## CLINICAL PRACTICE ARTICLE



# Use of strictureplasty technique for surgical treatment of

# ileal endometriosis: a case series [version 1; peer review: 1

# approved, 1 not approved]

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#### Abstract

Intestinal deep infiltrating endometriosis is the most frequent extragenital localisation and its traditional surgical treatment is segmental resection of the affected tract. The need for implementing alternative techniques in the treatment of intestinal endometriosis arises from those cases of multiple ileal and recto-sigmoidal localisations, in which removing excessive lengths of intestine could lead to a higher rate of adverse events. Ileal endometriosis represents 4.7% of all intestinal localisations, often associated with multiple lesions and yet, to the best of our knowledge, there are no data on techniques other than intestinal resection for its treatment. Since its capacity to solve fibrostenotic lesions without removing centimeters of intestine, strictureplasty is widely implemented in the management of Crohn's disease. We propose the use of stricture plasty for the treatment of ileal endometriosis. We performed surgical treatment for symptomatic deep infiltrating endometriosis in two patients with either ileal and sigmoidal localisations. We approached ileal nodules with strictureplasty technique, while sigmoidal nodules were removed by traditional segmental resection. No complications occurred and both patients are now asymptomatic after a 12 months-follow up. Therefore, strictureplasty could provide a tool to eliminate small bowel endometriosis maintaining a regular caliber of the ileal tract without modifying its length.

#### **Keywords**

ileal endometriosis, deep infiltrating endometriosis, resection, strictureplasty, obstruction

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Any reports and responses or comments on the article can be found at the end of the article.

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Author roles: Alboni C: Conceptualization, Formal Analysis, Investigation, Methodology, Project Administration, Supervision, Validation, Writing – Review & Editing; Camacho Mattos L: Data Curation, Formal Analysis, Methodology, Writing – Original Draft Preparation; Facchinetti F: Resources, Supervision, Writing – Review & Editing; Cabry F: Conceptualization, Formal Analysis, Methodology, Project Administration, Validation; Serra F: Methodology, Project Administration, Visualization; Ricciardolo A: Methodology, Project Administration, Visualization; Mabrouk M: Conceptualization, Investigation, Supervision, Writing – Review & Editing; Gelmini R: Conceptualization, Methodology, Project Administration, Supervision, Writing – Review & Editing

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#### Background

Endometriosis is a benign estrogen-dependent disease in which endometrial-like tissue with stroma and glands is found outside the uterus. It can present in a variety of forms, classified as typical, subtle, cystic and deep infiltrating endometriosis (DIE) with deep endometriosis being mostly associated with pelvic pain and severity of symptoms<sup>1</sup>. The intestine is a frequent localisation of deep infiltrating endometriosis, with nodules most commonly affecting the recto-sigmoid tract (83.1%), the appendix (6.4%), the caecum and ileocaecal junction (4.1%), the small bowel (4.7%), and the omentum  $(1.7\%)^1$ . Symptoms that patients with intestinal endometriosis could experience are dysmenorrhea, rectal pain extended to the perineum, diarrhea, catamenial rectal bleeding, constipation and subocclusive symptoms, nausea and vomiting, cramps and abdominal distension; acute occlusion is rare<sup>2</sup>. Traditional surgical treatment for intestinal localisation is resection of the affected tract. However, in those cases with multiple nodules the surgeon must consider the risks related to a wide or multiple intestinal resection and, to the best of our knowledge, yet there are no data on specific techniques other than intestinal resection for ileal infiltrating endometriosis treatment<sup>3</sup>.

#### Technique and our experience

To avoid excessive removal of intestine in the treatment of ileal endometriosis, we propose the use of a technique known as strictureplasty. This technique was firstly described in 1977 by Katariya et al., who published the results of stricture plasties in a series of nine patients with multiple tubercular strictures producing symptoms of obstruction<sup>4</sup>. Then in 1982, Lee and Papaioannou published the results of stricture plasty on nine patients with Crohn's disease and obstructive symptom<sup>5</sup>. At present, strictureplasty is widely implemented in the treatment of Crohn's disease for its capacity to solve multiple fibro-stenotic lesions without removing sections of the intestine. Several strictureplasty techniques can be performed depending on the length of the stricture, the most commonly used being the Heikene-Mikulicz technique<sup>6</sup>. It consists of making a longitudinal incision across the obstructed area extending into normal bowel on the antimesenteric side, creating an enterotomy which is sutured transversely<sup>7</sup> (Figure 1).

