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Case report and short review of the literature

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Rectal perforation caused by deep infiltrating endometriosis in non-pregnant woman: Case report and short review of the literature.

AIM: *The aim of this paper is to describe an unique case of deep infiltrating endometriosis of the rectum in non-pregnant woman with unusual clinical and pathological presentation resulting in spontaneous perforation.*
MATERIALS AND METHODS: *A female (20 years of age) with a two year history of chronic recurrent abdominal pain of unknown etiology treated by a psychiatrist underwent diagnostic laparoscopy which revealed many peritoneal implants of endometriosis involving the right ovarian fossa, the vesico-uterine pouch and sacrouterine ligament; the bowel wall showed no structural abnormalities. Peritonectomy of the broad and uterosacral ligaments was used and eight days after the operation, the patient developed crampy abdominal pain and enterorrhagia necessitating laparoscopic revision; pelvic haematoma and rectosigmoiditis were found. Over the next three days, perforation of the rectum resulted in the presence of fecal material in the surgical drain.*
RESULTS: *Lower rectal resection with ileostomy was performed. Microscopic examination revealed discrete small endometriotic lesions in submucosa, muscular layer and serosa of the rectum associated with perforation.*
DISCUSSION: *Laparoscopy and laparotomy may be insufficient in the case of an inactive endometriosis. Definitive diagnosis is thus reached only by the histological examination. The pathophysiology of the bowel perforation secondary to endometriosis is not entirely clear.*
CONCLUSION: *The presented case confirms the importance of interdisciplinary cooperation between surgeons, gynaecologists, and pathologists. We also want to emphasize the need for extensive pathological examination of the resected specimens which is essential for a proper diagnosis.*

KEY WORDS: Endometriosis, Rectum, Spontaneous perforation

Introduction

Endometriosis is defined as the presence of endometrium outside the uterine corpus [1,2]. It usually affects many organs as the ovaries (endometriomas), fallopian tubes, uterus, urinary bladder, rectosigmoideum, uterosacral ligaments, the pouch of Douglas, and peritoneum [3,4]. It is a common, chronic, oestrogen-dependent disease affecting between 5% and 20% of women of reproductive age [5]. The intestinal involvement by endometriosis occurs in 3% to 37% of patients. Up to 73% of cases affect the lower rectosigmoid colon followed by the rectovaginal septum, terminal ileum, caecum, and the appendix [6].

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Superficial intestinal diseases in the form of serosal implants usual do not cause any symptoms, but bulky, deeply invasive diseases can cause real problems. Spontaneous perforation of intestinal endometriosis is a very rare complication but occurs most frequently during pregnancy [7,8].

Case Report

Case Presentation

A female, 20 years of age, gravida 0, para 0, was admitted to a surgical unit. She was suffering from acute and worsening anorexia, vomiting and abdominal pain. She had been experiencing the symptoms for about three months. The abdominal pain was mainly in the right lower quadrant and epigastrium. The patient also complained of dysmenorrhea. A complete gynaecological ultrasound examination was inconclusive.
Esophagogastroduodenoscopy (EGD), c-reactive protein (CRP) and white blood cells (WBC) were within normal ranges. As there was no obvious etiology of her symptoms, the patient was referred for psychiatric evaluation. Her symptoms persisted and she later underwent a diagnostic laparoscopy. Endometriosis of the vesico-uterine pouch, right ovarian fossa, right sacrouterine ligament and Allen-Masters syndrome were identified (Fig. 1 a,b). There were no signs of inflammation, infection nor other structural abnormalities on the appendix, sigmoid colon or rectum. The revised American Society for Reproductive Medicine (rASRM) score (stage IV) was used to classify endometriosis. Two months later the patient underwent deperitonealization of the posterior compartment, bilateral ureterolysis and revision of the sacrouterine ligaments. Four days after the operation, the patient was discharged in a stable condition without complaints of hematochezia. Eight days after the operation, the patient was returned to hospital with cramping abdominal pain, rectal and vaginal bleeding. Ultrasound showed pelvic haematoma consistent with postoperative changes. CRP was 130 mg/l, WBC 16,500 and procalcitonin was negative. A computer tomography (CT) scan of the abdomen and pelvis confirmed haematoma in the pelvic cavity and the patient was found to have a distended sigma. Flexible sigmoidoscopy revealed nothing due to poor bowel preparation. Later, the patient deteriorated clinically with worsening leukocytosis, fevers and increasing abdominal pain suggesting acute abdomen. A repeat laparoscopic evaluation was undertaken. The findings included a pelvic abscess and secondary inflammation of the rectosigmoideum (Fig. 1 c,d). No evidence of bowel perforation was found. The patient's clinical status deteriorated further and the devel-

