BENIGN DUODENO-COLIC FISTULAE

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In 1857 Dr. Charles Murchison,¹⁵ in a review of alimentary fistulae, quoted a case in which 'the duodenum communicated with the colon through the gall bladder, the nature of the disease not being stated'. However, the first fully documented case of benign duodeno-colic fistula which we have been able to trace was reported by Sanderson in 1863.²⁸ The majority of the reported fistulae between the duodenum and colon have been malignant and benign duodeno-colic fistulae are rare. In a review of the literature we have been able to find only 25 cases (see table). There are some other cases in the literature which we have omitted from the table for the following reasons. We have not included the case reported by Blondeau¹ (quoted by Clayton and Thornton²), as it was thought to be malignant. It would seem that the case reported by Krock⁶ is the same as that of Olson.¹⁹ Garland and Wyatt⁴ reported a case following surgery for pancreatitis, but the site of the fistula would seem to be in doubt. Judd and Burden⁵ reported two cases of internal biliary fistulae where the duodenum connected through the gall bladder to the colon, but we are not including these two cases, as they were not fully documented. For a similar reason we are not including the two separate cases mentioned by Lyons⁹ and Crill³ in the discussion following the case report of Olson.¹⁹ We describe below two further cases of this uncommon condition and discuss the clinical features and diagnosis.

Case 1

The patient, a 40-year-old male clerk, was admitted on 19.9.58 from the out-patient clinic with a history of diarrhoea and abdominal pain. Two and a half years previously he had had intermittent lower abdominal pain with some looseness of the stools lasting for about a week. Six months later he had a second episode, also lasting for a week. Four months before he presented at the clinic he had a further attack of severe colicky lower abdominal pain, eased by passing flatus and associated with a feeling of abdominal distension. At first he had a normal, formed daily motion, but later the stools became fluid, although only once per day. During this illness he lost 20 lb. (9.1 kg.) in weight

and had a poor appetite. After five weeks his symptoms subsided until three weeks before he was admitted when, following a meal of mussels, he had a recurrence of the lower abdominal pain and his daily stool became loose. On no occasion had the stool been more frequent than once daily and he had noticed no blood. Two weeks prior to admission he vomited some brown fluid on two occasions and once a small clot of blood. Eight days before admission his stools became formed, three days later his abdominal pain ceased, and for four days he had no bowel action. During this last attack he lost a further 20 lb. (9.1 kg.) in weight. He had at no time suffered from dyspepsia.

Previous History. No serious illnesses.

On examination he was an ill-looking man, who had obviously lost weight. His tongue was furred and his mucous membranes were slightly pale. His blood pressure was 115/65 and there were no cardiac, respiratory or neurological abnormalities. There was no abnormal finding on abdominal examination. Rectal examination was normal. Sigmoidoscopy on 22.9.58 showed a rather dry, granular mucosa which was unduly reddened, but not friable. It was thought that the appearances were abnormal, but that they were not typical of ulcerative colitis. A mucosal biopsy was taken from the sigmoid colon. On histological examination this revealed an excess of inflammatory cells, but no abnormality of the glandular pattern.

Laboratory Investigations. Haemoglobin 12.5 g. per 100 ml.; mean corpuscular haemoglobin concentration 29.7%; blood film, normochromia and some anisocytosis; white cell count 13,000 per mm.³ with 78% polymorphonuclear neutrophil leucocytes; E.S.R. (Wintrobe) 45 mm. in the first hour; stool culture, no pathogens isolated.

Radiology. On 26.9.58 a barium enema examination demonstrated a fistula between the colon and the upper alimentary tract. There was no other colonic lesion and, in particular, no radiological evidence of ulcerative colitis. A chest radiograph was normal. On 1.10.58 a barium meal examination showed no abnormality of the oesophagus or stomach. The duodenal cap was deformed and an ulcer was present in the second part of the duodenum with kinking of the duodenum and some hold-up of barium at this site. The remainder of the duodenum emptied rapidly and showed a jagged contour. No fistula was seen at this examination. On 6.10.58 a repeat barium enema examination demonstrated a fistula between the region of the ulcer in the second part of the duodenum and the proximal part of the transverse colon (Fig. 1).

Course. For the first three days the patient had a low-grade pyrexia of 99.6°F. (37.6°C.) and had no bowel action. Thereafter he was afebrile and had a daily formed motion.

