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Case Report

Ileocecal endometriosis: diagnosis and management

Ana López Carrasco [a, *], Alicia Hernández Gutiérrez [a], Paula A. Hidalgo Gutiérrez [b], Roberto Rodríguez González [a], José L. Marijuán Martín [c], Ignacio Zapardiel [a], Javier de Santiago García [a]

[a] Gynecology Department, La Paz University Hospital, Madrid, Spain
[b] Radiology Department, La Paz University Hospital, Madrid, Spain
[c] General Surgery Department, La Paz University Hospital, Madrid, Spain

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ABSTRACT

Objective: Ileocecal endometriosis is rare. Symptoms range from no symptoms, cramps, vomiting, to acute intestinal obstruction. Our objective was to review our cases, clarify, and resume its most appropriate management focusing on the factors to determine diagnosis. This is a retrospective study by revision of medical charts of all ileal endometriosis cases of our unit from 2006 to 2014.

Case Report: Seven cases were found; three (43%) had previous endometriosis laparoscopic diagnosis, four (57%) had partial bowel obstruction episodes, three (43%) had chronic pelvic pain, and one developed acute intestinal obstruction in postoperative ileostomy closure. In three (43%), the diagnosis was made with magnetic resonance imaging (MRI) and double contrast barium enema, in one (14%) only with MRI, and the other three (43%) during surgery. All patients underwent resection of the ileum and evolved favorably.

Conclusion: Variability in symptoms hinders diagnosis. The gold standard for diagnosis is MRI, but clinical suspicion optimizes imaging test diagnosis. Segmental resection should be indicated in the majority of the cases.

Introduction

Endometriosis is a chronic gynecological disease characterized by the presence of endometrial tissue, which lies outside the uterine cavity. It affects 10–15% of women of reproductive age [1].

The most frequent location is the ovary, followed by the Douglas cul-de-sac and the uterosacral ligaments [2]. The bowel is the most affected extragenital location (3–12%), 50–90% in the rectosigmoid junction; however, it can also affect the small bowel (2–16%), appendix (3–18%), and cecum (2–5%) [3].The ileum is affected in 4.1% patients [4].

The clinical features that patients with bowel endometriosis present add to the usual symptoms (cyclic pelvic pain, dysmenorrhea), others that are more specific of bowel involvement, such as rectal pain extended to the perineum (rectosigmoid location), which worsens with defecation, sitting, and especially during menstruation (52%), constipation, diarrhea, catamenial rectal bleeding (15–20%), and subocclusion symptoms (12%). Acute occlusion is rare [5]. When ileal involvement is added to other locations, symptoms overlap and the diagnosis can be omitted; in the cases where it is presented separately, unspecific symptoms (cramps, vomiting, abdominal distension) will require the patient to undergo numerous tests for differential diagnosis with other intestinal diseases, leading to frustrating results and inadequate treatment for many years [6].

The purpose of this study was to review our experience with regard to the ileal endometriosis focusing on the importance of clinical suspicion for the diagnosis.

Case report

After the Institutional Review Board approval, we retrospectively reviewed the medical charts of all ileal endometriosis cases operated and followed-up at the Endometriosis Unit of La Paz Universitary Hospital from 2006 to 2014.

During the study period, 150 patients were operated for symptomatic deep endometriosis at our center. Among these, 73 patients

* Corresponding author. Obstetrics and Gynecology Department, La Paz University Hospital, Paseo de la Castellana 261, 28046 Madrid, Spain.
E-mail address: analopezcarrasco.lopez@gmail.com (A. López Carrasco).

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had bowel endometriosis and only 7 patients had ileum involvement; their characteristics are given in Table 1.

Four (57%) patients had previous surgeries that allowed the diagnosis of endometriosis: one patient for endometrioma resection 4 years before; one patient for segmental resection of the rectum due to endometriosis 3 months before, and two patients had a diagnostic laparoscopy, one several months before the ileum resection (it allowed to establish diagnostic suspicion) and the other with rectal biopsy and cystectomy 9 years before.

Three (43%) patients presented with the main symptom episodes of catamenial intestinal pseudo-occlusion. One patient presented an acute bowel obstruction during the postoperative care from the closure of a prophylactic ileostomy. Four (57%) patients had chronic pelvic pain; one of them suffered two episodes of intestinal pseudo-occlusion short before the scheduled surgery.