In clinical practice and in literature ileal lesions are surgically removed in different ways but, to the best of our knowledge, no technique is standardized apart from segmental resection. Herein, we performed and standardized a technique which is already fully consolidated in general surgery for the treatment of diseases which produce ileal lesions similar to those produced by endometriosis. No devices other than common surgical instrumentation have been used. The decision to use this technique was taken with the intention to treat the patients described below, and not with the aim of testing the procedures performed. Therefore, as a common clinical practice in our institution in general surgery, there was no need for consultation of IRB for approval for implementing the procedure that we describe.

We present our experience on two patients with either sigmoidal endometriosis and ileal localisations.

#### Case 1

A caucasian 36-year old cashier was followed at the Endometriosis referral Centre of Policlinico di Modena for deep infiltrating endometriosis which caused light dysmenorrhea, dyspareunia and heavy dyschezia. She was diagnosed with transvaginal ultrasound to have a single sigmoid localisation of endometriosis of 3 centimeters in longitudinal diameter, with partial infiltration of the sigmoid wall. Surgery was planned to remove endometriosis localisations and the patient was put on the waiting list. On June 2018, after 4 months from the first examination, she presented at Gynaecologic Emergency Department for heavy abdominal pain, dysmenorrhea and constipation from 3 days. She underwent plain abdominal radiograph which showed multiple air-fluid levels and an abdominal computed tomography (CT) showing dilation of the proximal tract of the small bowel with no clear sign of obstruction. The patient was diagnosed with suspected intestinal occlusion. Conservative treatment with fasting and hydration has been established but in the proceed 12 hours the patient conditions declined with worsening of symptoms. An urgent laparoscopy was performed. During the intervention, the patient was found to have three ileal lesions suggestive of endometriosis localisation as well as the known sigmoid localisation. The mean diameter of the ileal lesions was 5 millimeters and they were all placed on the antimesenteric side of the ileal wall, from 20 to 50 centimeters from the ileo-cecal junction, causing partial stricture of that portion of the intestine. The surgical approach has been decided together in an equipe of expert gynaecologists and general surgeons. Ileal nodules have been approached by removing the trocar from the right lateral laparoscopic access and widening it to five centimeters, therein exteriorizing the interested ileal tract. The ileal nodules have been completely removed by lancing the ileal wall in the shape of a lozenge with its major axis longitudinal to the intestine itself, sparing the mucosal layer of the intestine which was not involved in the lesion and therefore not creating an enterotomy in the wall of the strictured tract. Afterwards, the ileal wall was closed transversely with a single layer, interrupted stitches, monofilament absorbable suture, 3-0. Eventually the ileum was re-inserted in the abdominal cavity through the enlarged lateral access. The same technique was performed for each of the three ileal nodules. In such a manner, it was possible to radically remove all endometriosis lesions maintaining a regular caliber of the ileal tract and without modifying its length. The intervention has been completed by resecting the sigmoid nodule with a traditional technique with Knight-Griffen<sup>8</sup> anastomosis.

Post-operative period was uneventful: the patient developed no fever or any sign of infection; she started a light diet on the second day after surgery and underwent complete re-canalisation on the fourth day after surgery. She was discharged on the fifth day after surgery with a regular diet and the prescription of Leuproreline acetate 3.75 mg, one injection per month for three months, followed by continuous combined oral contraceptive (Nomegestrol acetate 2.5 mg/Estradiol 1.5 mg). One month after surgery the patient came for a post-operative examination: she indicated no symptoms and regular alvum; no sign of relapse of the disease