After preliminary phthalyl-sulphathiazole and oral streptomycin, a laparotomy was performed on 7.10.58

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FIG. 1.—Case 1. Barium enema examination showing filling of the duodenal cap and the distal part of the stomach from the colon.

by Mr. P. H. R. Ghey through a right paramedian incision. The hepatic flexure of the colon was bound to the duodenum by many adhesions. Dissection disclosed a fistulous track from the second part of the duodenum to the region of the proximal transverse colon. This was divided and the colonic opening closed. A Polya gastrectomy was then performed with closure of the duodenal stump just distal to the ulcer.

The post-operative course was uneventful and the patient is now symptom-free and has regained his normal weight.

Case 2

The patient, a farmer aged 33 years, attended the out-patients' department on 30.6.55, complaining of diarrhoea for seven weeks, peri-umbilical pain on defaecation and loss of weight. He had seven or eight pale motions at night but not by day and there was no slime or blood present in the stool. There was no vomiting. He had lost a lot of weight but was uncertain of the amount. During the year preceding his attendance at clinic he had had several attacks of abdominal pain with slight tenderness in the umbilical region and was admitted to another hospital during one such attack, being diagnosed as a case of colitis. At no time during these attacks was he pyrexial. Five months previously

FIG. 2.—Case 2. Barium enema examination outlining the obstructive lesion in the proximal transverse colon.

he was treated for a lumbar disc lesion and in February 1955, he had an acute attack of dysentery when *Shigella Sonne* was isolated from his stools. It was subsequently proved that his whole family were also infected but were symptom-free. Since then, two negative stool cultures had been obtained from the patient.

Previous History. No serious illnesses.

On examination the patient was thin, wasted and definitely ill with some distension of the lower abdomen. Peristalsis was active. No anaemia. B.P. 120/80. Rectal examination was normal and sigmoidoscopy was not performed. The stools were clay coloured.

He was admitted to hospital for investigation. A barium enema on 15.7.55 showed an obstructive lesion in the proximal transverse colon (Fig. 2).

At operation on 21.7.55 the surgeon, Mr. J. F. R. Withycombe, found a large mass of inflammatory tissue involving the caecum and ascending colon with a fistulous connection between the duodenum and the region of the hepatic flexure. The fistula was closed and a right hemicolectomy was performed with anastomosis of the terminal ileum to the mid-transverse colon. Following the operation he did well until the tenth post-operative day when he developed a subcutaneous abscess

Author	Sex	Age	Aetiology	Clinical Features				Method of Diagnosis	Site of Fistula		
				Pain	Vom.	Diarrhoea	Wt. Loss		Part of Duodenum	Colon	
Sanderson	1863	M	30	Duodenal ulcer	+	++ (faecal)	—	?	Autopsy	First	Transverse
Rees	1933	F	62	? Typhoid ulcer	+	+	—	+	Operation	Second	Transverse
Ormandy and Borgen	1939	M	31	Ulcerative colitis	+	+	++	++	Autopsy	First	Transverse
McPeak	1940	M	58	Duodenal ulcer	—	+	++	+	Barium enema	First	Hepatic flexure
McPeak	1940	M	46	Duodenal ulcer	+	—	++	+	Barium enema	Second/ Third	Transverse
McClinton	1944	M	47	Duodenal ulcer	+	+	+	+	Barium enema	Second	Transverse
Lovell	1947	M	55	Cholecystitis	+	+	?	+	Barium enema	First	Transverse
Masters	1948	M	40	Regional ileitis	+	—	++	++	Operation	Second	Transverse
Railton	1948	F	45	Duodenal ulcer	+	+	—	?	Barium meal	First	Transverse
Ogilvie	1950	F	52	Tuberculous gland	—	—	++	++	Barium meal	Third	Ascending
Ogilvie	1950	M	54	Tuberculous gland	—	+	++	++	Barium enema	Third	Ascending
Rife	1951	M	45	Duodenal ulcer	—	—	++	++	Barium meal and enema	Second	Transverse
Ransom	1951	M	28	Ulcerative colitis	—	—	+	+	Barium enema	Unknown	Transverse
Ransom	1951	M	12	Ulcerative colitis	?	—	+	+	Barium enema	Unknown	Transverse
Winfield	1951	M	?	?	—	—	+	?	Barium enema	Third	Sigmoid
Olson	1951	F	68	? Duodenal ulcer	+	+	+	++	Barium meal and enema	Second	Hepatic flexure
Clayton and Thornton	1953	M	48	Appendicitis	+	—	++	+	Barium meal and enema	Second/ Third	Caecum
Neville	1954	M	?	Cholecystitis and gall stones	+	+	—	—	Barium enema	First	Hepatic flexure
Rosenqvist and Sjöberg	1955	F	26	? Foreign body	—	+	++	+	Barium enema	Second/ Third	Transverse
Rosenqvist and Sjöberg	1955	M	61	? Tuberculous gland	+	—	++	+	Barium enema	First	Transverse
Nash and Daland	1956	M	39	Cholecystitis and gall stones	+	+	—	+	Operation	Second	Hepatic flexure
Pautler	1958	M	72	Duodenal ulcer	?	—	++	++	Barium enema	First	Transverse
Michell	1959	M	43	?	—	—	+	?	Barium enema	Second	Transverse
Own case 1		M	40	Duodenal ulcer	+	—	++	+	Barium enema	Second	Transverse
Own case 2		M	33	Regional ileitis	+	—	++	+	Operation	?	Hepatic flexure

