

Appendiceal endometriosis as a rare cause of abdominal pain – a case report and literature review

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ABSTRACT: Endometriosis is an estrogen-dependent inflammatory disease, common in young women, characterized by the presence of endometrial tissue outside the uterine cavity. This ectopic endometrial tissue is most commonly found in the ovaries, peritoneum, uterosacral ligaments and rectovaginal cul-de-sac, with extremely rare involvement of the appendix. The main symptom is chronic abdominal pain, and the diagnosis is often made later, after the result of the histopathological examination. This study reports a 34-year-old patient complaining of chronic pelvic pain refractory to medical treatment, having undergone diagnostic laparotomy. During the surgery, we observed the presence of endometrioma fixed to the uterine wall, and the appendix was enlarged, but without evidence of inflammation. Endometrioma resection and appendectomy were performed, with good postoperative recovery. The anatomopathological exam showed endometriosis in the cecal appendix.

Keywords: endometriosis; gastrointestinal tract; appendix; abdominal pain; appendectomy.

RESUMO: Endometriose é uma doença inflamatória estrogênio-dependente frequente em mulheres jovens, caracterizada pela presença de tecido endometrial fora da cavidade uterina. Esse tecido ectópico de endométrio é mais comumente encontrado nos ovários, peritônio, ligamentos uterossacros e fundo de saco retovaginal, sendo o acometimento do apêndice cecal extremamente raro. O quadro clínico predominante é o de dor abdominal crônica, sendo muitas vezes o diagnóstico feito posteriormente, após o resultado do anatomopatológico. Relatamos o caso de uma paciente de 34 anos com queixa de dor pélvica crônica, refratária ao tratamento clínico, tendo sido submetida à laparotomia exploradora diagnóstica. Durante o ato cirúrgico, observamos a presença de endometrioma fixo à parede uterina, bem como apêndice cecal aumentado de volume, porém sem evidência de sinais flogísticos. Procedeu-se à ressecção do endometrioma e apendicectomia, com boa evolução pós-operatória. O resultado do exame anatomopatológico revelou a presença de endometriose no apêndice cecal.

Palavras-chave: endometriose; trato gastrointestinal; apêndice; dor abdominal; apendicectomia.

INTRODUCTION

Endometriosis is an estrogen-dependent inflammatory disease that affects 5 to 10% of women at reproductive age¹, characterized by the presence of endometrial tissue outside the uterine cavity²⁻⁶. It is most commonly found in the peritoneum and the ovaries^{5,7}.

It affects the gastrointestinal tract (GIT) around 12% of the cases⁴, but the involvement of the cecal appendix is rare^{4,6,7-10}.

When the appendix is affected, symptoms may vary from acute abdominal pain (simulating appendicitis)^{3,4}, to chronic (significant reduction in the quality of life)^{2,5,7,9-11} or even asymptomatic abdominal pain^{3,5}.

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Regarding the diagnostic investigation, endorectal ultrasound and laparoscopy can add important information when patients present with unexplainable abdominal pain^{6,9,12}. Typical treatment consists of suppressed ovulation and, if required, surgical excision^{1,7}.

In cases of appendix involvement, intraoperative inspection may not show any alteration that suggests endometriosis^{3,9,11}. Then, the definitive diagnosis is confirmed through anatomopathological analysis^{3,6,7,9,11,12}.

Some authors describe patients with abdominal pain who, after undergoing appendectomy, presented improvements and their abdominal symptoms disappeared^{3,4,6,9-11,13}. However, incidental appendectomy during the surgical treatment of pelvic endometriosis is a controversial theme⁹.

Based on these considerations, this study reports a rare case of appendiceal endometriosis in a woman with chronic pelvic pain.

CASE REPORT

A 34-year-old female patient, with history of chronic pelvic pain related to the menstrual period for one year, with worsening of symptoms in the last four months. She came to our service with intense pain in hypogastric region and right iliac fossa for seven days.

At the physical examination, she had regular general health, normal vital signs, no fever, flat abdomen, presence of bowel sounds, pain at deep palpation of the hypogastrium and right iliac fossa and absence of peritoneal signs.

At vaginal exam, the patient had anteverted uterus, fibroelastic colon, intense pain at uterine mobilization and at palpation of posterior and right lateral cul-de-sac; non-palpable attachments. She was submitted to abdominal ultrasound, which showed normal attachments, normal size of the uterus, with a small quantity of free fluid in the cavity.

Based on that, the treatment choice was hospitalization and conservative treatment with analgesics, anti-inflammatory medication and intravenous hydration. After two days, the pain increased and, although the patient had no peritoneal signs, exploratory laparotomy was performed. The findings were: presence of a small amount of blood in the pelvic region, endometrioma measuring around 1.0 cm in diameter fixed to the lateral wall of the uterus and enlarged cecal appendix, but no signs of inflammation. Endometrioma resection and appendectomy were performed.

