Rare presentation of primary coloduodenal fistula

Veronika Pronisceva1 • Nipin Bagla2 • Siddappa Lakshmaiah1 • Elizabeth Sharp1

1QEQM Hospital – General Surgery, Margate CT9 4AN, UK
2William Harvey Hospital – Histopathology Department, Ashford, Kent TN24 9NB, UK
Correspondence to: Veronika Pronisceva. Email: vpronisceva@yahoo.com

Primary benign coloduodenal fistula is rare. We report a single case where a patient underwent emergency laparotomy and made complete recovery.

Case presentation

We report a case involving a 54-year-old female office administrator, who presented to A&E with a 5-day history of multiple episodes of vomiting (in the last 24 hours it had become feculent), upper abdominal pain, tenderness and generally being unwell. On clinical examination, patient had epigastric and right upper quadrant abdominal pain and tenderness. The abdomen was not distended. Blood results showed haemoglobin of 15.4 g/dl, White cell count of 11.7 10^9/l, Neutrophils of 9.4 10^9/l and a Calcium of 2.7 mmol/l. C-reactive protein and the rest of the biochemical profile were normal. An abdominal film did not reveal any obvious pathology (Images 1 and 2).

Patient had initially presented to her GP with a history of vague abdominal pain, diarrhoea and vomiting. Her symptoms were attributed to stress at work and depression. She was started on PPI and anti-sickness medication. As the symptoms did not settle and the patient developed right upper quadrant pain, she was investigated with Ultrasound (US), diagnosed with gall stone disease and was referred to the general surgical clinic for consideration of laparoscopic cholecystectomy.

Her past medical history included investigation for endometriosis and treatment for depression. She had a family history of scleroderma (two 1st degree relatives), breast cancer (1st and 2nd degree relatives), Raynaud’s disease (two 1st degree relatives) and multiple sclerosis.

While she was waiting for a clinic appointment, she was admitted to the hospital with acute cholecystitis and had conservative treatment. At that time she had raised inflammatory markers and US showed thickened gall bladder containing stones and nothing else. She was discharged and placed on waiting list for a cholecystectomy.

During her admission a history of weight loss and lethargy was noted. The patient was referred for outpatient computer tomography (CT) examination to exclude malignancy. CT reported thickening of the wall of the hepatic flexure and transverse colon suggestive of colitis, but its exact significance was uncertain.

A colonoscopy was organized with a view to obtaining a definite diagnosis, but the patient was acutely unwell again and had to be admitted. By this time it was more than 6 months since the initial symptoms began. Patient was monitored and treated with intravenous fluids, nasogastric tube, input and output monitoring. After 24 hours her condition was found to deteriorate.

Based on results of recent CT scan, our first differential diagnosis was an obstructive upper Gastrointestinal (GI) neoplasm. It was decided not to have further preoperative CT scan as it would not add additional diagnostic value but only delay definitive treatment. Patient was taken for surgery.

Intraoperative findings were a large inflammatory mass in right upper quadrant area which involved the proximal transverse colon, third part of duodenum, and part of the liver medial to the gall bladder bed. Dissection revealed a...
coloduodenal fistula. A limited right hemicolectomy was performed and the duodenal defect was repaired and over sewn with an omental patch. A Robinson’s drain was placed in the sub hepatic area, and nasogastric and nasojejunal tubes were sited. Patient had a small anastomotic leak on the CT scan on the 4th postoperative day, and it was treated conservatively with total parenteral nutrition and subsequently recovery was uneventful.

Examination of the right hemicolectomy specimen by pathologists revealed thickening and stenosis of the distal 75 mm. This corresponds to the bowel wall thickening seen by CT scan at the hepatic flexure. The wall thickening was due to hypertrophy of the muscularis propria, increased mural lymphoid aggregates and muscularization of the submucosa. The mucosal lining in the narrowed segment showed mild distortion of the crypt architecture with formation of hypertrophic mucosal islands (pseudopolyps) and crypt abscesses. The small and large bowel mucosa away from the stenosed segment was normal. There were no granulomata.

We followed the patient up for 2 years after surgery, but no definite histological or clinical diagnosis of inflammatory bowel or other disease was identified. Follow-up endoscopy (oesophagogastroscopy and colonoscopy) results come back normal.

**Discussion**

The causes of benign coloduodenal fistulae could be varied: malignancy, duodenal ulcers, Crohn’s disease, Ulcerative colitis, infective (tuberculosis, typhoid), parasites (amoebiasis, hydatid), foreign body, duodenal diverticulum, colonic diverticulum, spontaneous, iatrogenic or appendicitis.1,4,5 Acute presentation of a primary benign coloduodenal fistula is a very rare pathology in surgical practice and could be misdiagnosed initially. In our case, immediate diagnosis was difficult. This patient’s initial management was complicated by diagnosis of acute cholecystitis. When we trawled through the past medical history and reviewed symptoms in retrospect, we raised theories that the patient may have had an
undiagnosed duodenal ulcer, bowel inflammatory disease, diverticula of the transverse colon or a congenital abnormality.

