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CASE REPORT

A boy with duodenocolic fistula mimicking functional gastrointestinal disorder

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Abstract
Duodenocolic fistula (DCF) is a rare disorder defined by the presence of an internal fistula between the duodenum and colon. Colon cancer, Crohn's disease, diverticulum and duodenal ulcer are common causes of DCF, and vomiting and diarrhea are its main symptoms. We report a 14-year-old boy with DCF who had been treated for a functional gastrointestinal disorder (FGID). The boy had often experienced episodes of vomiting and diarrhea since infancy, and had been diagnosed with FGID. He was referred to our hospital because of a 2-month exacerbation of persistent vomiting and diarrhea. Upper gastrointestinal contrast revealed no abnormalities. Eventually, esophagogastroduodenoscopy detected a duodenal fistula, and DCF was diagnosed by endoscopic fistulography. Colonoscopy showed a diverticulum in the ascending colon near the fistula. In addition, a C^{13} urea breath test for *Helicobacter pylori* infection was positive. One hypothetical pathogenesis of his DCF was perforated colonic diverticulitis. Adhesion between the fistula wall and colonic diverticulum near the fistula strongly suggested a relationship between the fistula and the diverticulum. However, he never presented with symptoms of colonic diverticulitis. Thus, a congenital origin was also suspected. After confirming temporary relief from the symptoms by endoscopic closure, surgical closure was performed.

Keywords Child · Congenital · Diverticulum · *Helicobacter pylori* · OTSC (over-the-scope clip)

Introduction

Duodenocolic fistula (DCF) is a rare disorder defined by the presence of an internal fistula between the duodenum and colon. It commonly occurs as a complication of colon cancer, Crohn's disease, or duodenal ulcer [1]. Vomiting and diarrhea are its main symptoms [1].

Functional gastrointestinal disorders (FGIDs) are common disorders characterized by recurrent or chronic gastrointestinal (GI) symptoms that cannot be attributed to structural or biochemical abnormalities [2]. Especially in school-age children, FGIDs are the most common cause of visits to pediatric gastroenterologists by patients complaining of abdominal symptoms [3]. "Functional nausea and vomiting disorders" is one of the disorders in children and adolescents classified according to the Rome IV criteria [4].

We herein report the case of a boy with a DCF who had been treated for FGID.

Case report

A 14-year-old Japanese boy was referred to our hospital because of a 2-month exacerbation of persistent vomiting and diarrhea, and 5 kg of body weight loss. He had often experienced episodes of vomiting and diarrhea since infancy. His symptoms worsened at around 9 years of age. He had been diagnosed and treated as FGID after visiting a local doctor. At 14 years of age, his symptoms worsened at the end of his summer vacation, and from the beginning of the subsequent semester, he tended to be absent from school. His vomiting was induced by overeating, and it was prolonged for few days once it occurred. He had never experienced bilious or bloody vomit.

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At his first visit to our hospital, his height was 166.5 cm (0.13 SD) and his weight was 47.0 kg (−0.73 SD). Five kilograms of body weight loss over 3 months was noted. Physical examination revealed the following findings: body temperature, 36.3 °C; blood pressure, 98/65 mmHg; and heart rate, 103 bpm. Mild abdominal tenderness was noted in the epigastric region.

Laboratory tests revealed the following findings: WBC count, 6400/μl; RBC count, 486×10^4/μl; Hb, 14.1 g/dl; Hct, 42.4%; Plt count, 29.7×10^4/μl; aspartate aminotransferase (AST), 37 IU/L (reference value; 11–30); alanine aminotransferase (ALT), 40 IU/L (4–30); gamma-glutamyl transpeptidase (GGT), 11 IU/L (< 70); serum amylase, 103 U/L; total bilirubin, 0.66 mg/dl (0.40–1.50); total protein, 7.7 g/dl (6.6–8.1); albumin, 5.1 g/dl (4.1–5.1); total cholesterol, 121 mg/dl (127–258), C-reactive protein, 0.01 mg/dl (0.00–0.14); serum amyloid A protein, < 8.0 μg/L; and erythrocyte sedimentation rate, 5 mm/h. A fecal occult blood test, fecal fat staining, fecal calprotectin, and fecal bacterial culture revealed no remarkable findings.

