

CASE REPORT

Ovarian involvement in Crohn's disease: A rare complication

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ABSTRACT

Background: The transmural condition of Crohn's disease predisposes to fistulae or abscesses. The internal fistula incidence is about 15%. Among them, enteroovarian fistula is rarely described on the literature. Herein, the authors present three cases of enteroovarian fistulas.

Case reports: Two women are diagnosed with ileal Crohn's disease that presented a pelvic abscess diagnosed by ultrasound and CT. On surgery, an inflammatory mass involving the ileum and the ovary was found. The third woman was operated because of a tuboovarian abscess and was diagnosed with ileal Crohn's disease afterwards. In the three cases, the histopathological analysis of the ovary showed granulomas with abscess compatible with Crohn's disease. In one of the cases, multinucleated giant cells were found in the foreign body reaction to vegetable matter. A right ileocelectomy and an adnexectomy were performed in all three cases. No further involvement of the contralateral ovary or other gynaecological complications was observed.

Discussion: The treatment of Crohn's disease complications should be individualized. In the case of ovarian involvement, surgical treatment should include adnexectomy.

Key words: Crohn's disease. Enterovarian fistula. Fistula.

INTRODUCTION

Crohn's disease is characterized by a chronic inflammatory process with different evolutive patterns: inflammatory, stricturing or penetrating (1). Being a transmural condition, it results in penetrating injuries, such as fistulae, phlegmons or abscesses.

Fistulae incidence varies between 17 and 50% of the cases, being most of them perianal (2). Moreover, internal fistulae occur in about 15% of the cases. The main types are enteroenteric, enterocutaneous, enteroabdominal, enterovaginal and enterovesical (2,3). However, entero-

ovarian fistulas are rare. Only very few cases are reported on the literature (4).

In this paper, we will present three cases, with emphasis on the diagnosis and management.

CASE REPORTS

Case report 1

A 31-year-old woman was diagnosed with ileal Crohn's disease and initially treated with corticosteroids and 5-ASA. She was admitted five weeks later with abdominal pain, fever and a tender mass in right iliac fossa. Laboratory results showed leukocytosis and elevated CRP. Ultrasound examination revealed an inflammatory mass with an associated abscess and a terminal ileum wall thickening.

In elective surgery, an inflammatory mass constituted by the terminal ileum, and the right parametrium and ovary, was found. It had associated 7 cm purulent collection. Thus, a right ileocelectomy and adnexectomy were performed. She was discharged with Azathioprine.

Histopathological features were: 14 cm segment of the terminal ileum with thickened wall, transmural chronic active inflammation with ulceration, edema and fibrosis and non-specific lymphoid hyperplasia. The ovary showed xanthogranulomatous inflammation with abscessification and multinucleated giant cell foreign body reaction to vegetable matter, suggesting direct fistulization from the bowel. The right fallopian tube was strongly adhered to the ovary.

After three years, a disease recurrence appeared at the preanastomotic ileum segment. She continued the treatment with azathioprine and adalimumab.

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Because of the subocclusive symptoms, the patient underwent a new ileocolic resection twelve years after the first surgery. She had many postoperative complications because of an anastomotic leak and an intestinal fistula requiring a new resection.

The patient is currently well and asymptomatic, treated with azathioprine.

Case report 2

A 31-year-old woman was diagnosed with ileal Crohn's disease. Initially she was treated with budesonide and 5-ASA. During the following four years she had several relapses meanwhile she was using antibiotics and corticosteroids. Then, she had azathioprine as maintenance treatment.

After this, the patient presented again postprandial abdominal cramps, occasional fever and pain on the right iliac fossa. Colonoscopy showed an ileum stenosis. Digital rectal exam revealed a painful mass in the pouch of Douglas. Ultrasound showed a pelvic inflammatory mass including the right ovary with gas bubbles inside (Fig. 1). CT displayed a 4 cm inflammatory mass in the pouch of Douglas with internal gas that could not be seen separately from the right ovary and it was adjacent to the ileum (Fig. 2).

