The quality of the available evidence on medical treatment for intestinal endometriosis is suboptimal. Most studies were not comparative. There are very few reported cases in which infiltrating deep intestinal endometriosis has been resolved by non-surgical management when symptoms persist or worsen, and with the disadvantage that treatment cannot be interrupted[14].

Medical treatment should not be suggested if the lesion is located above the middle part of the rectum, the degree of lumen stenosis is > 60%, if the lesion infiltrates > 50% of the intestinal circumference, or if the largest diameter of the nodule is > 3 cm, the same rule is followed in the locations in the small intestine[7,14-16].

Surgical treatment depends on the degree of intestinal involvement and the clinical condition of the patient at the time of diagnosis. Ideally, it consists of intestinal resection of the affected ileal segment and primary anastomosis; however, cases have been reported in which right hemicolectomy is performed with or without anastomosis, preferring in either case the minimally invasive approach. When there is doubt of malignancy, resection with oncological criteria is justified and should be considered at the discretion of the surgeon[17-22].

Although successful cases have been presented with medical management, high recurrence rates continue to be shown in contrast to patients who undergo resection, have significant and persistent long-term improvement in pelvic pain, gastrointestinal discomfort, and quality of life, as well as a negligible recurrence rate[14,23,24].

Pharmacological therapy plays a fundamental role as a complement to surgical intervention, both in the pre-operative period and, more significantly, in the post-operative phase. In the pre-surgical context, its use can contribute to the reduction of the size of the lesions, thus facilitating the intervention. Subsequently, in the post-operative period, pharmacological therapy is essential to reduce the size of residual implants, control the progression of the disease in cases where surgical resection has not been able to be carried out completely, as well as to prevent the recurrence of the disease[25-27].

There are multiple therapeutic alternatives available for the management of endometriosis. Non-steroidal anti-inflammatory drugs are a widely used option in the treatment of chronic inflammatory diseases and have been shown to be effective in relieving primary dysmenorrhea. Combined oral contraceptives (COCs) and progestins, which are available in various presentations and routes of administration, represent the first line of hormonal treatment due to their efficacy and safety profile.

In cases where these therapies are not sufficient, the second line is mainly composed of gonadotropin-releasing hormone agonists. Although these agents have shown positive results in women who do not respond to COCs or progestins, it is important to mention that they require the addition of complementary treatments. On the other hand, the use of danazol has decreased significantly due to the availability of hormonal options with a superior safety profile and better tolerability. Since there are few data available on the long-term efficacy and safety of aromatase inhibitors, they should be given only to women with symptoms refractory to other conventional therapies in a clinical research setting[26,28-31].

Studies have shown that vitamin C and vitamin E supplementation is effective in reducing the severity of dysmenorrhea and improving dyspareunia, as well as decreasing the intensity of pelvic pain in patients with endometriosis[32-34].

Conclusion

Endometriosis with terminal ileum involvement is a rare and difficult to diagnose entity that can mimic multiple entities. This pathology should be suspected in all women of childbearing age who show intestinal symptoms without apparent causes traditionally demonstrable as if it were a functional digestive disorder, especially because of the implications that emergency surgery can have. Minimally invasive surgery should be considered the standard of management due to its widely demonstrated benefits, as well as a follow-up in conjunction with the gynecology service.

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Conflicts of interest

The authors declare no conflicts of interest.

Ethical considerations

Protection of human subjects and animals. The authors declare that no experiments on humans or animals were performed for this research.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patient, and secured approval from the Ethics Committee. SAGER guidelines have been followed as applicable to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing or creation of the content of this manuscript.

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