Abdominal X-ray studies, abdominal computed tomography (CT) and brain magnetic resonance imaging (MRI) which was performed to exclude brain tumors showed no significant abnormalities.

Upper gastrointestinal (GI) contrast and enhanced abdominal computed tomography, which was performed 1 month after his first visit to our hospital, revealed no remarkable findings (Fig. 1).

Medication with mosapride citrate and traditional Chinese medicines prescribed for a presumed diagnosis as FGID was ineffective.

Eventually, esophagogastroduodenoscopy (EGD) detected a fistula in the descending part of the duodenum without any gastrointestinal erosions or ulcers (Fig. 2a–c), and a diagnosis of DCF was made based on endoscopic fistulography (Fig. 2d). EGD showed the reflux of colonic contents, including fecal juice, into the duodenum through the fistula (Fig. 2a, c). Colonoscopy (CS) showed a diverticulum in the ascending colon near the fistula (Fig. 3a), and subsequent endoscopic fistulography enhanced the duodenum (Fig. 3b). There were no tumors or ulcers (including ulcer scars) in the stomach, duodenum or colon.

In addition, a C^{13} urea breath test for *Helicobacter pylori* (*H. pylori*) infection was positive.

He underwent endoscopic closure using an over-the-scope clip (OTSC) (Ovesco Endoscopy AG, Tubingen, Germany) in conjunction with the cauterization of fistula margin using argon plasma coagulation. Complete relief from his symptoms was obtained at 3 months, after endoscopic closure and his body weight increased from 47 kg to 56 kg. However, EGD performed 3 months after treatment revealed that the clip was disengaged from the fistula. He began to vomit again after meals, so finally underwent surgical closure of the fistula. The fistula was resected by wedge resection, after which the defect was closed directly. The histological findings of the resected specimens revealed the presence of a transition point in the mucosal membrane from the small intestine to the colon with a well-marked muscular layer. There were no findings of epithelioid cell granuloma formation, malignancy or *H. pylori* infection.

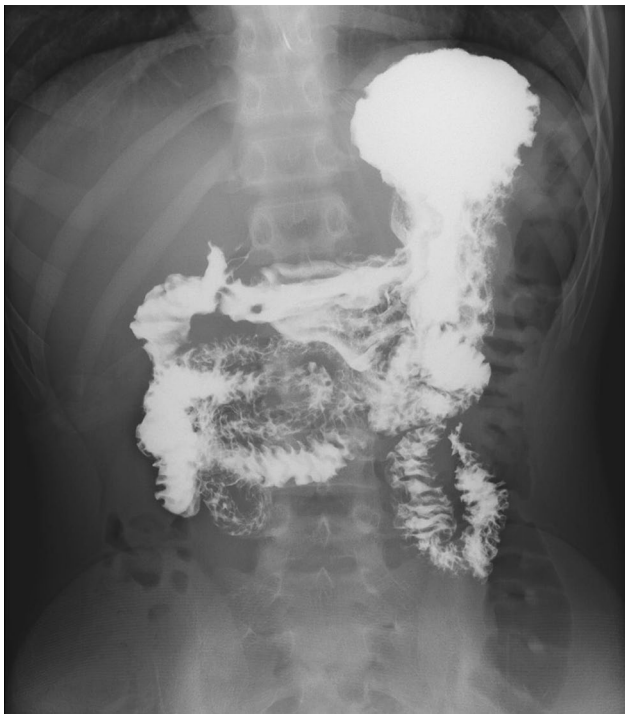

Fig. 1 Upper gastrointestinal (GI) contrast. No leakage from the duodenum into the colon was observed in the upper GI contrast study. However, a double contrast examination was not performed

Discussion

The clinical course of this child with DCF illustrated two notable issues: first, his symptoms and the results of screening examinations mimicked those of FGID; and second, the fistula was not detected by conventional upper GI contrast in this case.

