International Surgery Journal
Agrawal TL et al. Int Surg J. 2024 Mar;11(3):453-456
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

Case Report

DOI: https://dx.doi.org/ 10.18203/2349-2902.isj20240576

Endo the line: stricturing terminal ileum endometriosis

Tushar L. Agrawal[1]*, Richard Maguire[1], Sooraj Pillai[2], Arun D. Naik[1]

[1]Department of General Surgery, Gold Coast University Hospital, Southport, QLD, Australia
[2]Department of Anatomical Pathology, Gold Coast University Hospital, Southport, QLD, Australia

Received: 12 February 2024
Accepted: 26 February 2024

***Correspondence:**
Dr. Tushar L. Agrawal,
E-mail: tushar.agrawal@health.qld.gov.au

ABSTRACT

Endometriosis is the implantation and proliferation of endometrial tissue outside the uterus, and while involvement of the small bowel is rare, it is particularly unusual for it to present with terminal ileum stricture mimicking Crohn's ileitis. We present the case of a 38-year-old female with recurrent mechanical small bowel obstruction with transition point in the terminal ileum. She was struggling with infertility issues and was not previously known to have endometriosis. The presumptive diagnosis was Crohn's disease due history of previous anal fissure, high faecal calprotectin level, and imaging findings of terminal ileitis with stricture, despite lacking tissue confirmation from colonoscopy. She underwent laparoscopic ileocolic resection with histology showing endometrial stricture and secondary mucosal inflammation, without any established features of Crohn's disease. This case demonstrates the potential diversity in presentation of endometriosis, including small bowel manifestations that can mimic Crohn's ileitis. If endometrioma can be confidently diagnosed based on characteristic imaging features or tissue sample, unnecessary treatment may be avoided, whilst appropriate specialist management improves endometriosis symptoms and fertility outcomes.

Keywords: Crohn's ileitis, Endometriosis, Small bowel obstruction, Stricture

INTRODUCTION

Endometriosis is the extra-uterine implantation and proliferation of endometrial tissue, and while these lesions are typically found in the pelvis, they can occur throughout the body, including more proximally within the gastrointestinal tract.[1] Endometriosis is common, with the prevalence ranging from 1-7% in asymptomatic individuals, peaking in women of reproductive age.[2] Although a benign condition, symptoms can be severely impactful on quality of life, with resultant inflammation causing dysmenorrhea, dyspareunia, chronic pain, and infertility.[2]

Rectovaginal disease is the most common intestinal manifestation, present in 5-25% of patients with endometriosis.[3] This represents a form of deep infiltrating endometriosis (DIE), which are endometriotic lesions more than 5 mm below the peritoneum. When there is bowel involvement, this correlates to invasion of the muscularis propria. It is important to note that most affected patients have multiple lesions, raising the possibility of disease in more than one bowel segment. When considering intestinal endometriosis, the rectum and sigmoid colon are the most common sites (>90%), followed by the appendix (2-10%), with small bowel involvement occurring in less than 2% of these cases.[4]

The pathogenesis of symptomatic endometriosis is suggested to involve local inflammatory changes with increased production of inflammatory and pain mediators.[5] This case demonstrates mucosal inflammation and subsequent stricture from a terminal ileum endometrial deposit as a mimic of inflammatory bowel disease (IBD), specifically Crohn's ileitis.

CASE REPORT

A 38-year-old female presented to the emergency department with a one-week history of abdominal pain, nausea and vomiting, and diarrhoea, on a background of episodic cramping lower abdominal pain for the previous

two years. Initially thought to be gastroenteritis due to a recent travel history and sick contacts at home, she was discharged home and advised to return should symptoms continue. The pain persisted and she went on to develop worsening abdominal distension, constipation, and obstipation, and re-presented to the emergency department three days later. Subsequent investigation showed a small bowel obstruction with a single transition point in the terminal ileum.

Her background was significant for an anal fissure with associated necrotic fibroepithelial polyp which was removed during examination under anaesthesia of the rectum four years prior. Short-term follow-up colonoscopy at that time did not find any further polyps or other colonic pathology. There was no family history of IBD or colorectal cancer. She was an active smoker but otherwise had no medical history and did not take any regular medications. She had been struggling with infertility issues for several years but had yet to seek assisted reproductive technologies, and at the time of presentation, had no known diagnosis of endometriosis.