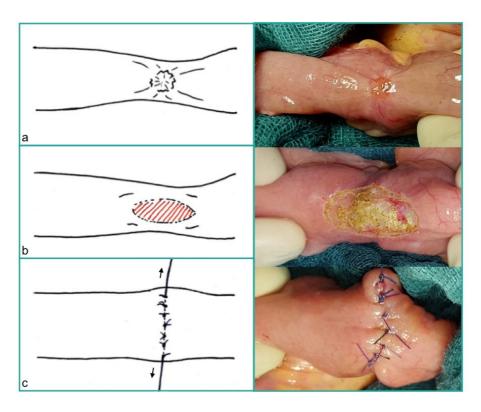


Figure 1. Strictureplasty technique for ileal endometriosis. a. Identification of the nodule/s by laparoscopic visualization and evaluation of the ileal wall by palpation once the ileal loop is exteriorized from the abdomen; b. Removal of the nodule lancing ileal wall in the shape of a lozenge with a longitudinal axis, with the mucosal sparing option; c. Transverse suture (interrupted stitches, absorbable suture, 3-0) to reduce the risk of ileal caliber reduction during healing process.

was found on sonographic evaluation. No long-term complications occurred in 12 months of clinical follow up, in terms of intestinal dehiscence, surgical site infection, weight loss, absorption discrepancies or relapse of the disease.

#### Case 2

On January 2019 a caucasian 36-year old school teacher came to the attention of the Endometriosis referral Centre of Policlinico di Modena for consultation. She presented with chronic pelvic pain, catamenial dyspareunia, cramps and abdominal distension. After accurate history collecting, physical examination and sonographic evaluation were performed the patient was diagnosed with suspected right ovarian endometriosis, recto-sigmoidal endometriotic localisation of 4 centimeters with complete infiltration of the bowel wall and thickening of right utero-sacral ligament. Surgery was planned and in the meantime the patient was prescribed with combined oral contraceptive (Nomegestrol acetate 2.5 mg/Estradiol 1.5 mg) on a continuous regimen. After four months the patient was called in for the surgery; at that time, she indicated improvements for all symptoms with the exeption of abdominal distension, which remained constant and occasionally associated with vomiting. An operative laparoscopy was performed and all endometriotic lesions suspected on sonographic evaluation were confirmed. Moreover, a single hard nodule of ten millimeters in diameter was found on the antimesenteric surface

of the ileum, approximately at 10 centimeters from ileo-cecal junction, causing a thickened stricture of the intestine, supposedly the cause of patient's vomiting and abdominal distension. All lesions were removed, performing extensive adhesiolysis, right ovarian cystectomy, removal of right utero-sacral ligament and segmental resection of the affected recto-sigmoidal tract with a traditional technique with Knight-Griffen anastomosis. The ileal nodule was approached using strictureplasty technique: the affected ileal tract was temporarily extracted from the abdomen from the widened right lateral access; the nodule was removed from the ileal wall in the shape of a lozenge with its major axis longitudinal to the intestine; ileal mucosa was macroscopically diseasefree and therefore it has been spared. A single layer suture with interrupted stitches in monofilament absorbable suture, 3-0, was performed transversely to the major axis of the incision. Eventually, the ileum was completely disease free and showed a regular caliber along the whole tract.

Post-operative course was regular: fasting was stopped after two days and a light diet was commenced; complete canalization occurred on the fourth day and the patient was discharged on the sixth day after surgery. She was prescribed with a three-months course of Leuproreline acetate 3.75 mg, one injection per month, followed by continuous combined oral contraceptive pill (Nomegestrol acetate 2.5 mg/Estradiol 1.5 mg). One month after surgery, the patient was completely recovered and asymptomatic. Further follow-up examinations have been performed at 3, 8 and 12 months after surgery and the patient indicated no symptoms. She is still following the prescribed therapy with no side effects. No long-term complications occurred.

#### Discussion

Given the significant impact on the quality of life, the young average age of the patients affected by endometriosis and the need to restore a physiologic condition when treating this disease, intestinal endometriosis is far from being an easy challenge. The most common surgical treatment for intestinal localisations is segmental resection and yet, to the best of our knowledge, there is no standardized alternative for the surgical treatment of ileal endometriosis, neither laparotomic nor laparoscopic. In those cases of multiple infiltrating nodules, the decision of the type of surgical strategy needs to take into account the risk of removing excessive lengths of the intestine. This assumption conveys the necessity of implementing intestine-sparing alternative techniques in the surgical treatment of these localisations of endometriosis, thus to reduce the risk of adverse events such as short bowel syndrome9, anastomotic dehiscence, bowel obstruction and reduction in quality of life. As a matter of fact, strictureplasty is an effective and widely applied surgical technique developed as an alternative to segmental resection for patients at risk for short bowel syndrome.

