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Case Report

A rare case of isolated duodenal and proximal jejunal Crohn's disease – Learning experiences from the challenges faced in diagnosis and management

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Introduction

Crohn's disease (CD) is an idiopathic, chronic inflammatory bowel disease characterised by transmural inflammation of the bowel wall with inflammatory, perforating or stricturing disease phenotypes. Any part of the gastrointestinal tract, from mouth to anus, can be affected by CD with the terminal ileum and colon being the commonest. Isolated gastroduodenal CD is extremely rare with an incidence of less than 0.07% of all patients with CD [1]. Here we present a case of isolated duodenal and proximal jejunal CD which posed difficulties in diagnosis and management.

Clinical presentation

A 42-year-old female presented with progressive upper abdominal pain, vomiting and low-grade fever of 2 months duration. Constitutional symptoms, such as loss of appetite and malaise, were noted. There was no alteration of bowel habits. She had no past history of previous bowel disease. The symptoms were progressive despite numerous hospital visits and symptomatic management. She presented to our unit as an emergency with severe abdominal pain and vomiting.

On admission, the patient was febrile, dehydrated and tachycardic. On abdominal examination, tenderness and guarding were noted in the epigastrium and left hypochondrium. Her blood pressure and urine output were normal. The blood panel showed an elevated white cell count of $32x10^3$/µL, haemoglobin of 7.8g/dL and elevated CRP of 224IU. Blood cultures were negative. A contrast enhanced CT was performed and the 3rd and 4th parts of duodenum (D3, D4), DJ flexure and proximal jejunum were noted

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to be grossly inflamed and thickened. Inflammatory fluid was noted between the bowel loops and the mesentery appeared thickened and inflamed with multiple enlarged mesenteric lymph nodes. There was no evidence of bowel gangrene or pneumatosis. Radiological imaging was suggestive of either inflammatory bowel disease, infective enteritis or lymphoma of small bowel. We proceeded with enteroscopy and the D3 and D4 segments of the duodenum were noted to be grossly inflamed with haemorrhagic and necrotic areas on the mucosa (Figure 1). A biopsy obtained from the duodenal mucosa demonstrated a severely inflamed mucosa with evidence of focal ischaemia but no evidence of malignancy was detected. The faecal calprotectin level was markedly elevated.

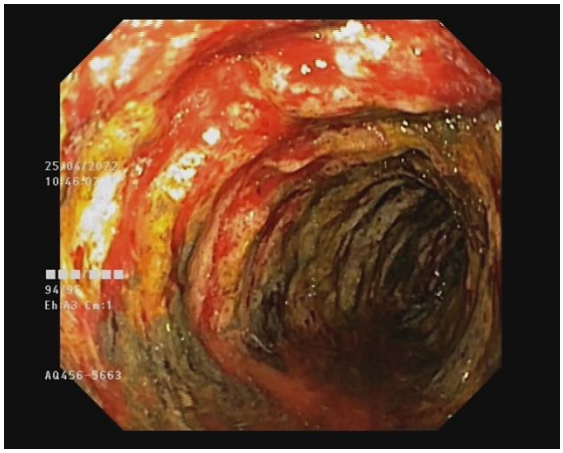

Figure 1: Enteroscopic view of the inflamed duodenum

With no proper diagnosis as yet, the patient was initially managed conservatively with fluid resuscitation, intravenous antibiotics and parenteral nutrition. Vital parameters and lactate levels were regularly monitored for evidence of bowel ischaemia and perforation. Despite conservative measures the patient failed to show improvement and an explorative laparotomy was performed. Intraoperatively the distal duodenum, DJ flexure and about 15cm of the proximal jejunum were found to be grossly inflamed and thickened (Figure 2). A few skip lesions were noted in the proximal jejunum, distally. The oedematous and thickened mesentery showed a creeping fat appearance and contained multiple enlarged lymph nodes. The clinical appearance of the bowel was suggestive of CD. There was no evidence of abscess formation or bowel gangrene.

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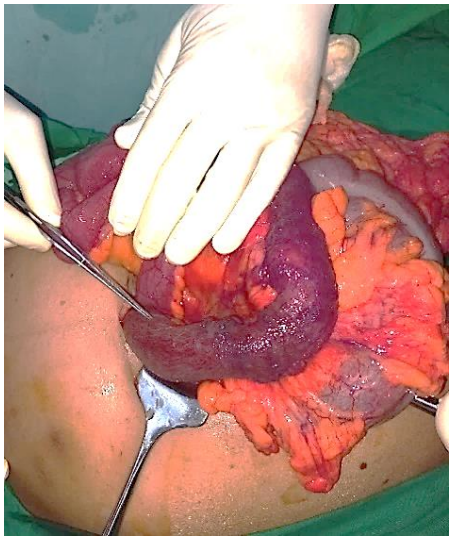

Figure 2: Grossly inflamed thickened proximal jejunum

Following thorough peritoneal survey, the surgical options were considered. After considering multiple factors including current physiological status and safety, we decided to biopsy multiple mesenteric lymph nodes and create a retro colic, iso-peristaltic gastro-jejunostomy to bypass the affected bowel segment. Gastric contents would now pass from the stomach into the distal small bowel bypassing the inflamed segment. Direct biopsy from the affected bowel segment was impossible due to the high risk of leakage.

