

Case Report

DOI: <https://dx.doi.org/10.18203/2349-2902.ijssurgery20214770>

A rare case of idiopathic multiple small bowel strictures in Indian subcontinent: a case report

Garima Dwivedi*, Sanjeev Kumar Singla, Rohit Virmani

Department of Surgery, BPS GMC Khanpur Kalan, Sonipat, Haryana, India

Received: 03 October 2021

Accepted: 29 October 2021

***Correspondence:**

Dr. Garima Dwivedi,
E-mail: dwivedi.garima6@gmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Small bowel ulcers and strictures are uncommon but when they occur they can be an important cause of morbidity. We reported a case of a multiple strictures in small bowel that was treated surgically pain for 15 years. She also had hypoalbuminemia and iron deficiency anemia. A diagnosis of small bowel stricture was made on CT and surgery was advised. Resection of the 100 cm long segment of small bowel was done. Approximately 10 smooth strictures were identified in the resected segment. Histology did not identify any specific cause. A 43 year old Indian female was diagnosed with subacute small bowel obstruction. She had a history of chronic abdominal pain for 15 years. She also had hypoalbuminemia and iron deficiency anemia. A diagnosis of small bowel stricture was made on CT and surgery was advised. Resection of the 100 cm long segment of small bowel was done. Approximately 10 smooth strictures were identified in the resected segment. Histology did not identify any specific cause. Subacute small bowel obstruction can be associated with various factors. Multiple idiopathic small bowel strictures can be considered as one of the causes. In present case no cause of ulcers and strictures could be found. Idiopathic multiple bowel strictures with fecalith is rare. Histopathology and other investigations revealed no specific cause. We recorded no postoperative complications 2 months after surgery by resection and anastomosis

Keywords: Idiopathic small bowel stricture, Multiple fecaliths, Indian subcontinent

INTRODUCTION

Small bowel ulcers and strictures can occur in adults and children.¹ These can be prolonged use of NSAIDS leading to small bowel diaphragm disease as described by Lang et al.² There are many other causes also but when no specific cause is found a diagnosis of cryptogenic multifocal ulcerous stenosing enteritis can be made after excluding other diseases.¹ However management of this condition is not well established and recurrences are quite common. We reported a case of 43 year old female with prolonged course of dull aching abdominal pain, presented with subacute small intestinal obstruction without any apparent cause.³

CASE REPORT

At the time of admission a 43 year old female had a history of chronic intermittent abdominal pain and episodes of subacute intestinal obstruction for 15 years. She had complains of pain associated with vomiting, which was yellow in color and projectile in nature, that was getting relieved after medication. No history of constipation, diarrhea was present with pain. She also had pain in suprapubic region which was associated with urinary retention, burning micturition and dribbling of urine for 1 and half month before presentation.

CECT abdomen was done which showed thickening of proximal small bowel (jejunum) wall thickening approximately 9.6 mm (Figure 1). A transitional zone with narrowing was seen with intermittent dilation of

multiple jejunal loops largest measuring approximately 43 mm in left para umbilical region. Multiple calcified fecoliths were seen in small bowel loops. Multiple centimeter sized fecoliths were seen measuring 9.4 mm in short axis diameter. Minimal free fluid was seen in abdominal cavity she had Hb=5.7, total protein=4.56, albumin=2.72 and globulin=1.84. She was having microcytic hypochromic anemia with koilonychia. She was managed conservatively with blood transfusion and her anemia and hypoproteinemia was corrected.

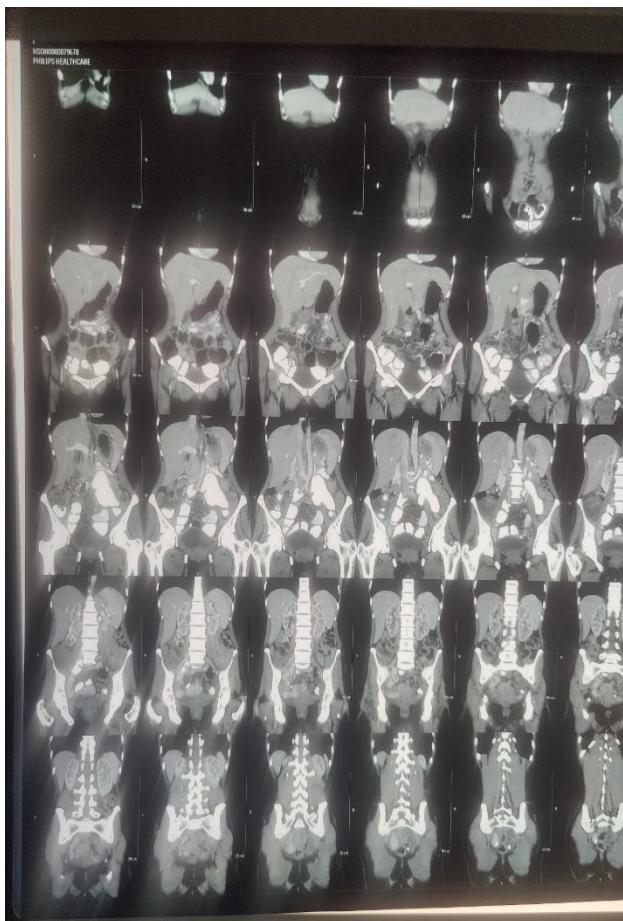


Figure 1: CE-CT of the abdomen.