On surgical examination, an inflammatory mass containing cecum, terminal ileum, right ovary and fallopian tube was found. A small amount of pus was removed on debridement and a fistulous tract between terminal ileum and right ovary was observed. A right ileocelectomy and adnexectomy were performed.

Histopathology reported Crohn's disease involving the terminal ileum with fissures and abscesses. Some lymph nodes contained granulomas. The ovary and fallopian tube was affected seriously with abscesses and granulomatous inflammation by local spread of Crohn's disease.

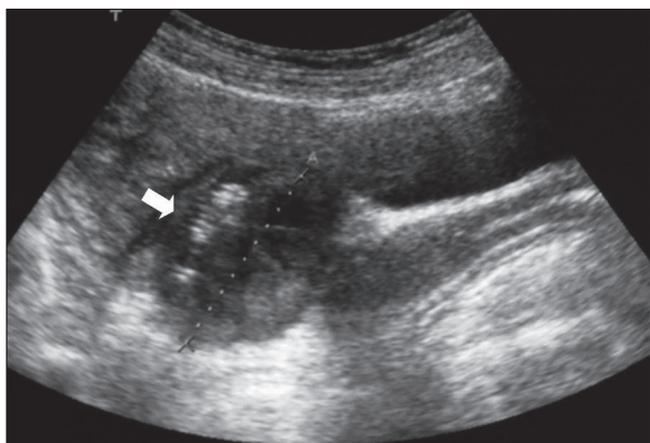


Fig. 1. Ultrasonography. Right ovary with internal gas.

Since surgery, the patient has been treated with azathioprine. After seven years, she is still asymptomatic.

Case report 3

A 25-year-old woman, without significant history, was admitted to another hospital with pelvic pain, fever, leukocytosis and elevated CPR. Ultrasound examination revealed a right tubo-ovarian abscess. On emergency surgery she presented a right ovarian abscess with involvement of an ileal loop that showed edematous change. This was easily manually released, and a right salpingo-oophorectomy was performed.

Histopathology analysis showed a 5 cm right ovary with a 2 cm cystic structure with ragged inner wall surface. The fallopian tube of 7x1.5 cm was edematous. The right ovary and Fallopian tube showed granulomata with abscess formation, which were negative to PAS and Ziehl-Neelsen stain techniques and compatible with Crohn's disease.

Seven days after surgery, the patient was admitted to our hospital with a painful mass in the right iliac fossa. Laboratory results showed leukocytosis and CRP levels of 100; ultrasound and CT revealed wall thickening in distal ileum and cecum, suitable with Crohn's disease, with no associated collection. She was treated with antibiotics, corticosteroids, 5-ASA and parenteral nutrition. She was discharged well after ten days.

Three months later, she was readmitted to surgery ward for fever and a painful mass in the right iliac fossa. Neutrophilia and leukocytosis were found. Ultrasound examination showed a collection in the right iliac fossa adjacent to the rectus abdominis that extended to the thickened loop of ileum and the mesentery. The patient underwent elective surgery, in which an inflammatory plastron of terminal ileum fistulized to abdominal wall



Fig. 2. TC. Pelvic inflammatory mass with internal gas (arrow) and adjacent ileum (arrowhead).

and to an ileal loop was found, with no intra-abdominal abscess. A right ileocelectomy was performed and the abdominal wall fistula was resected. Postoperative evolution was satisfactory.

Histopathology of 60 cm segment of terminal ileum described adherent loops of bowel, with areas of ulcerated mucosa and thickened wall. In addition, there was a granulomatous inflammation with abscess formation and fissures compatible with Crohn's disease. The resected abdominal wall fistula showed areas of abscess and granulomatous changes, which were also consistent with Crohn's disease.

The patient remained asymptomatic for four years when she presented a recurrence in the preanastomotic ileum. Then, a treatment with azathioprine was prescribed. She remains symptom-free.