In three (43%) patients, diagnosis was reached by magnetic resonance imaging (MRI; Figure 1) and double contrast barium enema (DCBE; Figure 2), in one (14%) patient just by MRI, and three (43%) patients were diagnosed during surgery. One patient with negative MRI for an ileum lesion from 6 months before and who had a scheduled focus surgery on the rectosigmoid suffered two episodes of pseudo-occlusion prior to the surgery; a re-evaluation of the MRI was requested from the radiology department without visualizing a lesion at that level.

The seven patients received hormonal treatment; Patients 1, 2, and 5 received combined oral contraceptives, Patients 3 and 7 got levonorgestrel intrauterine device, and patients 4 and 6 received GnRH analogs.

In all the patients, the surgery was performed in collaboration with the general surgery unit; surgery was performed by laparoscopy in five patients and by subumbilical midline laparotomy in two patients, obtaining histologic confirmation of the presence of endometriosis in the resected ileum segments in all the patients.

A resection of the ileum by end-to-end anastomosis was performed in all of them, and they all progressed toward recovery. Surgeries were performed by laparoscopy (single-port Olympus TriPort+; Figure 3) in Patients 1, 2, 3, 4, and 7 (71%). In the other two patients, the resection was performed by laparotomy due to excessive bowel distension; in Patient 5, emergency laparotomy was performed due to intestinal occlusion.

Three patients had deep lesions only in the ileum (43%). In two patients, adenomyosis was diagnosed by MRI (one of them had also several superficial peritoneal implants), and one had an ovarian endometrioma. Four patients had other deep lesions (57%), three of them (Patients 2, 5, and 7) in the rectosigmoid, two of which were resected during the same surgery, whereas in Patient 5, it was previously resected. In both patients, we performed a laparoscopic end-to-end anastomosis with CEEA-31 device. In Patient 2, we extracted the sectioned rectum through the colpectomy to perform the proximal section and place the anvil, while in Patient 5, this step was performed through a 4-cm suprapubic incision.

The painful symptoms of all patients improved significantly and the subocclusion symptoms were repeated in none of the patients. Only Patient 2 had surgical complications: a fistula due to dehiscence of the rectal anastomosis, which required a colostomy, leaving an ample vagina defect, and also a severe postoperative hemorrhage originating from a cervical artery that also required surgery. After a failed attempt of reconstruction, she continues with the ileostomy (probably permanent). Patient 1 is currently pregnant. Patients 2, 3, and 4 were treated with hormonal anticonceptives. Patient 5 received assisted reproductive treatment. Patient 6 received no treatment. Patient 7 underwent the hormone replacement therapy with estrogen and progestins.

Discussion

Infiltrating endometriosis affecting the terminal ileum is quite infrequent, accounting for 4.1% of all endometriosis cases that affect the bowel [4], which is 15–37% of the patients with pelvic endometriosis [7].

Indeed, since Melody [8] published the first case in 1956, multiple isolated cases [9,10] have appeared in the literature. Fedele

Table 1
Characteristics of the patients with an involvement of the ileum.

Patient	Age	Endometriosis previous diagnosis	Main symptom	Medical treatment	Diagnosis focus ileum	Associated lesions	Performed surgery	Via	ASRM
1	35	No	Catamenial pseudo-obstruction	Combined oral contraceptives	MR and DCBE	Adenomyosis (MR)	Ileum resection	SILS	I
2	30	LPSC	CPP	Combined oral contraceptives	LPSC (endometriosis)	Rectosigmoid, USL, and vagina	Ileum resection rectosigmoid resection partial colpectomy and USL resection	LPSC	IV
3	38	LPSC	CPP	LNG-IUD	MR and DCBE	Left USL and left ovarian endometrioma	Ovarian cystectomy + USL resection + ileum resection	LPSC	IV
4	31	No	Catamenial pseudo-obstruction	GnRH analogs	MR and DCBE	Small peritoneal implants and adenomyosis (MR)	Ileum and peritoneal implants resection	LPSC	I
5	34	LPSC	Bowel obstruction after closure of prophylactic ileostomy after LAR performed by endometriosis	Combined oral contraceptives	LAP (acute abdomen)	Rectosigmoid, USL and bilateral ovarian cyst	Ileum resection	LAP	IV
6	41	No	Catamenial pseudo-obstruction	GnRH analogs	MR	Right ovarian endometrioma	Right adnexectomy + myomectomy + ileum resection	LAP	IV
7	41	LPSC	CPP and 2 episodes pseudo-obstruction	LNG-IUD	LPSC	Rectosigmoid, USL, vagina and bilateral ovarian cystcs	HYS + partial colpectomy + Double adnexectomy + ileum and rectosigmoid resection	LPSC	IV