On examination, she was most tender in the right lower quadrant of the abdomen, without features of peritonism. Serum biochemical results were unremarkable with normal inflammatory markers (white cell count 5.3×10^9/l, C-reactive protein 5.5 mg/l), electrolytes (sodium 137 mmol/l, potassium 4.0 mmol/l), and renal function (creatinine 54 µmol/l, estimated glomerular filtration rate >90 ml/min/1.73 m^2). Faecal examination was negative for bacterial or viral infection, but the calprotectin level was raised at 2,100 µg/g. Computed tomography (CT) scan showed small bowel obstruction with transition point at the terminal ileum, secondary to terminal ileitis (Figure 1). There was associated mural thickening and submucosal hypodensity extending to the ascending colon, concerning for enterocolitis. There was no evidence of perforation, collection, or pneumatosis.

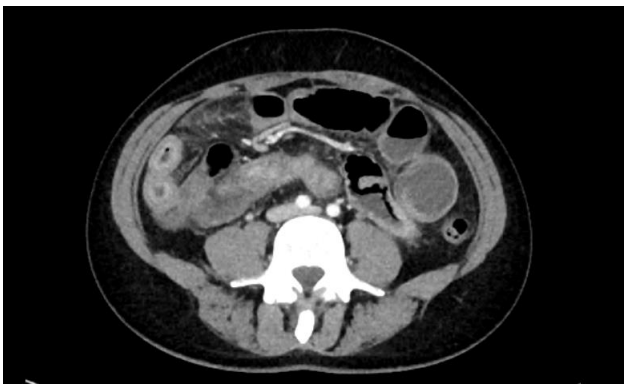

Figure 1: Axial CT image showing terminal ileitis with proximal small bowel obstruction.

Given the imaging findings of terminal ileitis, elevated faecal calprotectin, and history of anal fissure, the presumptive clinical diagnosis was Crohn's disease and gastroenterology opinion was sought. She was commenced on a short course of IV hydrocortisone which improved her symptoms, having otherwise conservative management for the small bowel obstruction. Inpatient colonoscopy showed a patch of mild inflammation 5-7 cm from the ileocecal valve with a narrow stricture through which the scope was unable to be passed (Figure 2). Histology of this stricture showed normal small intestinal mucosa. There were no extra-intestinal manifestations of Crohn's disease. She was commenced on exclusive enteral nutrition (EEN), a weaning prednisone regime, and azathioprine 75 mg once daily.

Figure 2: Endoscopic images of terminal ileal stricture taken during colonoscopy (a) caecum, (b) and (c) terminal ileum.

Subsequent outpatient magnetic resonance enterography showed a 2 cm stricture in the terminal ileum. Her case was discussed in the IBD multi-disciplinary team meeting which recommended the patient discuss surgical options. However, prior to being seen in the colorectal surgery outpatient clinic, she was admitted to hospital with a recurrent small bowel obstruction with mechanical transition point immediately proximal to the known terminal ileum stricture (Figure 3). She was managed conservatively in the first instance and underwent elective laparoscopic ileocolic resection three weeks later. Intra-operatively, a short segment of terminal ileum stricturing with proximal dilatation was found. 10cm of small bowel was resected with side-to-side functional ileocolic anastomosis performed. She had an uneventful recovery and was discharged from hospital on post-operative day seven.

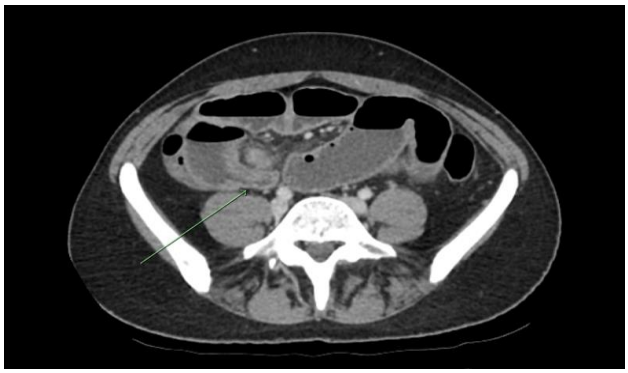

Figure 3: Axial CT image showing terminal ileal stricture causing small bowel obstruction.

The specimen consisted of a 10 cm segment of terminal ileum, caecum, appendix and proximal ascending colon. There was a stricture with ileo-ileal wall adhesion over a length of 50 mm. The strictured area had a narrow lumen with diameter of 7mm and wall thickness of 15 mm. The mucosal surface showed secondary inflammation but was otherwise unremarkable. Sections of small intestine showed evidence of endometrial glandular epithelium with associated stroma within the muscularis propria of the strictured segment (Figure 4). There was also endometriosis seen in one of the sampled lymph nodes. There were no granulomata, lymphoid aggregates, neuronal hyperplasia, or other established features of Crohn's disease.