Figure 2: Segment of the small bowel resected.



Figure 3: Segment of the small bowel resected with multiple fecolith and multiple stricture.

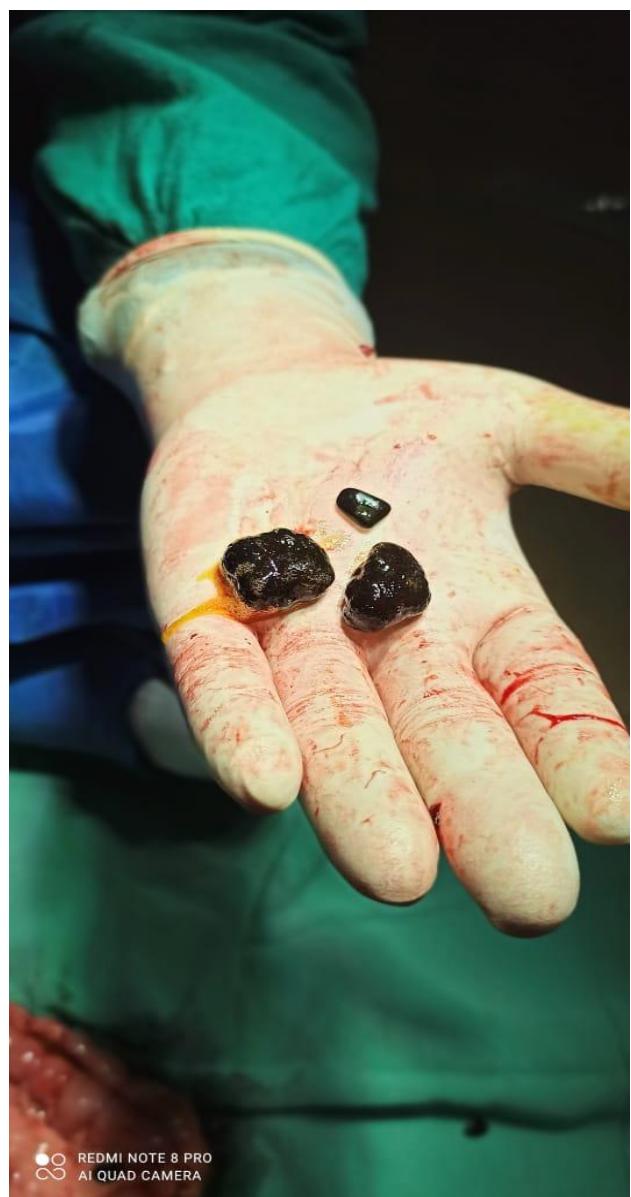


Figure 4: Extracted fecolith.

She was operated under GA and midline incision was given from xiphisternum to 5 cm above pubic symphysis. Multiple strictures from 50 cm distal to DJ to 100 cm proximal to the ileocecal junction were found. Resection and anastomosis of the remaining segment was done (Figure 2-4).

The resected specimen was sent for histopathology which showed stricture of intestine showing ulceration of mucosa in few focal areas and ischemic necrosis and collection of lymphoid aggregates causing destruction of muscularis mucosa at places. The submucosa showed areas of congestion multiple lymph nodes showing reactive hyperplasia. Postoperative period was uneventful.

On postoperative day 4 patient passed flatus and motion. On postoperative day 7 drain was removed. And alternate stitches were removed. On postoperative day 9 remaining stitches were removed and patient was discharged.

DISCUSSION

Ulcerations and isolated strictures of the small intestine are uncommon.³ CMUSE is a rare cause of strictures and ulcerations which can present as GI bleeding, small bowel obstruction, anaemia and perforation.⁴⁻⁷ These were mostly diagnosed by radiological investigations like contrast enhanced CT scan (as in our case) or macroscopic examination via capsule enteroscopy, however this capsule enteroscopy should only be advised to patients with iron deficiency anaemia.⁸⁻¹¹

Differential diagnosis of CMUSE included congenital malformations, lymphoma, carcinoma, tuberculosis, radiation enteritis, incarcerated hernias, trauma, chemical or drug irritation, vascular abnormalities, ischemia and Crohn's disease. This had been mostly reported in Mediterranean region. Some unexplainable strictures were termed as nonspecific or idiopathic. An entity included in this group, multifocal idiopathic stenosing enteritis was first described by Debray et al and consisted of the following clinicopathologic characteristics: onset in young people; iron-deficiency anaemia; recurrent, intermittent intestinal obstruction; absence of malabsorption; multiple, short annular strictures usually limited to the ileum; mucosal and submucosal ulceration involving the mesenteric margin and absence of villous atrophy between the strictures.¹² This uncommon clinical entity had been described mostly in the Mediterranean region.¹²⁻¹⁶ Many reasons have been given for nonspecific ulcers and strictures in small intestine. These explanations included vascular disease, neuropathy, infection, trauma and hormonal or autonomic influences. No evidence of infection or trauma were found in our cases. An often reported cause was focal ischemic etiology for development of strictures and ulcerations. Secondary stenosis of the small bowel can be caused by partial necrosis of small bowel. Subacute or chronic focal ischemia of the small bowel may cause the development

of collateral circulation. This could prevent a transmural infarct, but the partial necrosis of the intestinal wall could cause secondary stenosis. The fact that multifocal idiopathic stenosis had been described, mostly in Mediterranean countries, could imply a genetic or environmental component in the pathogenesis of these lesions.