TABLE 1.
DETAILS OF REPORTED CASES OF DUODENO-COLIC FISTULAE

which was drained satisfactorily. He was discharged from hospital a week later, apyrexial, gaining weight and feeling very well. He has had no further complications.

Pathology Report (Dr. R. A. Parker). 'The specimen included caecum, appendix, terminal ileum and the proximal 16 cm. of colon. Six cm. from the ileo-caecal valve, in the narrowed segment, there was considerable thickening of the bowel wall apparently due to submucous fibrosis, without complete destruction of the muscularis mucosae. Outside the bowel wall in this region a certain amount of fibrosis was present in the mesocolon. Similar submucosal thickening was present in the proximal 2 cm. of the appendix and in the distal 4.5 cm. of the ileum. A number of superficial ulcers were also present in the ileum, caecum, appendix and ascending colon with shelving edges and with necrotic, haemorrhagic floors formed by the submucous layer. One such ulcer was situated in the narrowed segment mentioned above and led to a fistula which communicated with the duodenum. The histological appearances were those of a chronic inflammatory lesion, composed mainly of follicular aggregations of lymphocytes with centres composed of epithelial cells'. The lesion was diagnosed as regional ileitis.

Clinical Features

Aetiology

It will be seen from Table 1 that 20 of the reported cases of benign duodeno-colic fistulae occurred in men, and five in women. Such fistulae

can occur at any age ranging in the above reported cases from 12 to 72 years.

As might be expected, duodenal ulceration is the commonest cause of the fistulae, being the cause in nine of the above reported cases and a possible cause in two others. Cholecystitis and ulcerative colitis have each been reported in three cases, and regional ileitis in a further two. Tuberculous glands, typhoid ulceration, appendicitis, duodenal diverticulum and foreign bodies have been postulated as other causes.

Symptomatology

The typical features of duodeno-colic fistulae include diarrhoea, weight loss, vomiting and abdominal pain. Diarrhoea may be gross and was prominent in 19 out of the 25 reported cases. Understandably, blood was present with the stools in two of the cases of ulcerative colitis, but it was also noted in two other cases where the aetiology was not due to colonic disease. This point is discussed later. In such fistulae it might be expected that the stool would contain undigested food, but this has not been reported.

Weight loss may be severe, ranging from 11 lb.

(5 kg.) to 84 lb. (38.2 kg.) in 20 out of the 25 above cases.

Vomiting was not so frequent, being reported in only 12 cases, but when the vomitus was faecal, as it was in half of these cases, it was a useful diagnostic point. Pain was rather non-specific and seemed in most cases to depend on the cause of the fistula.

Diagnosis

In our first case the provisional diagnosis was that of ulcerative colitis, although the weight loss seemed out of proportion to the severity of the diarrhoea. Although the sigmoidoscopic appearances were abnormal, they were not quite typical of this condition. A mucosal biopsy was taken which showed inflammation, but no diagnostic features of ulcerative colitis. Barium enema showed a duodeno-colic fistula, although this was not demonstrated by a barium meal examination.