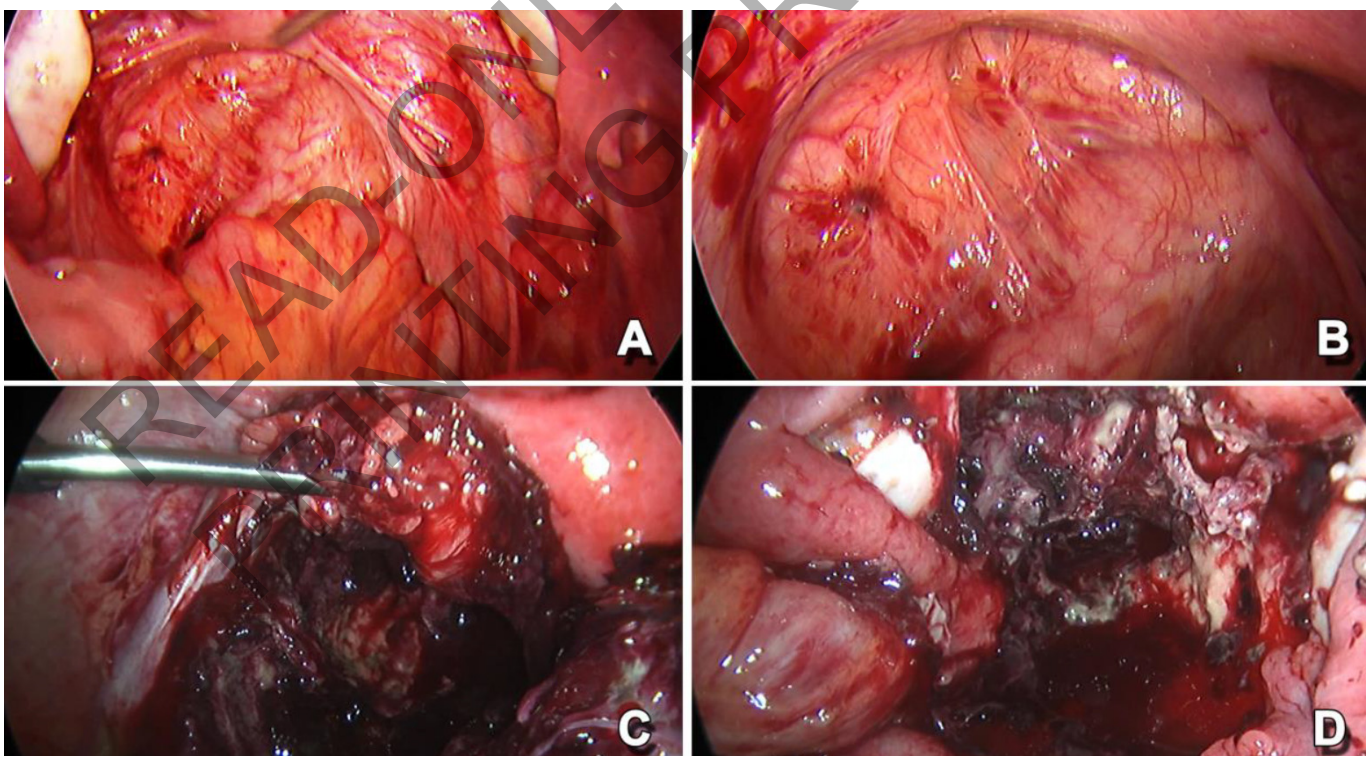

Fig. 1: Intraoperative laparoscopic images.
A,B: Initial diagnostic laparoscopy showing endometriosis of the lesser pelvis – vesico-uterine pouch, right ovarian fossa, right sacrouterine ligament, Allen Masters syndrome.
C,D: Laparoscopic revision of the abdominal cavity on the 8th postoperative day – pelvic haematoma and rectosigmoiditis.

opment of sepsis and stercoral peritonitis required emergent exploratory laparotomy. Acute pelviperitonitis along with a small perforation of the anterior rectal wall was identified. Low anterior resection of the rectosigmoid colon with protective ileostomy was indicated. After three months the patient underwent revision and reconstruction of the rectosigmoideum and is currently without symptoms.