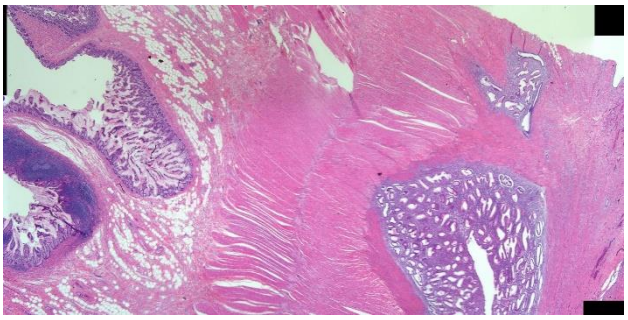

Figure 4: Small intestinal tissue with normal native mucosa and endometriosis in the muscularis propria.

Her IBD treatment was ceased, and she did not have any further gastrointestinal symptoms at one-year follow-up. She was referred to the gynaecology team for further investigation and management, as this unexpected diagnosis was likely contributing to her infertility issues. Pelvic ultrasound showed ovarian adhesions, uterine adenomyosis, and features of DIE within the left uterosacral ligament. A levonorgestrel-releasing intra-uterine device was inserted as medical management and she was referred to an in-vitro fertilization specialist.

DISCUSSION

Endometriosis is the implantation and proliferation of endometrial tissue outside the uterus, and although involvement of the small bowel is rare, it is particularly unusual for it to present with terminal ileum stricture mimicking Crohn's ileitis. Endometriosis is a common condition in women of reproductive age, with an incidence of 1-7% found on laparoscopy in asymptomatic individuals, and as high as 60-90% in patients with chronic pelvic pain and infertility.[2] The most common gastrointestinal manifestation is rectovaginal disease present in 5-25% of all cases, while small bowel involvement is rare, representing less than 2% of lesions found in the gastrointestinal tract.[3]

The pathogenesis of endometriosis involves the implantation and growth of ectopic endometrial cells to elicit an inflammatory response. The development of endometriosis is likely multifactorial involving aberrant production and transportation of endometrial cells, deficient cellular immunity causing an imbalance between proliferation and apoptosis, endocrine signalling, and genetic factors.[6] Sampson's theory of retrograde menstruation proposes ectopic endometrial cells occur when cells flow backwards into the peritoneal cavity during menstruation.[7]

However, there are likely other factors involved as endometriosis can be found in premenarchal girls, and due to the relatively low prevalence of endometriosis when compared to retrograde menstruation. Other theories involve coelomic metaplasia and Mullerian remnants, but none explain the diversity of presentations seen with endometriosis.[7] Genetic studies have found abnormalities in certain proto-oncogenes including KRAS, PIK3CA, and ARID1A which, despite being a benign condition, may explain the aggressive nature of some endometrial lesions.[8]

The diagnosis of endometriosis can be presumptive or surgical. Presumptive clinical diagnosis is based on characteristic symptoms, examination findings, and imaging features, and is gaining favour for patients with mild to moderate symptoms who do not have endometriomas, as it is less invasive and reduces treatment delay.[9] While laparoscopy is still the gold standard for diagnosis, ultrasound and magnetic resonance imaging (MRI) are increasingly recommended as early diagnostic tests in emerging guidelines. MRI has high sensitivity (90%) and specificity (91%), particularly for lesions containing blood products, while ultrasound can be similarly effective depending on location.[10] Surgical diagnoses with laparoscopy is preferred for patients with more severe symptoms that have not responded to medical treatment, and has the advantage of being both diagnostic and therapeutic.

Though the specific cellular and molecular mechanisms of inflammation in endometriosis remain to be clarified, there is local production and upregulation of inflammatory and pain mediators. There is significant elevation of macrophages within tissue samples from both normal peritoneum and peritoneal lesions in women with endometriosis.[11] There are also significantly more nerve fibres in DIE with strong expression of factors involved in neurogenesis including nerve growth factor, Trk-A, and p75, to increase peripheral nerve sensitization.[12] In addition, the Wnt/β-catenin signalling pathway has been shown to mediate fibrogenesis in endometriosis, leading to scarring, chronic pain, and altered tissue function.[13]

Chronic inflammation with fibrosis in the intestines can lead to stricture formation and is the proposed mechanism for the pathology seen in this case. This is similar to the pathogenesis of stricture formation in Crohn's ileitis, which is driven by cytokine mediated smooth muscle proliferation and collagen deposition.[14] Small bowel

endometriosis poses a diagnostic dilemma, particularly when biopsies taken during colonoscopy failed to demonstrate features of neither endometriosis nor Crohn's disease. Imaging findings can also be similar, with MRI showing mural hyperenhancement, restricted diffusion, and intra-mural oedema.[15]