ASRM = American Society for reproductive medicine classification; CPP = chronic pelvic pain; DCBE = double-contrast barium enema; HYS = hysterectomy; SILS = single-incision laparoscopic surgery; LAP = laparotomy; LAR = low anterior resection; LNG-IUD = levonorgestrel intrauterine device; LPSC = laparoscopy; MR = magnetic resonance; USL = uterosacral ligaments.

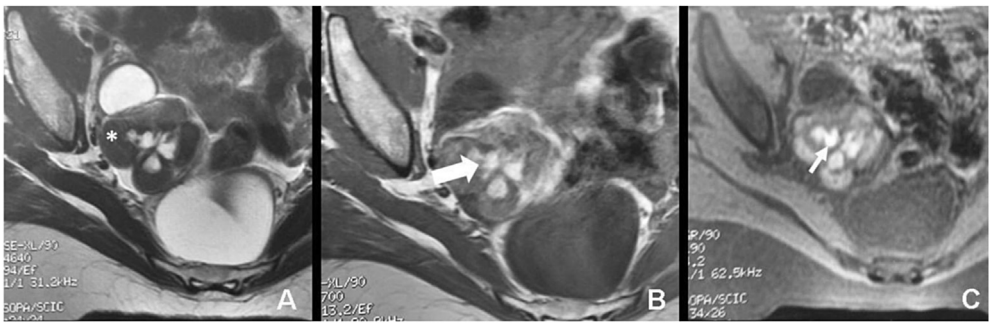

Figure 1. Different appearance of ileal endometriosis in (A) T2, (B) T1, and (C) T1-weighted images with (D) fat suppression. Pelvic ileum is thickened (*), with rounded images hyperintense in T2 and T1 that correspond to deep endometriosis foci (thick arrow). Axial T1-weighted image with fat suppression demonstrates improved contrast and conspicuity of the same lesions due to bloody content (thin arrow).

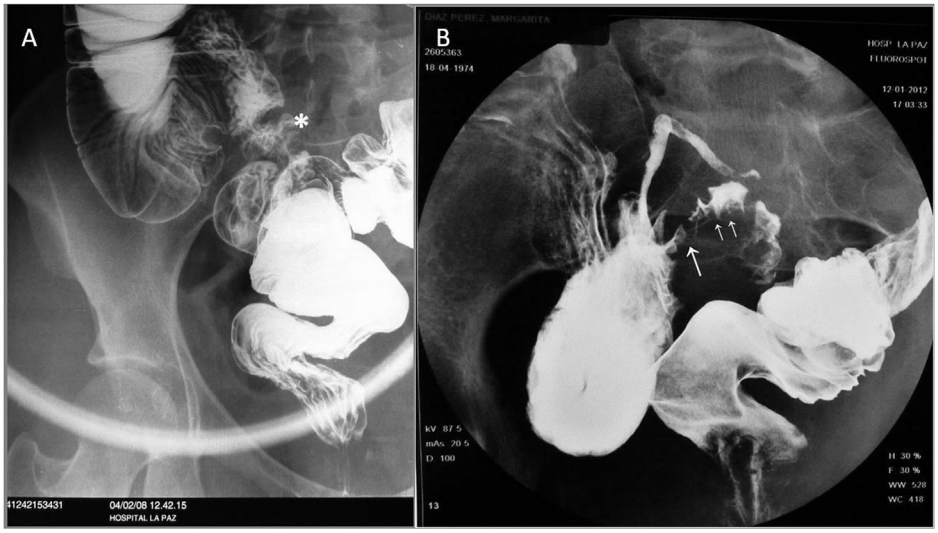

Figure 2. Double-contrast barium enema in two different patients with endometriosis of the ileum. (A) Lack of compliance and parietal spiculation of the mucosa in the mesenteric border of the ileum consistent with involvement by endometriosis. (B) In the mesenteric border of the ileum, digitiform images (small arrows) and filling defect (large arrow) due to involvement by endometriosis of the terminal ileum's wall.

