Case report / *Prikaz primera*

Ileo-salpingeal Fistula with Ovarian Abscess in Crohn's Disease

Ileo-salpingealna fistula z ovarijskim abscesom pri Crohnovi bolezni

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***Ključne besede**: vnetna črevesna bolezen, Crohnova bolezen, ileo-salpingealna fistula, ovarijski absces*

Abbreviations used in this paper: *CD, Crohn's disease; IBD, Inflammatory Bowel Disease; TNF-α, tumour necrosis factor alpha; UC, Ulcerative Colitis; TB, Tuberculosis; ITB, Intestinal Tuberculosis; MRI, Magnetic Resonance Imaging; CT, Computed Tomography; IUS, Intestinal Ultrasound.*

Conflicts of interest

All authors have none to declare.

ABSTRACT

Introduction

The clinical presentation of Crohn's disease (CD) depends significantly on the location and severity of inflammation. While fistulising forms of CD affect 17–50% of patients, involvement of gynaecological

IZVLEČEK

Klinična slika Crohnove bolezni (CB) je močno odvisna od lokacije in resnosti vnetja. Medtem ko fistulizirajoče oblike CD prizadenejo 17–50 % bolnic, je prizadetost ginekoloških struktur – kar lahko povzroči medenične ciste, mase ali abscese – redka. V literaturi so poročali o manj kot 20 primerih granu-

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structures – which may lead to pelvic cysts, masses, or abscesses – is rare. Fewer than 20 cases of granulomatous ovarian disease and only 5 cases of enterosalpingeal fistulisation have been reported in the literature.

A 26-year-old female was admitted with right lower abdominal pain. During hospitalisation, a colonoscopy was performed, which revealed no pathological changes in the colonic mucosa, but ileal stenosis was confirmed. A biopsy sample from the altered ileal mucosa could not be obtained. CT enterography showed terminal ileitis with luminal stenosis, along with suspected inter-intestinal fistulas and abscesses. Pelvic MRI indicated ileal thickening, an abscess in the right ovary, and pelvic fluid collections. The patient was taken to the operating room, where a fistulising lesion of the terminal ileum involving the right fallopian tube and an abscess of the right ovary was identified.

If a patient with CD develops a pelvic abscess, TNF-α inhibitors should be avoided, and appropriate antibiotics should be initiated. Surgical intervention or drainage may be necessary. The management of fistulas is typically surgical, but oophorectomy can be avoided if there is no fistulisation to the ovary.

lomatozne bolezni jajčnikov in samo o 5 primerih enterosalpingealne fistulizacije.

26-letna ženska je bila sprejeta z bolečino v desnem spodnjem delu trebuha. Med hospitalizacijo je bila opravljena kolonoskopija, ki ni pokazala patoloških sprememb na sluznici debelega črevesa, potrdila pa je stenozo ileuma. Biopsijskega vzorca iz spremenjene sluznice ileuma ni bilo mogoče dobiti. CT enterografija je pokazala terminalni ileitis z luminalno stenozo, skupaj s sumom na medčrevesne fistule in abscese. MRI medenice je pokazala zadebelitev ileuma, absces v desnem jajčniku in zbiranje medenične tekočine. Bolnico so odpeljali v operacijsko sobo, kjer so ugotovili fistulizirajočo lezijo terminalnega ileuma, ki vključuje desni jajcevod, in absces desnega jajčnika. Če se pri bolniku s CD razvije medenični absces, se je treba izogibati zaviralcem TNF-α in uvesti ustrezne antibiotike. Morda bo potreben kirurški poseg ali drenaža.

Zdravljenje fistul je običajno kirurško, a se je mogoče izogniti ooforektomiji, če ni fistulizacije jajčnika.

INTRODUCTION

The CD is a chronic inflammatory condition that can affect any region of the gastrointestinal tract, from the mouth to the anus, though it most commonly presents in the ileum and colon. Together with ulcerative colitis (UC), CD is classified as an inflammatory bowel disease (IBD). Although the precise cause remains unclear, the onset of CD is believed to result from the interplay between genetic susceptibility, environmental factors, and interactions with the gut microbiota (1, 2).

The clinical presentation of CD depends significantly on the location and severity of the inflammation. Symptoms and signs can vary widely, ranging from anorexia, abdominal pain, and fatigue to more severe manifestations such as rectal bleeding, bloody diarrhoea, and the development of perianal lesions like ulcerations and fistulas (3). The incidence of fistulas varies from 17% to 50%, with most being perianal (2). Internal fistulas occur in approximately 15% of cases, with the main types being enteroenteric, enterocutaneous, enteroabdominal, enterovaginal, and enterovesical (2, 3).