In our second case an inflammatory intestinal lesion was suspected on clinical grounds. An obstructive lesion in the ascending colon was shown on barium enema, but the duodeno-colic fistula was not revealed. In this case the diagnosis was made at operation.

In the 25 reported cases, as shown in the table, 19 were diagnosed by barium examination. Of the others, four of the fistulae were demonstrated first at operation and the remaining two at autopsy.

The fistula was demonstrated by barium enema examination in 17 (85%) of the 20 cases where it was performed, whereas 16 barium meal examinations showed the fistulae in only six (37.5%) cases. On no occasion did a barium meal examination reveal a fistula which a barium enema had failed to show. These results confirm the contention of Medhurst¹³ that the most useful diagnostic method is barium enema examination. This is probably due to the higher pressure gradient that obtains between the two viscera on barium enema examination.

Treatment

Operative treatment was performed in 22 out of 25 cases with only one post-operative death, due to a pelvic abscess. Operative procedures included simple ligation or excision of the fistula, sometimes combined with partial or total colectomy or with gastro-enterostomy or partial gastrectomy. At the time of the original reports the 21 patients were in good health. Death ensued in the three patients where operation was not performed.

Discussion

Diarrhoea and loss of weight are often the presenting complaints in patients with fistulae between the upper alimentary tract and the colon.

Until recent years the explanation of these features was assumed to be the short-circuiting of food and gastric secretions into the colon. Convincing evidence, however, has been presented by several authors that the diarrhoea is due to retrograde passage of colonic contents through the fistula with resultant contamination of the small bowel. Lowden⁸ reports an inflammatory jejunitis and steatorrhoea in cases of gastro-jejunocolic fistulae. Rosenqvist and Sjoberg²⁶ also believe that the clinical features of diarrhoea and malabsorption in such fistulae are due to jejunitis with resultant rapid passage of food through the small bowel.

In our first case, however, there was histological inflammation of the recto-sigmoid mucosa. Whilst accepting that the main cause of the diarrhoea in duodeno-colic fistulae is retrograde passage of faeces through the fistulae with resultant jejunitis, we feel that sometimes the pressure in the duodenum may exceed that of the colon, causing intermittent flow of acid gastric juice into the colon with resultant colitis and that this may contribute to the diarrhoea. In two other reported cases, without colonic disease, where blood was present in the stool, it may be that there was also some acid-induced colitis.

Summary

The literature of benign duodeno-colic fistulae reviewed and two further cases are added. This condition is rare, the commonest cause being duodenal ulceration. The patient usually complains of diarrhoea with associated weight loss, which may be severe. The method of choice of diagnosis is by barium enema. The condition is usually curable by surgery. The mechanism of the diarrhoea in this condition is discussed.

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normal, and this symptom cannot be attributed to continuing glandular fever. Cases 8 and 9 have been included to show the dramatic effect of prednisone subjectively and on the pyrexia, but they are too recent for continued improvement to be reported. We can vouch for the good health of Case 10 who is a medical colleague.

Dosage

Our patients usually started with between 20-60 mg. prednisone daily in divided dosage, to be reduced slowly over the next few weeks. Case 3 might have responded to continued prednisone but we felt bound to change to ACTH for the reasons stated. The apparent superiority of ACTH over oral steroids here, and in other diseases such as ulcerative colitis, may well lie in the matter of effective dosage rather than any specific therapeutic effect. We consider that some authors have not used a sufficient initial dose, nor have they continued the therapy for long enough, and for this reason we would not agree with Mason and Adams⁷ that the speed of recovery is similar to those cases treated with bed rest only. We also feel that the dosage should be lessened slowly, and Cases 2 and 7 demonstrate the relapse which occurs if the drug is withdrawn too quickly.

Until more is known of the cause and mechanisms involved in glandular fever, any interpretation of the apparent beneficial effect of steroids in the treatment of the disease, especially of its complications, can be no more than conjecture. We suspect

that the benefit may arise from the known hypoplastic effect of steroids on lymphatic tissue, on hepatitis and encephalitis, as 'antistressor' agents, and possibly by reduction of unhelpful antigen-antibody responses.

Summary

Ten patients with glandular fever are reported in whom steroid therapy had a beneficial effect; three of these had severe complications.

The indications for steroid treatment are discussed together with suggestions for effective therapy.

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