Pathology

Three separate specimens were evaluated at the department of pathology: rectosigmoid colon resection specimen, 18 cm in the length (1), colon resection rings (2) and appendix (3). Macroscopic investigation of the colon revealed regressive and inflammatory changes including one 8 mm perforation. Thirty one tissue sections in total were stained with hematoxylin-eosin and extensively investigated. Cardinal histologic finding represented discrete endometriotic lesions in the submucosa, muscular layer and serosa of the rectum associated with full thickness bowel wall necrosis and acute inflammation (Fig. 2 a,b). These findings were confirmed by using immunohistochemistry - both stromal and glandular immunopositivity for oestrogen and progesterone receptors (ER, PR) (Fig. 2c), only stromal immunopositivity for CD10 (Fig. 2d), only glandular immunopositivity for cytokeratin 7 (CK7) and stromal and glandular immunonegativity for cytokeratin 20 (CK20).

The final diagnosis was of deep infiltrating endometriosis (DIE) of the rectum causing perforation and fibrinous-purulent peritonitis.

Discussion

Intestinal endometriosis typically involves areas where the peritoneum is irregularly folded, such as the rectovaginal septum, rectum, and sigmoid colon [1,6]. Most cases occur during surgical intervention or are revealed incidentally by pathological examination of tissues removed for different surgical indications [9].

The symptoms of gastrointestinal tract involvement by endometriosis are nonspecific and depend on a) the severity and b) the location of the disease. Superficial intestinal endometriosis may be asymptomatic or cause cyclical spastic pain [10]. When endometriosis deeply invades the bowel wall, it causes a scarring and retraction and can form a mass lesion which partially obstructs the bowel wall [11]. In such cases symptoms may include constipation, diarrhoea, melena, rectal bleeding, meteorism and tenesmus. It is very rare that the colon is perforated by endometriosis. When searching the literature, 12 cases of perforation of the small bowel, 16 cases of perforation of the large bowel and 3 cases of perforation of the appendix, due to intestinal endometriosis were found (a total of 31 cases). The first case report was published in 1931 by Haufler. A 30-year old women was reported with jejunal perforation due to rupture of