The differential diagnosis between intestinal tuberculosis (ITB) and CD is crucial, as misdiagnosis can lead to fatal outcomes. Both CD and ITB can result in fistulisation to adjacent organs, including other segments of the small or large bowel, the urinary bladder, the vagina, skin, as well as the stomach and duode-

num (4–6). While fistulising disease in CD affects 17–50% of patients, involvement of gynecologic structures – which may lead to pelvic cysts, masses, or abscesses – is rare. Fewer than 20 cases of granulomatous ovarian disease and 5 cases of enterosalpingial fistulising disease have been reported in the literature. We present a surgically treated case of CD with a tubo-ovarian abscess and ileo-salpingeal fistula (7).

CASE REPORT

A 26-year-old female was admitted to the hospital with right lower abdominal pain. Four months prior, she had visited a gastroenterologist for the first time due to the same symptoms – abdominal pain, diarrhoea, and weight loss. Initial laboratory tests showed elevated leukocytes and C-reactive protein (CRP). A colonoscopy was performed, which revealed no significant pathological changes in the colonic mucosa, and it was not possible to enter the ileum due to suspected ileal stenosis. Computed Tomography (CT) enterography showed terminal ileitis. Faecal calprotectin levels were elevated, while the Quantiferon test and stool analysis for tuberculosis (TB) were negative. Antibiotics and budesonide were prescribed, but the patient was lost to follow-up. She also underwent a gynaecological examination due to a CT-verified ovarian cyst. Four months later, she was readmitted to the hospital with right lower abdominal pain, diarrhoea, and elevated leukocytes and CRP. Although she had felt some improvement on the prescribed therapy, she missed the suggested follow-up gastroenterological examinations. During this time, she was followed by a gynaecologist, who did not recommend any additional diagnostic procedures, aside from an ultrasound. Two months later, the patient discontinued budesonide on her initiative. Upon hospitalisation, a colonoscopy was performed again. It showed no significant pathological changes in the colonic mucosa, and a biopsy of the altered ileal mucosa could not be obtained due to stenosis. CT enterography revealed terminal ileitis with luminal stenosis and suspected inter-intestinal fistulas and abscesses. (Figure 1).

Figure 1. CT enterography: terminal ileitis with luminal stenosis and suspected inter-intestinal fistula and abscesses

The faecal calprotectin levels were elevated again. Due to the inability to obtain pathohistological confirmation of CD endoscopically – which is crucial before initiating specific biologic therapy – and the presence of suspicious abscesses, a surgeon was consulted, who recommended surgical treatment. Before surgery, a pelvic Magnetic Resonance Imaging (MRI) was performed, which revealed ileal thickening, an abscess in the right ovary, and pelvic fluid collections (Figure 2). The patient was taken to the operating room, where terminal ileum disease was found to be fistulising to the right fallopian tube, with an abscess of the right ovary. Right salpingectomy, ovariectomy, ileocecectomy, ileocolic anastomosis, and repair of the enterotubal fistula were performed.

The pathohistological diagnosis confirmed IBD as the primary diagnosis, with CD identified as the specific subtype. Additionally, a tubo-ovarian abscess with granulomatous inflammation of the right ovary was found, as a complication of the fistulising form of CD. Reactive lymphadenopathy was also noted in the intramural and mesenteric lymph nodes.

