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General Surgery

A RARE CASE OF SMALL BOWEL DIAPHRAGM DISEASE PRESENTING AS IRON DEFICIENCY ANAEMIA

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ABSTRACT Here we report a case of a 12 year old male child who was being evaluated for iron deficiency anaemia and pain abdomen, and who had undergone repeated endoscopies and CT scans; was referred to Department of Surgery for management of forgotten and impacted capsule endoscope from Gastroenterology. Patient had undergone capsule endoscopy one year back for evaluation of iron deficiency anaemia, but due to technical error no images were delivered from the capsule. Patient was lost to follow-up but returned a year later with complaints of easy fatiguability and pain abdomen. A abdomen erect X-ray revealed the forgotten capsule. A mini-laparotomy with Intraoperative push enteroscopy revealed a string of short segment strictures (>10) with circumferential ulcerations involving upto 60cm of small bowel. Resection of the involved bowel segment, with retrieval of the capsule with side to side anastomosis was done. Diagnosis of diaphragm disease was further confirmed on histopathological examination of the resected bowel segment. Small bowel diaphragm disease is a rare disorder caused by chronic NSAID use and is characterised by strictures throughout the small bowel leading to luminal narrowing and occasionally even obstruction. Given its atypical presentation and rarity, it is usually missed unless the surgeon keeps an high threshold of suspicion.

KEYWORDS : capsule endoscopy, chronic anaemia, push enteroscopy, diaphragm disease

INTRODUCTION

Small bowel diaphragm disease was first identified by Lang et al.[1] in 1988 who described it as an NSAID: (non-steroidal anti-inflammatory drug) induced enteropathy, which consisted of inflammation, erosions, fibrosis, and formation of diaphragm-like strictures of the small bowel. This disease has been reported to present with many manifestations such as iron deficiency anaemia, acute gastrointestinal hemorrhage, perforated peritonitis and rarely even subacute intestinal obstruction. Most cases reported attribute chronic NSAID use to the diaphragm disease. Here, we report a case of 12 year old male child with no known history of NSAID use presenting with chronic iron deficiency anaemia, unresponsive to medical therapy.

CASE REPORT

- In 2019, 12 year old male child was referred from a peripheral health centre for work up and management of chronic iron deficiency anaemia(IDA) not responding to standard treatment.
- Patient had presented with complaints of recurrent easy fatiguability, fever, lethargy, loss of appetite. Routine blood work up was suggestive of iron deficiency anaemia with a hemoglobin (Hb) of 4g/dL.
- Detailed blood work-up for anaemia including hemoglobin electrophoresis and a peripheral smear returned normal.
- Patient was given blood transfusions after which Hb improved to 9 g/dL and patient reported resolution of symptoms.
- Further evaluated with CECT abdomen, upper esophagogastroduodenoscopy, & colonoscopy - no significant abnormality detected.
- 2020: patient presented with recurrent complaints - blood work-up again suggestive of fall in Hb and IDA. Blood transfusions given. Repeat upper GI endoscopy was suggestive of duodenal D1 and D2 segment stricture - CRE dilatation was done.
- 2021 : patient returned yet again with similar complaints with documented fall in Hb. After stabilisation, capsule endoscopy was planned. Due to technical errors, no images could be retrieved from the device. Patient was discharged and asked to follow-up for a repeat capsule endoscopy. Patient was lost to follow up thereafter.
- 2022: Patient returned with same complaints of easily fatiguability, lethargy and loss of appetite and also complaining of mild pain abdomen. An X-ray abdomen erect done which revealed the forgotten and impacted capsule endoscope. Patient was then referred to department of Surgery for management of the impacted capsule.
- In discussion with medical gastroenterologists, a plan was made for mini-laparotomy and intraoperative push enteroscopy.
- Intraoperative findings were as follows:

1. 80cm from ileocaecal junction(ICJ) capsule endoscope was found impacted within a submucosal tunnel.

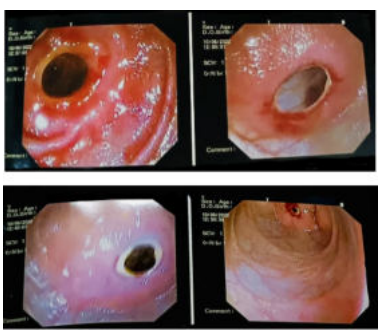

Figure 1a & 1b : Push enteroscopy images demonstrating the classic "diaphragms"; multiple, mucosal membrane like ulceration at the luminal margin.

2. Externally, small bowel loops were normal without any evidence of strictures or adhesions.
3. Multiple lumen compromising strictures (>10 in number) found in ileum and jejunum - starting from 110cm from DJ flexure to up to 60cm from ICJ. Figure 1a &1b
3. Terminal 60cm of ileum, ICJ, caecum, and rest of the colon appeared to be normal.

- 60cm of the affected small bowel loop, worst affected segment where severe ulcerations were noted and scope was passed with difficulty, resected and side-to-side anastomosis done in 4 layers.

Resected specimen was sent for histopathological examination.

- Patient had an uneventful recovery and was discharged on post-operative day 8.
- HPE of the respected specimen confirmed the diagnosis of diaphragm disease.
- Patient was regularly followed upto 1 month post-op and again later at 6 months post-op : was complaints free and repeat complete blood counter was normal.

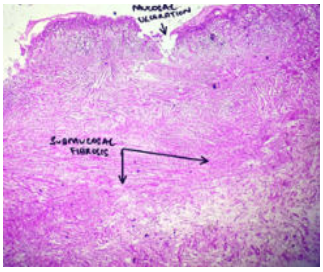

Figure 2 : Histopathological image (H&E stain) of the ileal diaphragm.

DISCUSSION:

Small bowel diaphragm disease is an under diagnosed entity that can present in a variety of ways. CT abdomen, laparoscopy and other radiological modalities usually remain inconclusive[2]. Capsule endoscopy however clinches the diagnosis pre-operatively. Most commonly diagnosis is established retrospectively on histopathological examination of the tissue specimen. HPE characterised by : thin circumferential membranes resembling plica circularis,[1]composed of mucosa and submucosal with accompanying fibrosis, mucosal ulcerations at the diaphragm segment, with villous atrophy and eosinophilic enteritis. [3] Figure 2 Although most cases are found to be associated with chronic NSAID use, there are reported cases of diaphragm disease where there no history of NSAID use[4]. Treatment modalities include - withdrawal of the offending NSAID, surgical resection, stricturoplasty, double balloon enteroscopy. High index of suspicion must be maintained by surgeons in patients presenting with vague gastrointestinal symptoms with inconclusive radiological studies to avoid mis-diagnosing patients.

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