The pathological changes described above indicate chronic repeated bouts of inflammatory injury to the colonic mucosa adjacent to fistula with healing intervals, manifested in the form of hypertrophic mucosal islands. This is usually attributed to chronic idiopathic inflammatory bowel disease if no primary cause for the repeated chronic inflammation is found. If we assume this chronic inflammation to be purely idiopathic, the pathological features favour Crohn’s disease over ulcerative colitis because: (a) The small and large bowel away from the narrowed segment was normal indicating segmental colitis, (b) Distal colonic biopsies without steroid treatment (after six months) showed normal mucosa, and (c) the patient presented with fistula indicating deep transmural inflammation (Images 3 and 4).

On the other hand, it is quite possible that the fistula had been there for some time with intermittent small leakage of duodenal contents into the colon causing repeated bouts of mucosal injury and repair simulating the pathological features of Crohn’s disease. In this context the segmental colitis would be a consequence rather than the cause of fistula. In absence of any previous history or diagnosis of duodenal ulcer, it is difficult to confirm this theory. However, the omental patch was successful in sealing the duodenal defect; and the patient has been symptom free for the past two years without steroid treatment. Therefore, this is less likely to be Crohn’s disease and more likely to be a previously undiagnosed deep duodenal ulcer presenting as coloduodenal fistula and chronic segmental colitis.

Ideally it would have been very useful to have had gastroscopy or contrast studies prior to the emergency surgery; but firstly there was no clinical suspicion of the diagnosis of a benign fistula and secondly the patient was too sick and would not tolerate procedures.

In retrospect, review of the post contrast CT scan showed short segments of inflamed duodenum and transverse colons. There were no malignant appearing mass lesions in these regions. The serosa of opposing duodenum and transverse colons appeared tethered and pulled towards each other. Furthermore, a small extra luminal gas pocket could be seen between these segments. This is a nonspecific finding which may be seen with local perforation but, in patients with typical history, could suggest possible fistula. In this case to report a coloduodenal fistula without primary clinical suspicion would be challenging. In summary, primary coloduodenal fistulas are rare entities. Its diagnosis is difficult but should be suspected if the opposing segments appear inflamed. The presence of extra luminal gas,
although nonspecific, is particularly relevant for the diagnosis if the patient has feculent vomit (Images 5–9).

Very few case reports are published about benign spontaneous/primary coloduodenal fistulae and those which are, usually present with sub-acute symptoms and longstanding aetiology (such as inflammatory bowel disease/Duodenal ulcers), and there is opportunity for
investigations prior to surgical treatment. Most coloduodenal fistulae are treated on an elective basis and there are no guidelines or recommendations regarding surgical treatment of primary Coloduodenal fistulae on an acute basis. That is why every case is unique in management experience. Generally, treatment options could vary from conservative to operative treatment. In this particular patient we were not able to effect conservative or elective treatment. A literature search showed that high pressure in the colon causes large bowel contents to go rapidly into the duodenum via the fistula, where pressure is low. It resulted in initial profuse faecal vomiting and uncontrolled faecal output via nasogastric tube. Very high output led to electrolyte imbalance prompting emergency surgical treatment.

Based on our theory and histological suspicion of possible bowel inflammatory disease, the patient was referred to the gastroenterologist for follow-up and future investigations. After 2 years of close monitoring our patient was discharged and cleared from diagnosis/suspicion of bowel inflammatory disease, leaving us with the only diagnosis of a primary coloduodenal fistula with a debatable cause.

Learning points from this case report:

1. Treat a patient not X-ray, even if it is normal.
2. It is very unlikely to have upper GI obstruction with frank faecal output and flat abdomen. So if symptoms do not go with provisional diagnosis think again;
3. Do not request unnecessary imaging if it would not change your management. It can delay surgery possibly making the patient unfit for any procedure. Trust your clinical judgement and experience;
4. Quite often patients blame their abdominal pain or sickness on a stressful job or depression. Do not let that mislead you and interpret symptoms incorrectly;
5. It is very difficult to diagnose pathology with nonspecific symptoms at an early stage, especially with co-diagnosis of cholecystitis. If you encounter a case of primary coloduodenal fistula on a laparotomy for nonspecific symptoms, then our case is an example to learn from. Despite availability of a variety of diagnostic tools and imaging, definite preoperative diagnosis can be uncertain. Operative experience and technical skills are irreplaceable.

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