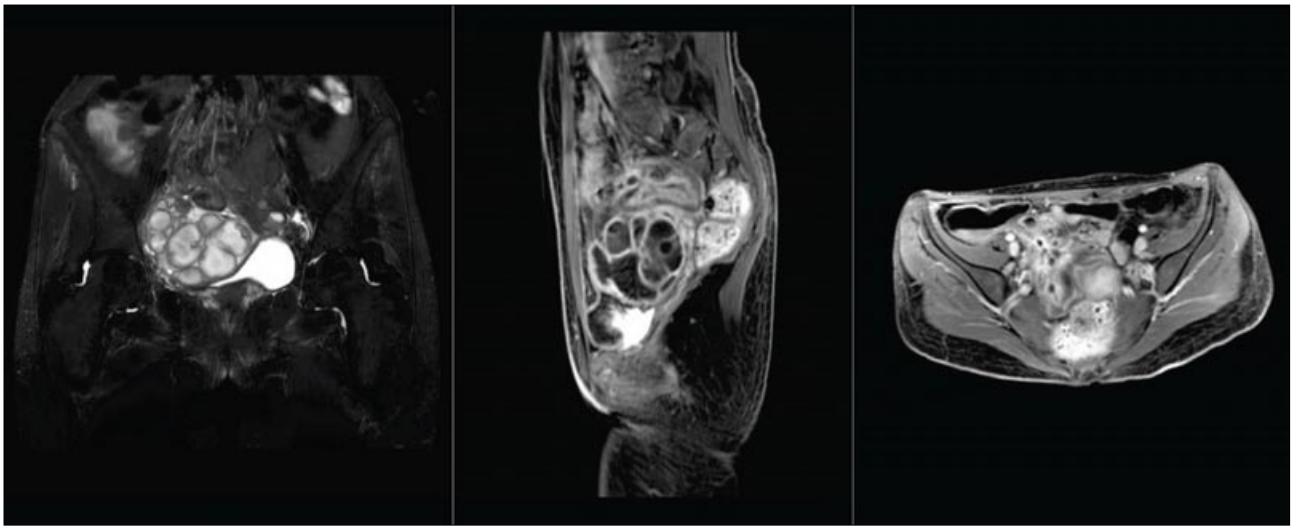

Figure 2. MR pelvis: Ileal thickening, right ovary abscess and pelvic fluid collections

DISCUSSION

Patients with CD frequently experience gynaecological disorders. Therefore, abdominal or pelvic pain in women may be attributed to gynaecological conditions such as menstrual disorders, pelvic inflammatory disease, endometriosis, or ovarian pathologies.

In this case report, we present a rare complication of CD—ovarian involvement, likely due to direct fistulisation between the bowel and the fallopian tube, leading to the formation of an ovarian abscess. Similar cases have been reported by authors from the United States, Canada, the United Kingdom, Europe, Tunisia, and Japan (7–10). The choice of diagnostic methods depends on the availability of resources and the patient's medical status. In a systematic review of the diagnosis of intra-abdominal fistulising lesions, cross-sectional imaging demonstrated the following accuracies: for CT (with surgery and endoscopy as reference standards), the sensitivity was 70% and specificity 97%; for MRI (with surgery or endoscopy as reference standards), the sensitivity was 76% and specificity 96%; and for intestinal ultrasound (IUS) (with surgery, barium studies, and colonoscopy as reference standards), the sensitivity was 74% and specificity 95% (10). For detecting intra-abdominal or pelvic fistulae, CT and MRI are preferred over ultrasound, with the added benefit of no radiation exposure with MRI enterography (11). In our case, after a colonoscopy, which revealed stenotic ileum, MRI enterography was performed and showed ileal inflammation, stenosis, and inter-intestinal abscesses. Before surgery, a pelvic MRI was also performed, which described an ovarian abscess. Fistula formation in CD is a complex condition to manage and treatment must be tailored to the various forms of the disease. Biologic therapy is the most well-documented treatment for fistulising CD, and it is often administered in higher doses in contrast to luminal CD. Antibiotics may be beneficial for a short duration and should be combined with biologics or immunomodulators to enhance treatment response (12). Medical therapy alone is rarely sufficient, but it can be initiated before surgery and continued after the surgical intervention. In most cases a combination of surgery and medical therapy provides the best outcomes; however, the ideal treatment strategy has not been definitively established (13). For ovarian involvement in CD, surgical treatment is generally indicated.

In the presented case, pelvic abscesses were caused by fistulisation of ileal loops affected by CD, with involvement of the right ovary, which also showed abscesses and typical granulomas, as confirmed by histopathological examination. This finding led to the decision to proceed with surgical treatment, addressing both the abdominal abscess and the fistulae by resecting the affected intestinal segment. Ovarian involvement was managed with adnexectomy, as described

in a few cases in the literature (7, 9, 10). Oophorectomy should be considered only if the inflammatory mass significantly affects the ovary (7).

CONCLUSION

CD can clinically and radiographically mimic gynaecological conditions such as endometriosis, ovarian cysts, and pelvic inflammatory disease. Gynaecologic involvement in CD should be strongly considered when ipsilateral bowel wall abnormalities are present. A multidisciplinary approach involving gastroenterologists, surgeons, and radiologists is essential for optimal patient care.

If a patient with CD develops a pelvic abscess, TNF-α inhibitors should be avoided, and appropriate antibiotics should be initiated. Surgery or drainage may be necessary, depending on the severity. Managing the fistulas is typically surgical; however, oophorectomy can be avoided if the ovary has no fistulisation. The patient's desire for fertility preservation should be assessed. Further research is needed to understand the impact of fistulising CD on ovarian reserve and fertility.

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