In all three cases, right adnexectomy plus ileocelectomy have been successful treatments, without further involvement of the contralateral ovary or other gynecological complications.

DISCUSSION

Gynecological disorders are common in patients with Crohn's disease. Therefore, abdominopelvic pain in women can be attributed to gynecological pathologies, such as menstrual disorders, pelvic inflammatory diseases (as seen in the third case), endometriosis or ovarian pathologies. This occurs when inflammatory bowel disease has not been previously diagnosed due to similar symptoms (5-7). It should be noted, that evolution of fistulizing Crohn's disease can affect any abdominal organ. Therefore, any complication can involve the female genital tract due to the proximity of the ileum, colon and rectum. Fistulae to uterus, adnexa, vulva and perineum have been described (6,8,9). Granulomatous affectation of the ovaries is rare, unless it results from fistulization (4,10) from the bowel, as seen with the first two patients, or the extension of the process by contiguity (11), as described in the third case.

Diagnostic methods will depend on their availability and the clinical status of patients. CT and MRI are the standard methods to assess the extent and severity of the disease. Moreover, internal fistulae can be diagnosed with highly accuracy with them (1,12). Although ultrasonography (1) possess less precision, it is also useful with a sensitivity of 87% and 100% for the detection of fistulas and abscesses, respectively.

Preoperative diagnosis of fistulae is rare (3). The most likely diagnosis is a plastron or an abscess, as happened in the first case, in which ovarian involvement was not expected to appear on ultrasound. On the second case, CT examination showed a plastron with pelvic abscess. However, ultrasound confirmed ovarian involvement. In the third case, this complication was only demonstrated surgically and after histopathological analysis.

Medical and surgical treatment for both internal and external fistulae in Crohn's disease remains a matter of discussion (2) because there are no diagnostic methods for a reliable assessment of healing. Furthermore, the healing rate varies significantly due to the heterogeneity of the patients, the different drugs, the surgical techniques used and the definitions on the criteria of healing.

Management of internal fistulae was surgical until two decades ago. However, the introduction of new drugs and immuno- and biological therapies, have caused an increase of a conservative treatment (2,3). This is the case when the fistula is asymptomatic or symptoms are only mild and temporary.

Surgery is effective for enterovesical and enteroenteric fistulas, with a high healing rate, despite being burdened by a high morbidity rate (2). Nevertheless, the effectiveness of surgery is lower for anovaginal or rectovaginal fistulae (3,13).

The abscess resulting from an intestinal fistula implies that medical treatment has only relative and short duration effectiveness. The fistula often reactivates when treatment stops. Consequently, a surgical treatment should be performed with an abscess (2,3,13).

In the first two cases presented here, there were pelvic abscesses due to fistulae of ileal loops affected by Crohn's disease, with involvement of the right ovaries, which also showed abscesses and typical granulomas, even containing vegetal fibers in one of the cases as histopathological examination proved. This is the reason why the most suitable therapeutic option in the two cases was surgical treatment, overcoming the abdominal abscess and the fistula problems by resecting the affected intestinal segment, as well as the ovary involvement by an adnexectomy, as described in the few cases in literature (4,10,11). Avoiding oophorectomy should only be considered if the ovary has not been affected by the inflammatory mass (4).

In the third case, adnexectomy was justified because there was an ovarian abscess, although Crohn's disease was not suspected at the time of excision. However, even when typical lesions such as abscesses and non-caseating granulomas are present, the differential diagnosis of ovarian affectation by tuberculosis, actinomycosis or fungal infection must be considered (14). Then, the suspicion of Crohn's disease with usual methods should be confirmed.

In conclusion, the treatment of complications of Crohn's disease should be individualized depending on the type of complication, the extent of the disease and the concerned structures. In the case of ovarian involvement by Crohn's disease, surgical treatment should include salpingo-oophorectomy.

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