The patient did well postoperatively, and was discharged from the hospital without symptoms on the second day after the surgery. The anatomopathological exam concluded that there was an endometrioma of the uterus and appendiceal endometriosis (Figure).

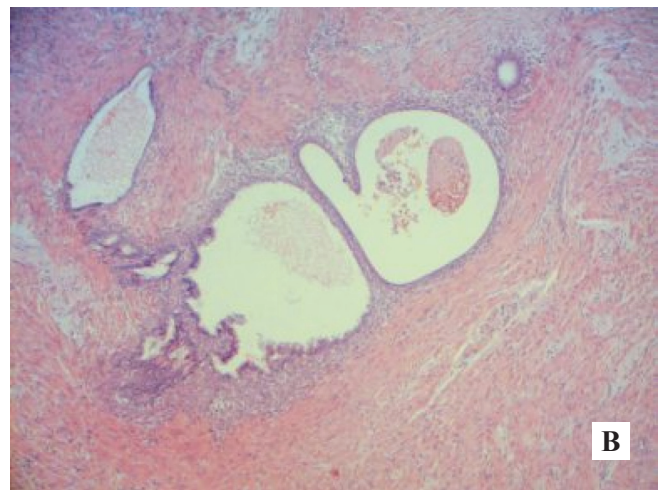
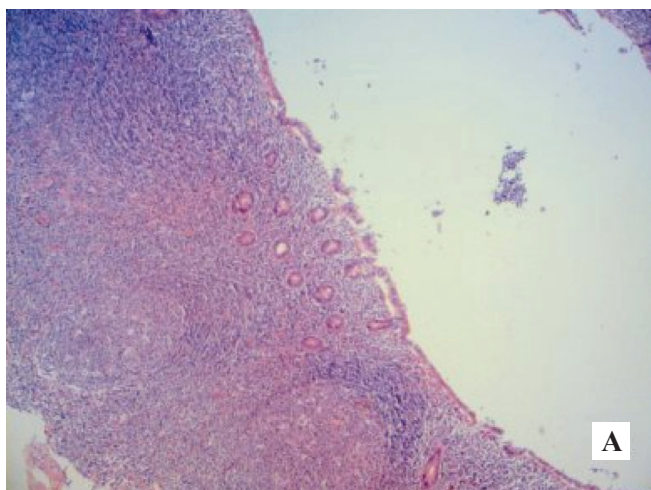


Figure. Photomicrograph of cecal appendix. (A) Mucosa of the appendix without alterations. (B) Presence of endometrial tissue in the middle of the tunica muscularis of the appendicular wall (hematoxylin and eosin staining, enlarged 40x).

DISCUSSION

Endometriosis is defined as the ectopic presence of endometrial tissue outside the uterine cavity²⁻⁶. There is no consensus regarding its histological origin^{1,4,5}. It may be inherited, as the disease prevalence in women related to affected females is seven times greater if compared to women without family history¹. Histologically, endometriosis is characterized by the ectopic presence of an endometrial stroma, chronic hemorrhage and signs of inflammation, and it may occur separately or in combination with other affections^{1,7}. This inflammation may cause pelvic nerve damage and consequent pain¹. It is usually found during explorations in the abdominal cavity due to pelvic pain, pelvic mass or infertility⁷.

Prevalence rates vary according to the investigated population, ranging from 0.7 to 45% in asymptomatic women; 20 to 40% in infertile women; 6 to 18% in women submitted to sterilization and 15 to 70% in patients with chronic abdominal pain¹⁴. In general, the disease affects around 5 to 10% of women, with annual incidence of 1.9 cases among 1,000 women, and aged between 15 and 49 years old^{1,14}. Studies showed that Afro-American women present lower incidence of the disease when compared to Caucasian American women. On the other hand, the disease seems to affect more Asian women than Caucasian women¹⁴.

Although the disease traditionally involves pelvic organs, its location and extension may vary considerably. The occurrence outside the genital tract is named extragenital endometriosis, and it may affect surgical scars, bladder, heart, pulmonary pleura, diaphragm and GIT^{5,7}.

In the GIT, the main affected site is the rectum and the sigmoid colon (95%)¹². The involvement of the cecal appendix is rare^{3,4,6-10}, varying from 0.8 to 20%^{10,15}. In anatomopathological studies of appendectomies, the incidence of endometriosis is low (varying from 0.15 to 1%)^{4,7,16}.