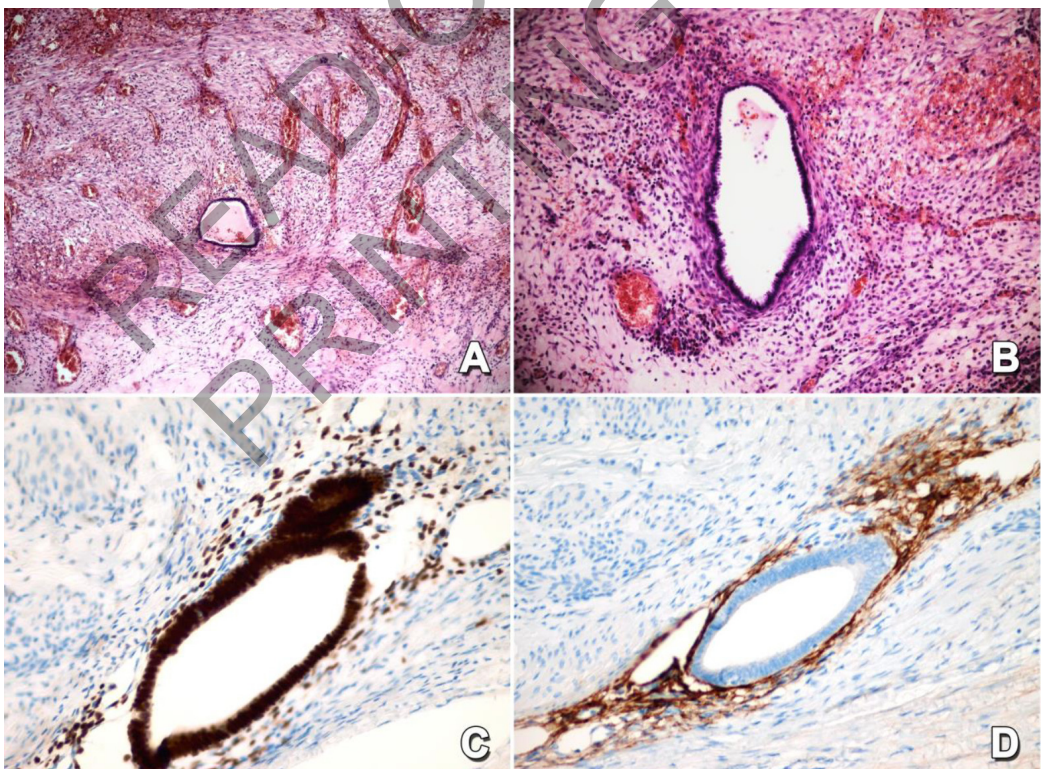

Fig. 2: Intestinal endometriosis, immunohistochemical expression of oestrogen receptors and CD10 in intestinal endometriosis.
A,B: Discrete endometriotic lesion and florid regressive and inflammatory changes in the subserosa of the bowel wall (hematoxylin-eosin staining, original magnification x100, x200).
C: Oestrogen receptors stain both endometrial gland and endometrial stromal cells (immunohistochemistry, original magnification x200).
D: CD10 stains a few endometrial stromal cells adjacent to the gland in the muscular layer of the bowel wall, confirming the diagnosis of intestinal endometriosis (immunohistochemistry, original magnification x200).

an endometriotic cyst during the sixth month of pregnancy [12]. Most recently, in 2017, Marujo et al. described a case of transmural perforation of the rectum wall by the left fallopian tube in a patient with DIE. A 38-year-old women, nulliparous, with a history of primary infertility, complained of chronic pelvic pain, dysmenorrhea, dyschezia, dyspareunia and the occasional rectal bleeding. On gynaecological examination, a painful retrocervical nodule and elastic mass on the left adnexal area was discovered. Given the clinical probability of DIE, the patient was initially treated with hormonal contraceptives. Using magnetic resonance imaging (MRI), rectal endoluminal formation was described; a biopsy taken during colonoscopy revealed an inflammatory polyp of the colon. Consequently, because of the symptoms and the presence of lesions compatible with DIE of the rectovaginal septum, with probable intestinal infiltration, the patient was recommended for surgery. During adhesiolysis it was found that the rectum had been pierced by the tube. Histological examination showed tubal endometriosis; involvement of the excised rectal segment by endometriosis was not diagnosed [13].

We have a few comments on this case. It is felt that a conservative approach should not be the first-choice of treatment (in this clinical context). Currently, many papers approve of the strategy of completely removing the bulky deeply invasive disease by laparoscopy in trained centers, followed by in vitro fertilization (IVF) in the case of sterility or hormonal treatment [14-18]. It is widely accepted that, because endometriosis is a systemic disease, the division between deep and superficial endometriosis is an anachronism. Endometriosis, in the form of DIE, does not show any respect for organ borders. The disease merges all concerned tissue into one bulky mass consisting of an active part of endometriosis, inflammatory tissue and hyalinisation. Pathological findings of the resected specimens are regionally heterogeneous and a large number of sections are required to determine the correct diagnosis. We would recommend a second-look histopathological examination of the resected rectal specimen focusing on the area of the perforation. Piercing is a very dubious mechanism of rectal penetration and we lack a competent commentary from the pathologist on this subject.

The current paper is interesting from two points of view: clinical and pathological. Misinterpretation of abdominal symptoms, initially treated by a psychiatrist and not surgically. Macroscopically, endometriosis involved the lesser pelvis, not the bowel, caused by inapparent lesions of inactive endometriosis and absence of fibrous adhesions within the lesser pelvis. Microscopically, discrete small lesions of endometriosis, in contrast to the cases of extensive transmural involvement that have so far been published, and absence of decidualized stromal cells. Spontaneous perforation of the rectum occurred after a simple surgical procedure – peritonectomy, without any manipulation with the bowel (e.g. rectal shaving) and with no use of coagulation devices, i.e. monopolar or bipolar.