The postoperative period was complicated and prolonged. In the early postoperative period, the patient developed high fever spikes and persistent abdominal pain. This was attributed to the diseased bowel segment and a flare up of the underlying disease. A CECT was repeated to assess for complications but there was no evidence of intrabdominal abscess formation or disease progression. The patient was continued on antibiotic therapy and symptomatic management. The decision to start immunosuppressive therapy was delayed until the biopsy reports were available. However, lymph node biopsy turned up to be inconclusive with non-specific inflammatory cells and lymphoid aggregates with no evidence of malignancy or lymphoma. The characteristic granuloma formation of CD was absent. The patient was discussed at a multi-disciplinary board with input from the gastroenterologist and was managed as inflammatory bowel disease.

Despite the drainage procedure (gastro-jejunostomy) the patient experienced recurrent episodes of bilious vomiting and high nasograstric drainage. Enteral feeding was significantly impaired due to this and the patient had to be restarted on parenteral nutrition. A gastrograffin study and upper GI endoscopy were performed to assess patency of the anastomosis and gastric emptying. There was no evidence of obstruction at the level of the anastomosis but poor gastric emptying with stasis was noted. Both the proximal and distal limbs of the gastro-jejunostomy were patent. A trial of intravenous

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neostigmine was attempted to counter the gastric stasis, but the response was temporary. The patient was managed with long-term intravenous metoclopramide and NG drainage with intermittent clamping while parenteral nutrition was continued. Continuation of parenteral nutrition in this patient was complicated with line sepsis, fluid overload and electrolyte imbalances.

While on medical management, the patient's symptoms gradually resolved over a period of 4 weeks and oral feeding was re-established. The patient was discharged after a total stay of 5 weeks. Follow up plan includes re-evaluation of symptoms and optimization of medical management.

Discussion

Numerous challenges were encountered in the management of this patient. This acutely ill patient required simultaneous diagnostic workup, resuscitation and timely therapeutic intervention. Therapeutic interventions could not be delayed until a definitive diagnosis was made. In this scenario we were unable to establish a diagnosis preoperatively. Radiological imaging, endoscopy findings and metabolic workup were in favor of an acute exacerbation of inflammatory bowel disease but there was no histological confirmation. The biopsies we obtained returned as negative for both Crohns and malignancy. However, characteristic histological features of CD is not always seen and is not mandatory for diagnosis. The characteristic non-caseating granulomas are only seen in 60% of all Crohn's patients [2]. The possibility of immune-mediated disease or vasculitis was ruled out by screening for immune markers such as ANCA, ds-DNA etc.

Another learning experience was the importance of considering multiple factors intraoperatively prior to surgical decision making. Intraoperative findings were more in favor of CD, and we considered several factors before deciding our surgical approach. Available surgical options were either resection of the affected segment or a staged surgical intervention with a bypass procedure and relevant biopsies. Excision of the affected segment required a major surgical procedure (a Whipple's procedure) which would not have been tolerated by an already physiologically unstable patient. In addition, a surgical complication (e.g. anastomotic leak) in this setting would have drastically increased the mortality risk of the patient. Also, in CD surgical resections are mainly reserved for complications while medical management plays a larger role.

However, resection of the affected bowel segment would have reduced the disease burden and would have reduced postoperative morbidity. Analysis of the resected specimen would also have established the diagnosis. In this patient, we opted for a safer option with biopsy and a bypass procedure. Even though this contributed to postoperative morbidity and prolonged hospital stay, mortality was prevented. Another learning point was the importance of taking adequate representative biopsies to avoid diagnostic difficulties.

Prolonged postoperative gastric stasis despite a patent anastomosis posed another challenge postoperatively. Delayed-return gastric emptying (DRGE) is a well-known

phenomenon after gastrojejunostomy. A literature review revealed that factors such as age over 60, non-gastric resection, intraoperative vagotomy and malnutrition are risk factors for delayed return of gastric function [3]. Majority of these cases improve overtime. The current recommendation for management of DREG is conservative since reoperation is associated with further worsening of gastroparesis [4].

Postoperative gastric stasis in this patient severely affected enteral feeding and nutrition. Enteral nutrition is always preferred over parenteral nutrition. Enteral feeding is associated with faster patient recovery and protects gut mucosal integrity while avoiding the complications of parenteral nutrition. In retrospect, placement of a feeding jejunostomy in this patient during the primary surgery would have improved postoperative nutrition.

Learning points

- Presentation of Crohn's disease is varied and it may present as an acute abdomen.
- Mainstay of management of Crohn's disease is medical. Surgical intervention is limited to the management of complications.
- Avoid malnutrition in acutely ill patients. Ensure that necessary steps are taken to continue or supplement enteral feeding.
- When faced with a challenging situation during surgery, consider multiple factors including patient stability and safety. Take time to plan the surgical approach and obtain a second opinion if needed.

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