## **Case Report**

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# Tubular enteric duplication presenting as jejuno-colic fistula in a 38 years old adult

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#### **ABSTRACT**

Colo-enteric fistula is a rare entity, malignancy being the commonest cause. Both adenocarcinoma and rarely lymphoma have been known to cause it. Benign jejuno-colic fistulas are mostly secondary to tuberculosis or inflammatory bowel disease. Here we present a case of a young adult male, who presented with altered bowel habits. Colonoscopy and oesophago-gastro-duodenoscopy (OGD) revealed a fistulous tract between the proximal jejunum to the splenic flexure of colon. Surgery revealed a small fistulous connection between the above-mentioned parts of the bowel about 1cm long. Histopathology demonstrated it to have all the layers of the intestinal anatomy, hence indicating a possibility of congenital aetiology, probably enteric duplication of communicating tubular variety.

Keywords: Colo-enteric fistula, Jejuno-colic fistula, Enteric duplication, Benign intestinal fistula, Diarrhoea

#### INTRODUCTION

Gastrointestinal fistulas represent abnormal duct like communications between the gut and another epithelial-lined surface, such as another organ system, anywhere along the Gastrointestinal tract (GIT) itself (internal), or the skin surface (external). Internal GIT fistulas can be further divided into two types: intestinal and extraintestinal. Intestinal fistulas refer to a gut-to-gut connection and may consist of any combination of stomach, small bowel, and colon.<sup>1</sup>

It is also useful to classify fistulas as congenital and acquired, since their clinical settings and implications differ greatly.

Congenital gastrointestinal (GI) fistulas are best understood by realizing their embryologic origin and include such entities as branchial, tracheoesophageal, and omphalomesenteric fistulas.

The underlying causes of acquired GI fistulas are diverse and can include virtually any process resulting in bowel perforation from within or bowel penetration from an extraintestinal process. The majority of external (cutaneous) fistulas represent a complication of recent abdominal surgery.<sup>2</sup>

#### **CASE REPORT**

A 38 years old male presented with history of altered bowel habits for the last 5 years. Along with history of passing undigested food particles soon after food. He also complained of diarrhea which had progressively increased. Patient also complained of pain abdomen ranging from dull aching to colicky in nature, mostly restricted to the lower abdomen. He was evaluated for the same by general physicians and on many occasions received treatment for infective diarrhea. Stool for ova and cyst was negative in the investigations done prior to admission.

On examination he had no pallor or icterus. His abdomen was soft with minimal deep tenderness in the lower abdomen. There was no liver or splenic enlargement and no mass was palpable. Per-rectal examination was normal.

Based on the above findings a provisional diagnosis of chronic diarrhea was made and the patient was subjected for a colonoscopy, which revealed a fistulous tract at the splenic flexure of the colon. We were able to pass the scope into the fistulous tract and a healthy small intestinal mucosa was noted. Other than the fistula colonoscopy showed a couple of small diverticuli, otherwise the colonic mucosa appeared healthy with no evidence of inflammation or growth. An upper GI endoscopy was done on the patient with minimal difficulty the third part of duodenum was intubated and a fistular opening was noted, using suctioning and straightening of the scope the fistula was entered and colonic lumen was identified. Mucosa was found to be normal on the small intestinal side too.



Figure 1: Colonoscopic visualisation of the fistulous tract (F), colonic lumen (C).

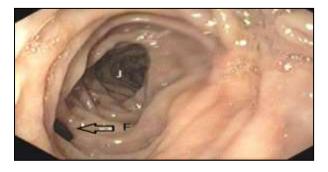


Figure 2: Upper GI endoscopy showing the fistula (F), jejunal lumen (J).

A contrast enhanced Computed tomography (CT) of the abdomen using oral and intravenous contrast confirmed the fistulous tract to be arising from the distal third part of the duodenum to the splenic flexure of the colon. Mesenteric inflammation was noted, and no obvious signs of malignancy were seen on imaging.

Further the patient was subjected to upper midline laparotomy and a tract of  $1\times1$  cm was noted connecting the jejunum just distal to the ligament of treitz to the splenic flexure of the colon. There was no inflammation or mass near the tract. The tract was arising on the mesenteric

aspect of both structures. The rest of the abdomen was found to be normal. No evidence of tuberculosis was found in the abdomen. Taking into consideration the benign appearance of the fistula, only the tract was excised, and the cut ends were closed using standard two layered bowel wall closure.



Figure 3: Interior of the fistulous tract visualised from the colonic aspect.

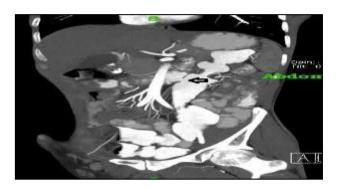


Figure 4: Coronal CT image showing fistula (arrow).



Figure 5: Sagittal CT image showing the fistula (arrow).

Histopathological examination of the fistulous tract showed the presence of all the layers of the intestinal wall, i.e., mucosa, submucosa, muscularis mucosa and serosa. No evidence of inflammation or granulomas was noted. Histopathological examination also did not find any foci of malignancy.

The patient's recovery was uneventful, and he got discharged on the 8th post-operative day. At the time of discharge patient was passing formed stools with no diarrhea. Patient's complaints were found to be relieved in the further follow up visits.