CONCLUSION

In our case no specific cause could be identified. No genetic co-relations, infections or history of trauma could be recognized. Patient had severe iron deficiency anaemia and hypoproteinemia with intestinal obstruction, severe malabsorption and multiple strictures as described by Debray, that was corrected preoperatively and remained corrected after resection and anastomosis..

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Kohoutová D, Bártová J, Tachecí I, Rejchrt S, Repák R, Kopáčová M, et al. Cryptogenic multifocal ulcerous stenosing enteritis: a review of the literature. *Gastroenterol Res Pract*. 2013;2013:918031.
2. Lang J, Price AB, Levi AJ, Burke M, Gumpel JM, Bjarnason I. Diaphragm disease: pathology of disease of the small intestine induced by non-steroidal anti-inflammatory drugs. *J Clin Pathol*. 1988;41(5):516-26.
3. Raskin JB, Pina V. Nonspecific enteric ulcers and strictures. In: Bockus H, Berk JE, eds. *Gastroenterology*. 4th ed. Philadelphia: Saunders; 1985: 1268-73.
4. Chung SH, Park SU, Cheon JH, Kim ER, Byeon J, Ye BD, et al. Clinical characteristic and treatment outcomes of cryptogenic multifocal ulcerous stenosing enteritis in Korea. *Dig Dis Sci*. 2015;60(9):2740-5.
5. Esaki M, Umeno J, Kitazono T, Matsumoto T. Clinicopathologic features of chronic nonspecific multiple ulcers of small intestine. *Clin J Gastroenterol*. 2015;8(2):57-62.
6. Perlemer G, Guillemin L, Legman P, Weiss L, Couturier D, Chaussade S. Cryptogenic multifocal ulcerous stenosing enteritis: an atypical type of vasculitis or a disease mimicking vasculitis. *Gut*. 2001;48(3):333-8.
7. Matsumoto T, Iida M, Matsui T, Yao T. Chronic nonspecific multiple ulcers of the small intestine: a proposal of the entity from Japanese gastroenterologists to Western enteroscopists. *Gastrointest Endosc*. 2007;66(3):99-107.
8. Slesser AAP, Wharton R, Smith GV, Buchanan GN. Systematic review of small bowel diaphragm

disease requiring surgery. *Colorectal Dis.* 2012;14(7):804-13.

- 9. Velchuru VR, Rimal D, Studley JGN, Sturzaker HG. Diaphragm disease of the ileum. *Int J Surg.* 2006;4(3):184-6.
- 10. Manetas M, O'Loughlin C, Kelemen K, Barkin JS. Multiple small-bowel diaphragms: a cause of obscure GI bleeding diagnosed by capsule endoscopy. *Gastrointest Endosc.* 2004;60(5):848-51.
- 11. Wang ML, Miao F, Tang YH, Zhao XS, Zhong J, Yuan F. Special diaphragm-like strictures of small bowel unrelated to non-steroidal anti-inflammatory drugs. *World J Gastroenterol.* 2011;17(31):3596-604.
- 12. Debray C, Besançon F, Hardouin JP, Martin E, Marche C, Khoury K. Entérite sténosante ulcéreuse plurifocale cryptogénétique. *Arch Fr Mal App Dig.* 1964;53:193-206.
- 13. Chagnon JP, Devars JF, Marche C, Vissuzaine C, Cerf M. Cryptogenetic multifocal ulcerous stenosing enteritis. *Gastroenterol Clin Biol.* 1984;98(3):808-13.
- 14. Doutre LP, Paccalin J, Périsat J, Traissac FJ. Entérite sténosante et ulcéreuse plurifocale (une nouvelle observation). *Arch Fr Mal App Dig.* 1966;55:537-40.
- 15. Raskin JB, Pina V. Nonspecific enteric ulcers and strictures. In: Bockus H, Berk JE, eds. *Gastroenterology.* 4th ed. Philadelphia: Saunders; 1985: 1268-73.
- 16. Chornet JC, Ximénez EG, Company RA, González JCC, Aracil JS, Quintana NU, et al. Etude photopléthysmographique de la circulation pariétale du tube digestif en conditions basales et de distension. Travail expérimental chez le chien Lyon Chir. 1988;84:247-51.

Cite this article as: Dwivedi G, Singla SK, Virmani R. A rare case of idiopathic multiple small bowel strictures in Indian subcontinent: a case report. *Int Surg J* 2021;8:3710-3.