Reporting these two cases, the authors propose strictureplasty as an option in the surgical treatment of deep infiltrating endometriosis with multiple intestinal nodules and ileal localisations. Heikene-Mikulicz technique seems to be appropriate for removing endometriosis lesions without reducing the length and caliber of the ileum. A specific strength of the procedure described is the possibility of sparing the mucosal layer of the intestine in those cases with no involvement of the mucosa, by implementing a modified Heikene-Mikulicz strictureplasty, as performed in our case series. However, due to the low incidence of small bowel endometriosis, the number of cases is still very small; therefore, larger studies are mandatory to verify the efficacy and safety of this technique and longer follow up periods are necessary to define the risks of intestinal relapse and recurrence of symptoms. Moreover, the small number of cases does not allow any precise indication on the dimension and position of ileal nodules which can be approached with this technique. It seems reasonable that nodules that transversely occupy more than one half (>50%) of the ileal diameter are more safely approached with resection; however, this assumption is only a speculation based on general surgeon experience and wider implementation of the technique with systematic reporting of the lesion dimensions and depth of infiltration is needed in order to give precise indications.

#### Conclusion

Gynaecologists need to be prepared to face and treat intestinal endometriosis with multiple recto-sigmoidal and ileal localisations. In those patients with no response or contraindication to medical therapy, a surgical approach has to be cautiously planned, considering the risks and benefits of eradication of all lesions. In order to reduce the risk of small bowel syndrome and improve the radicality of the intervention, strictureplasty technique could be an alternative to segmental resection approaching ileal nodules of small dimensions. This could provide less invasive impact on intestinal function and good results in terms of symptoms improvement and risk of recurrence.

#### Consent

Written informed consent for publication of their clinical details and clinical images was obtained from the patients.

#### Data availability

#### Underlying data

All data underlying the results are available as part of the article and no additional source data are required.

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Version 1

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## Philippe Koninckx 🔟

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The manuscript 'Use of strictureplasty technique for surgical treatment of ileal endometriosis: a case series' describes the use of strictureplasty for ileal endometriosis. It is an opinion paper and difficult to judge since there are little hard data.

My advice would be to reconsider the manuscript after a more balanced revision. Alternatively, a simultaneous comment would be necessary.

The fundamental problems are:

- Deep endometriosis is not defined. If deeper than 5mm is used as a definition, an important overlap exists with typical lesions. The multifocality of deep endometriosis is strongly associated with the definition.
- The deep endometriosis of the sigmoid is not clearly described and the reader might wonder whether a sigmoid resection was indicated.
- The lesions shown in the picture are small superficial lesions, which we used to treat superficially with a suture to reinforce the wall.

In conclusion:

- It is nice to emphasise that a bowel resection is not needed for this type of ileum lesion, especially since they are so rare that few people have experience.
- Technically it is simple destruction (depth to be debated) with a reinforcement of the wall.
- A similar full discussion for the sigmoid is much more difficult.
- Essential for the discussion, is the diagnosis and definition of obstruction/stenosis of small bowel, ileum and large bowel.

Is the background of the cases' history and progression described in sufficient detail? Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment? No

Is the conclusion balanced and justified on the basis of the findings?  $\ensuremath{\mathbb{No}}$ 

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: endometriosis surgery

I confirm that I have read this submission and believe that I have an appropriate level of expertise to state that I do not consider it to be of an acceptable scientific standard, for reasons outlined above.

Reviewer Report 20 July 2020

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## Michele Vignali

Department of Biomedical Sciences for Health, University of Milan, Milan, Italy

I read with great interest the paper titled 'Use of strictureplasty technique for surgical treatment of ileal endometriosis: a case series'. The Authors explain very clearly a new technique to remove ileal endometriosis nodules. This technique, called strictureplasty, derives from the experience of general surgeons in the treatment of Crohn's disease. The Authors report two patients treated with this technique who have had excellent results without post-operative complications and recurrence after 12 months of follow up. Excellent job!

Is the background of the cases' history and progression described in sufficient detail?  $\ensuremath{\mathsf{Yes}}$ 

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

F1000 Research

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment? Yes

Is the conclusion balanced and justified on the basis of the findings?  $\ensuremath{\mathsf{Yes}}$ 

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: gynecological surgery; laparoscopy; endometriosis;

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

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