The pathophysiology of the bowel perforation, being secondary to endometriosis, is not clear. Generally, two mechanisms should be considered. Firstly, endometriosis involving the intestinal tract may weaken the bowel wall while, on the one hand, the elasticity of the endometriotic tissue is impaired and causes a loss of elasticity and on the other hand, in the case of pregnancy, the whole endometriotic tissue is decidualised so that the elasticity of the tissue vanishes completely. Secondly, the intestine becomes a part of a convolute consisting of the uterus, adnexa and fibrous tissue (frozen pelvis) and is under tension of adhesions secondary to peristalsis. Both of these phenomenona together may compromise the intestinal wall integrity and result in perforation, especially during pregnancy [11].

There are few if any characteristic symptoms for intestinal endometriosis making it difficult to diagnose. FU et al. published a paper in 2007 recommending that intestinal endometriosis should be considered when a reproductive woman who has had a history of pelvic endometriosis developes a cyclical bowel discomfort [10]. In the case of chronic nonspecific abdominal pain, diagnostic laparoscopy is suggested. In accordance with some other papers, we recommend, in the case of asymptomatic superficial intestinal endometriosis, follow-up without any treatment. Resection of the involved bowel segment remains the treatment of choice for patients with intestinal endometriosis since the effect of gonadotropin-releasing hormone (GnRH) analogs and progestin is limited; also for patients with symptoms of obstruction or bleeding, and if malignancy cannot be excluded [7,18-20].

The most accepted etiological theories concerning endometriosis are still the peritoneal implantation of endometrium by retrograde menstruation, vascular dissemination or the possible metaplasia of peritoneal cells. Intestinal endometriosis predominantly involves the extra mucosal layers but may be found in all the layers of the bowel wall. Most endometriomas are ill-defined serosal and subserosal nodules that are rarely larger than 5 cm. On the cut surface, the spectrum of colors is quite broad, ranging from black through brown to red and white [21].

When examined microscopically, they consist of endometrioid glands and stroma that are often accompanied by fibrosis in the adjacent bowel wall.

Occasionally only endometrioid-type stroma, including a variety of changes such as decidua, smooth-muscle metaplasia, pseudodecidua, fibroblastic metaplasia and sarcoma, are present. Such lesions are referred to as stromal endometriosis and are considered to be most likely due to limited sampling [22]. In such cases a broad differential diagnosis should be considered including gastrointestinal stromal tumor or benign nerve sheath tumor [23].

From a histopathological point of view, it is necessary to differentiate between colorectal adenocarcinoma and endometriosis. Chen et al. in 2015 described a case report of a 39-year-old women with rectal mucosal endometriosis primarily misinterpreted as adenocarcinoma. Initial colonoscopy showed a rectal mass with ulceration and circum wall involvement. A combination of all the histological features, i.e. irregular glands with mucin depletion, nuclear stratification, subtile subnuclear vacuoles and spindle cells with abundant pink cytoplasm and an unclear boundary in the stroma, was subjectively interpreted as dysplastic glands in a desmoplastic setting with an initial suspicion of primary rectal adenocarcinoma. Subsequently, immunohistochemical examination with positivity for CK7, ER and CD10 identified the essence of ectopic endometrium [24]. As was shown in this case, the distinction between adenocarcinoma and endometriosis can be particularly challenging in mucosal biopsy specimens, mostly due to the limited amount of tissue present, misinterpreting of the reactice glandular changes and, because the endometrial tubules tend to be separated from their stroma, secondary to trauma from the biopsy procedure [23,25]. Routine light microscopy is usually sufficient to make the correct diagnosis. Malignancy is excluded by lack of significant cellular atypia, low mitotic activity and an absence of a desmoplastic stromal reaction. By the use of immunohistochemistry a diagnosis of colonic endometriosis can be confirmed. The first known case of pelvic lymph node endometriosis with aberrant immunophenotype with a complete loss of oestrogen and progesterone receptor expression of the endometrioid glands, mimicking metastasis of adenocarcinoma has been described by the authors [26]. Thus, it is important to keep in mind that the final interpretation must always be determined within the context of the morphological findings.
Laparoscopy and laparotomy, often considered as the gold standards for diagnosing pelvic endometriosis, may be insufficient in the case of an inactive endometriosis. As in the paper published in 2014 by Galazis et al, no active endometriosis of the bowel wall was seen during laparoscopy and laparotomy [27]. It is the histological examination that provides the definitive diagnosis. In our case, we initially found only regressive and inflammatory changes, including one perforation of the bowell wall. Due to unclear pathogenesis of these changes, another 15 sections of the resected colon were submitted. Evidence of endometriotic lesions in the submucosa, muscular layer and subserosa of the bowel wall were found in 3 hematoxylin and eosin stained slides. As a rule, in a non-neoplastic bowel resection specimen, it is necessary to submit representative sections of the proximal and distal margins and any focal lesions. It is also recommend that all areas of the bowel are sampled by submitting sections at regular 5-cm intervals. That way an extensive examination of the resection specimen is achieved leading to a proper diagnosis.