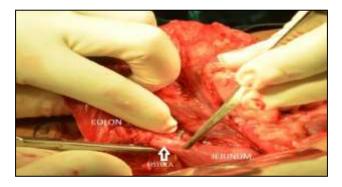


Figure 6: Intraoperative photographs of the fistulous tract.



Figure 7: Intraoperative photographs of the fistulous tract.

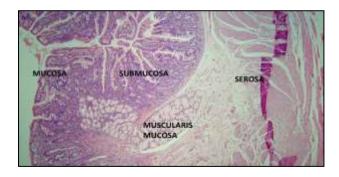


Figure 8: Histopathological picture showing all the layers of bowel, with no evidence of inflammatory or neoplastic focus.

#### DISCUSSION

Despite significant medical and surgical progress over the last two decades, gastrointestinal fistulae (GIF) continue to be associated with high morbidity and mortality.<sup>3</sup> The leading causes of internal fistulas in the industrialized world are acquired because of Crohn disease, diverticulitis, malignancy, or a complication of treatment of these entities.<sup>1</sup> Congenital fistulas can be dorsal-enteric fistula associated with split notochord syndrome and congenital entero-enteric fistula associated with an omphalocele.<sup>4</sup>

Intestinal (gut-to-gut) fistulas may involve combinations of the small bowel, colon, and stomach. The clinical manifestations of this subset may be subtle, since only the alimentary tract is involved. Diarrhoea, with or without abdominal pain, is the most common symptom overall.<sup>5</sup> However, if longer lengths of bowel are bypassed there will be significant metabolic and nutritional disturbances.<sup>6</sup> Gastric, lateral duodenal, ligament of treitz, and ileal fistulae are least likely to spontaneously close. Therefore, these fistulae require the most aggressive medical therapy and are likely to require operative intervention.<sup>7</sup>

Multiple imaging modalities and endoscopy can be used to identify internal fistulae. Contrast-enhanced fluoroscopic examination often remains the initial study of choice and is generally superior to endoscopy in demonstrating the presence and extent of a GI fistula.<sup>8</sup>

There are several factors that influence which segments of bowel are involved in the fistulous communication. In cases where a primary bowel abnormality is the underlying cause, the segment of diseased bowel will obviously be at highest risk. Proximity to the pathologic process, be it intestinal or extraintestinal, is also important. Finally, a pre-existing or preferred pathway, such as a connecting ligament or mesentery, between certain portions of the gut, can explain the predisposition for some intestinal fistulas to form.<sup>1</sup>

Every possible attempt should be made to rule out these pathological processes before categorising a fistula as congenital. Iatrogenic vascular disruptions have been known to cause congenital gastrointestinal anomalies. Anomalies arising after completion of organogenesis are often disruptions or deformations; otherwise the causes are not specific to any developmental period.

Enteric duplication cysts can occur anywhere along the length of the gut where they lie along the mesenteric border and share a common muscle wall and blood supply, though the duplications commonly occur in the distal ileum and esophagus. Duplication cysts are usually spherical lesions and less often tubular. The communication with the adjacent bowel is uncommon in spherical lesions and is more likely to occur with tubular duplications.

Tubular duplications may be short or may involve entire segments, such as the entire oesophagus and stomach combined, or the distal ileum, cecum, entire colon, and anus. Most are attached along the dorsal or mesenteric border. Some are attached to the lateral border, forming parallel, side-by-side segments referred to as double-barrel duplications. Rarely, the duplication has a separate mesentery and blood supply and is called loop duplication. Communications between the lumen of a tubular duplication and the normal lumen are common and may be located at the proximal or distal end of the duplication, or at both ends. Multiple communications may occur. Double-barrel duplications of the colon, rectum, and anus are associated with duplications of part, or all, of the

genitourinary tract and external genitalia, forming symmetrical or asymmetrical, right and left GI and genitourinary tracts and perineum. A tubular duplication with a proximal communication and a blind distal end forms an expanding mass that may obstruct the normal lumen or adjacent structures. Similarly, a cystic duplication may also cause obstruction of the involved segment.<sup>12</sup>

Histological analysis typically reveals at least one outer muscular layer with an inner gastrointestinal mucosal lining. The mucosal lining does not necessarily correspond to that of the adjacent normal intestine and may be comprised several different types of gastrointestinal mucosa. Because of the relative scarcity of such abnormalities, current literature consists mainly of small populations and case reports rather than any large single or multi-institutional series. 14,15,16

#### **CONCLUSION**

Benign colo-enteric fistulas are rare and mostly secondary to either tuberculosis or inflammatory bowel disease. Congenital internal GI fistulas presenting in adulthood are not reported in literature. In this case the possibility of a communicating tubular duplication cyst is likely as no pathological process could be identified even after a thorough search. Also, the fact that histologically the muscular layer was identified with presence of all the normal layers of bowel wall excludes fistula arising from inflammatory and neoplastic events. The origin of the fistula near the mesenteric border and with a short tract to a nearby structure consolidates the possibility of duplication cyst. However, the absence of any other congenital anomaly other than the mentioned fistula does not support a duplication cyst. Though as mentioned earlier, other anomalies are less common when the event occurs after organogenesis. In view of scarcity of literature, we intend to report this case.

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