In addition, co-existing disease can occur and has been described on several occasions in the literature, further complicating the diagnostic algorithm. Ultimately, patients with obstructive complications from small bowel stricture should proceed to surgical resection. If endometriosis is suspected, these lesions may regress with gonadotropin-releasing hormone agonist, but surgical resection remains the mainstay of treatment.[16]

CONCLUSION

This case demonstrates the potential diversity in presentation of endometriosis, including small bowel manifestations that can mimic Crohn's ileitis. If endometrioma can be confidently diagnosed based on characteristic imaging features or tissue sample, unnecessary treatment may be avoided, whilst appropriate specialist management improves endometriosis symptoms and fertility outcomes.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Mehmood S, Zhao S, Ain Q, Van Dellen J, Beggan C. Endometriosis of the Small Bowel: A Diagnostic Enigma. Cureus. 2021;13(6):e15520.
2. Hickey M, Ballard K, Farquhar C. Endometriosis. BMJ. 2014;348:g1752.
3. Seracchioli R, Poggioli G, Pierangeli F, Manuzzi L, Gualerzi B, Savelli L, et al. Surgical outcome and long-term follow up after laparoscopic rectosigmoid resection in women with deep infiltrating endometriosis. BJOG. 2007;114(7):889-95.
4. Pereira RM, Zanatta A, Preti CD, de Paula FJ, da Motta EL, Serafini PC. Should the gynecologist perform laparoscopic bowel resection to treat endometriosis? Results over 7 years in 168 patients. J Minim Invasive Gynecol. 2009;16(4):472-9.
5. McKinnon BD, Bertschi D, Bersinger NA, Mueller MD. Inflammation and nerve fiber interaction in endometriotic pain. Trends Endocrinol Metab. 2015;26(1):1-10.
6. Giudice LC. Clinical practice. Endometriosis. N Engl J Med. 2010;362(25):2389-98.
7. Vercellini P, Viganò P, Somigliana E, Fedele L. Endometriosis: pathogenesis and treatment. Nat Rev Endocrinol. 2014;10(5):261-75.
8. Anglesio MS, Papadopoulos N, Ayhan A, Nazeran TM, Noë M, Horlings HM, et al. Cancer-Associated Mutations in Endometriosis without Cancer. N Engl J Med. 2017;376(19):1835-48.
9. Agarwal SK, Chapron C, Giudice LC, Laufer MR, Leyland N, Missmer SA, et al. Clinical diagnosis of endometriosis: a call to action. Am J Obstet Gynecol. 2019;220(4):354.
10. Bazot M, Darai E, Hourani R, Thomassin I, Cortez A, Uzan S, et al. Deep pelvic endometriosis: MR imaging for diagnosis and prediction of extension of disease. Radiology. 2004;232(2):379-89.
11. Tran LV, Tokushige N, Berbic M, Markham R, Fraser IS. Macrophages and nerve fibres in peritoneal endometriosis. Hum Reprod. 2009;24(4):835-41.
12. Wang G, Tokushige N, Markham R, Fraser IS. Rich innervation of deep infiltrating endometriosis. Hum Reprod. 2009;24(4):827-34.
13. Matsuzaki S, Darcha C. Involvement of the Wnt/β-catenin signaling pathway in the cellular and molecular mechanisms of fibrosis in endometriosis. PLoS One. 2013;8(10):e76808.
14. Burke JP, Mulsow JJ, O'Keane C, Docherty NG, Watson RW, O'Connell PR. Fibrogenesis in Crohn's disease. Am J Gastroenterol. 2007;102(2):439-48.
15. Guglielmo FF, Anupindi SA, Fletcher JG, Al-Hawary MM, Dillman JR, Grand DJ, et al. Small Bowel Crohn Disease at CT and MR Enterography: Imaging Atlas and Glossary of Terms. Radiographics. 2020;40(2):354-75.
16. Razzi S, Luisi S, Calonaci F, Altomare A, Bocchi C, Petraglia F. Efficacy of vaginal danazol treatment in women with recurrent deeply infiltrating endometriosis. Fertil Steril. 2007;88(4):789-94.

Cite this article as: Agrawal TL, Maguire R, Pillai S, Naik AD. Endo the line: stricturing terminal ileum endometriosis. Int Surg J 2024;11:453-6.