Most patients with cecal appendix affected by endometriosis are asymptomatic^{3,5}. When symptomatic, the predominant symptom is chronic abdominal pain^{5,7,9-11} and, in few cases, it involves acute abdominal pain^{3,4}. Appendiceal endometriosis may occasionally appear as appendicitis^{4,7}, mucocele^{10,13}, local

peritoneal pseudomyxoma¹⁰, intussusception⁵, perforation^{4,10} and intestinal bleeding³.

The patients rarely present with acute appendicitis^{4,7}. Appendiceal endometriosis corresponds to less than 1% of the causes of acute appendicitis⁷. In a study that analyzed the cecal appendix histology of 1,225 patients submitted to surgical treatment with clinical diagnosis of acute appendicitis, only 0.25% of the analyzed specimens presented microscopic diagnosis of endometriosis¹⁷.

Thus, these findings agree with results of other studies that appendiceal endometriosis is more related to the diagnosis of chronic pelvic pain^{2,3,5-7,9-11,15} usually associated with pelvic endometriosis^{3,11}, and not to acute pain^{4,7}.

Therefore, the appendix is an important organ in the evaluation of non-diagnosed chronic pelvic pain^{7,6,11}. However, only 41% of the patients with appendiceal endometriosis complain of intermittent pain in the right lower quadrant⁷, which may – or may not – be related to menstruation^{2,3}.

Preoperative diagnosis of appendiceal endometriosis is uncommon^{3,18}. It is frequently found during the surgical treatment for pelvic endometriosis, similar to our case reported^{4,13,10}.

In our experience, the intraoperative inspection of the appendix did not show any sign suggesting the diagnosis, which was confirmed through microscopic exam, a fact also reported by other authors^{3,9,11}. Therefore, the definitive diagnosis is obtained through the anatomopathological study^{3,6,7,9,11}. The Table shows the studies in which the patients complained of abdominal pain, but endometriosis was not the preoperative suspicion, and it was diagnosed through the anatomopathological study.

The typical treatment of pain is a combination of suppressed ovulation and surgery^{1,7}. However, performing appendectomy in a macroscopically non-affected appendix during the surgical treatment for pelvic endometriosis is a controversial issue⁹.

Some authors justify the prescription of appendectomy to patients with chronic abdominal pain with undefined origin, even when the organ aspect is normal^{7,12}. Nisolle et al. favors organ resection when it is rigid as a result of deep infiltrating endometriosis¹⁵. The same author does not indicate prophylactic appendectomy to all patients submitted to laparoscopy

Table. Analysis comparing the case reports of appendiceal endometriosis.

Authors	Age (years)	Abdominal pain (time)	Associations	Supporting exams	Cecal appendix	
					Macroscopic aspect	Histopathological exam
Khoo et al. ³	40	5 days	Infertility; Fever: 37.9°C	TLC: 15,400/ mm ³	Congested	Endometriosis
Hasegawa et al. ⁴	35	3 days	Signs of peritonitis; Fever: 38.2°C	TLC: 12,500/mm ³ ; CT: Liquid in cavity	Perforated and adhered to the retroperitoneum	Acute appendicitis and endometriosis
Ljaz et al. ⁵	40	Several months	–	Colonoscopy: polyp in cecal region	Intussusception	Endometriosis
Krairy ⁶	33	10 days	–	Normal	Laparoscopy: appendicular mass	Endometriosis
Tumay et al. ⁷	24	2 years	–	TLC: 23,300/mm ³	Edematous covered by focal grey nodules with hemorrhagic areas	Endometriosis
Al Oulaqi et al. ⁹	25	15 months	Infertility	Normal	Normal serous	Endometriosis
Driman et al. ¹⁰	34	1 year	–	CT: paracecal mass	Perforated; Mucocele	Endometriosis
	31	Chronic	Infertility	–	Edematous; Mucocele	Endometriosis
Tez et al. ¹¹	42	2 months	Fever: 38.1°C	TLC: 14,500/mm ³ ; CT: cecal mass	Congested	Endometriosis
Chang-Hun et al. ¹³	42	4 months	Pelvic endometriosis	CT: periappendicular abscess	Loss of appendicular outline; Mucocele	Endometriosis

TLC: total leucocyte count; US: ultrasound.

due to chronic abdominal pain given the low probability of endometriosis in this organ¹⁵.

In our case, because of the presence of pelvic endometriosis combined with an enlarged appendix, the choice was appendectomy, with complete suppression of abdominal symptoms after the surgery, a fact also reported by other authors^{3,4,6,9-11,13}.

We concluded that endometriosis of the cecal appendix is rare and almost never diagnosed before the surgery, with the definitive diagnosis obtained through microscopic exam. However, it should always be taken into account for the diagnosis of chronic pelvic pain, especially in young women complaining of recurrent pain, history of infertility and pelvic endometriosis.

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