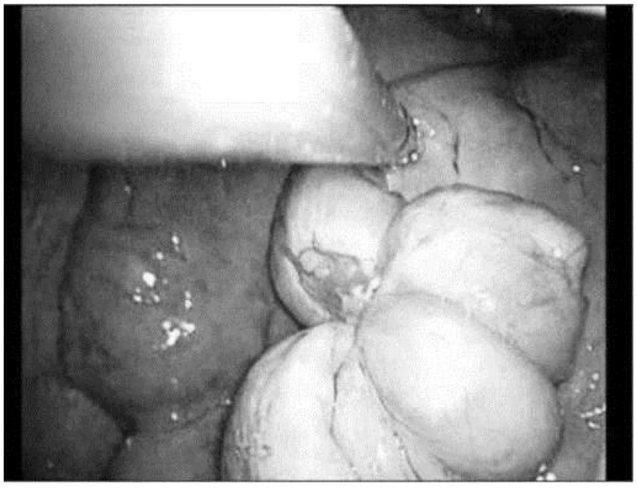

Figure 3. Laparoscopic vision of an implant of deep infiltrating endometriosis of the ileum (Patient 1).

et al [11] and Ruffo et al [12] published their case series of 8 and 31 cases, respectively.

In our hospital, 4% of the patients operated for deep infiltrating endometriosis had endometriosis of terminal ileum among 73 patients with bowel endometriosis. This coincides with what has been published previously [4].

As occurs in our series, in most cases, the disease in the terminal ileum is associated with other lesions [11,12]; therefore, the symptoms are related to their location but usually present associated with digestive clinical features [6]. Half of our patients presented episodes of pseudo-obstruction during menstruation associated with different dysmenorrhea; therefore, we agree with De Ceglie et al [13] regarding the way ileum endometriosis ought to be included in the differential diagnosis of pseudo-obstructive symptoms in patients at reproductive age. Three of our patients presented chronic pain; in one of them, diagnosis was possible using MRI and DCBE techniques, which were also useful for diagnosis in cases were clinical suspicion existed for pseudo-occlusion episodes. Although it is not a constant symptom, its absence must not exempt radiologists from searching for ileal endometriosis, especially when rectosigmoid endometriosis is found [14].

However, in two patients, ileocecal focus (both with chronic pelvic pain) was not diagnosed using MRI, and diagnosis was reached during a laparoscopy to perform an exeresis of a focus affecting the rectum; in one of them, the high clinical suspicion due to the two pseudo-occlusion episodes made us warn the patient of the high possibility of double resection. The association of several lesions requires the most accurate diagnosis to plan surgery properly and duly inform the patient about the surgical intention and its risks.

In the surgery of endometriosis, to reach our goal of removing all visible lesions, a careful exploration of the abdominal cavity is indispensable, especially when there exists already a diagnosis of rectosigmoid involvement, because lesions in the terminal ileum are frequently associated with them [12]. In spite of acknowledging this fact and having performed this inspection of the cavity, one of our patients debuted with an episode of acute bowel obstruction after the closure of a prophylactic ileostomy 3 months after performing a segmental resection of the rectum where an ileocecal lesion was not detected and had not been diagnosed by MRI either. A similar case was described by Dmowski et al [15]. This shows that the laparoscopic diagnosis is not easy and enables us to attribute, in some cases, the persistence of symptoms after surgery to this kind of lesions. Nonetheless, endometriosis of the terminal ileum does not frequently require emergency surgery [12].

Laparoscopy ought to be the technique of choice as it has demonstrated its viability and safety even in the double resection [12]. All our patients were using hormonal treatment to improve symptoms related to the endometriosis lesions or adenomyosis that were suspected. Nevertheless, finally they all needed surgery; therefore, we think that in the endometriosis of the ileum, probably the induction of the amenorrhea for the improvement of the symptoms is not enough, especially if obstructive symptoms exist. This way, in spite of the demonstrated efficacy of the hormonal therapy in the treatment of the symptoms in the deep endometriosis, sufficient improvement is not always obtained to avoid the surgery.

Clinical suspicion of ileal endometriosis is important to optimize the diagnosis by imaging tests. In these patients, surgical treatment was effective for the resolution of the symptoms, laparoscopy being the technique of choice. It is paramount to thoroughly search for lesions of ileal endometriosis during surgery, especially in patients with rectosigmoid involvement as they are frequently associated with other lesions and not detected by imaging tests.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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