Conclusion

Intestinal endometriosis should be considered in the differential diagnosis of any gastrointestinal or abdominal symptoms of every women. It is difficult to diagnose and may even mimic other diseases, including neoplasm. The risk of intestinal involvement by endometriosis is increased by simultaneous gynaecological symptons and diagnostic laparoscopy should be considered. Patients should be closely followed up after the operation.
We also want to emphasize the need for extensive pathological examination of the resected specimens which is essential for a proper diagnosis.
This report is believed to be a unique case of spontaneous rectal perforation secondary to endometriosis in non-pregnant women.

Riassunto

Viene descritto il caso di una perforazione spontanea del retto da infiltrazione endometriosica profonda, in una donna non gravida, e la sua insolita presentazione clinica e patologica,
Donna di 20 anni, con storia di dolore addominale cronico recidivante da due anni di etiologia ignora, trattata da uno psichiatra. A seguito di una laparoscopia diagnostica sono stati rilevati molti impianti peritoneali di endometriosi interessanti la fossa ovarica destra, la tasca vescico-uterina ed il legamento utero-sacrale, mentre la parete dell'intestino non mostrava anomalie strutturali.
Sottoposta quindi a peritonectomia dei legamenti larghi ed uterosacrali, dopo otto giorni dall'intervento sono insorti dolori addominali, crampi ed enterorragia che richiese una revisione laparoscopica che dimostrò un ematoma pelvico ed una rettosigmoidite, e tre giorni dopo una perforazione del retto con comparsa di materiale fecale nel drenaggio chirurgico. La paziente venne pertanto sottoposta a resezione del retto inferiore con ileostomia.
L'esame istologico ha dimostrato piccole e discrete lesioni endometriosiche nella sottomucosa, nello strato muscolare e nella sierosa del retto in associazione con la perforazione.
Laparoscopia e laparotomia possono essere insufficienti a riconoscere una endometriosi in fase di quiescenza, e la diagnosi definitiva è raggiungibile solo con l'esame istologico. La fisiopatologia della perforazione intestinale secondaria all'endometriosi non è del tutto chiara.
il caso presentato conferma l'importanza della cooperazione interdisciplinare tra chirurghi, ginecologi e anatomo-patologi, ed a questo proposito si sottolinea la necessità di un ampio esame patologico dei tessuti resecati, essenziale per una corretta diagnosi.

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Commento e Commentary

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The case described is very suggestive from a clinical and anatomo-pathological point of view. However, it is believed that the adjective "spontaneous" attributed to rectal perforation should only be accepted as an alternative hypothesis, both because the area of the perforation site had been modified by the deperitonization of the first operations, second because of the limited time interval between intervention and symptoms of rectal perforation preceded by prodromal symptoms.
The protean clinical presentation with a history of over two years, and the extreme variety of multifocal anatomo-pathological involvement of tissues, even deep in the rectal wall, confirms the diagnostic difficulties of this relatively rare disease.

* * *

Il caso descritto è molto suggestivo dal punto di vista clinico ed anatomo-patologico. Si ritiene però che l'aggettivo "spontaneo" attribuito alla perforazione rettale vada accettato solo come una ipotesi alternativa, sia perchè l'area della sede della perforazione era stata rimaneggiata dalla deperitoneizzazione del primo interventi, sia per il limitato intervallo di tempo intercorso tra l'intervento stesso ed i sintomi della perforazione rettale preceduta da sintomi prodromici.
La proteiforme presentazione clinica con una storia di oltre due anni, e l'estrema varietà di coinvolgimento anatomo-patologico multifocale dei tessuti, anche in profondità della parete rettale, conferma le difficoltà diagnostiche di questa relativamente rara patologia.