Benign DCF is defined as a fistula that is created with no malignant disease, and is quite a rare entity in children [5]. The main symptoms of DCF, and the percentage of patients who they affect are as follows: chronic abdominal pain (79%), diarrhea (75%), body weight (64%), and nausea/vomiting (50%) [1, 6, 7]. The passage of colonic contents through the fistula to the duodenum is considered to be dependent on the intestinal pressure gradient between the colon and duodenum. Subsequent bacterial overgrowth in the small intestine is considered to lead to diarrhea,

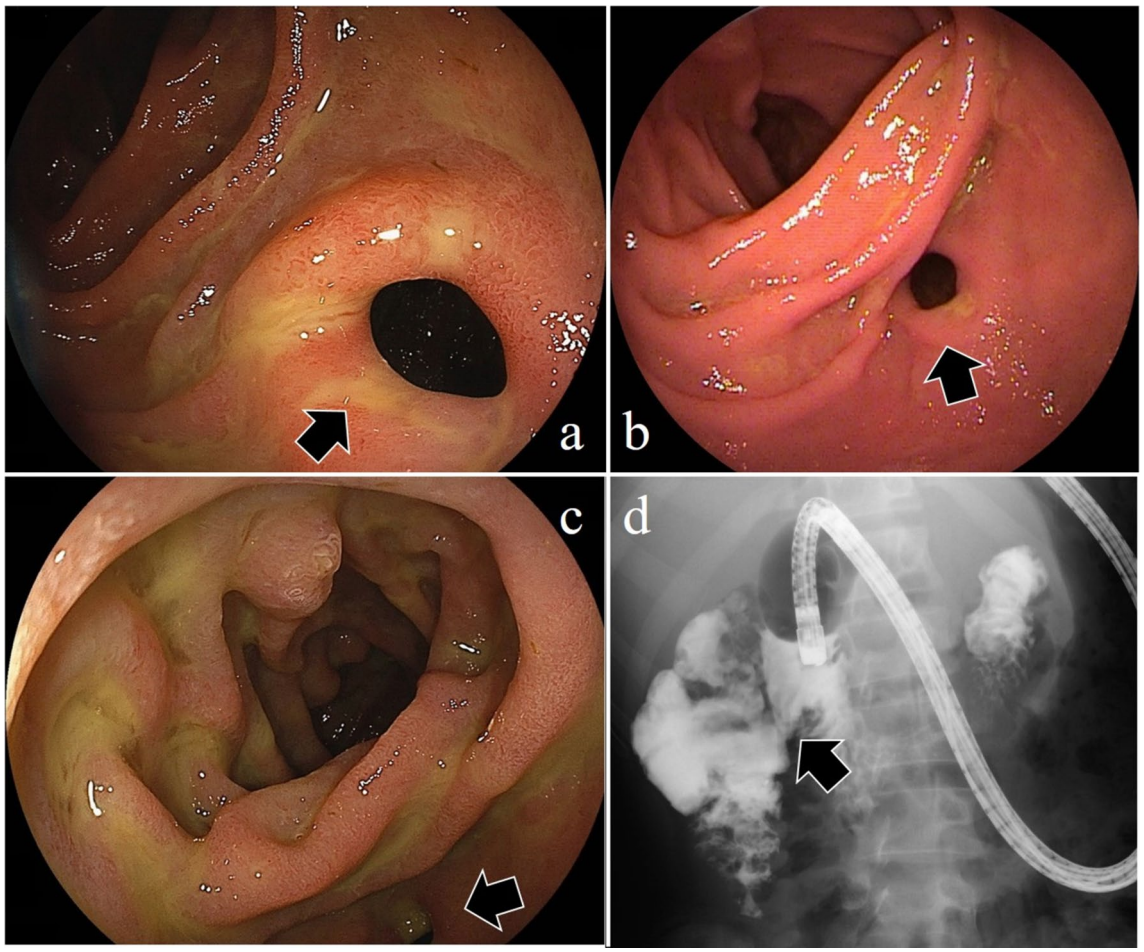

Fig. 2 Esophagogastroduodenoscopy (EGD) and endoscopic fistulography. **a–c** The EGD findings of the patient. The fistula was located at the second portion and opposite side of the duodenal papilla. **a** and **c** Fecal juice was refluxed to the duodenum via the fistula. **d** Fistulography through esophagogastroduodenoscopy enhanced the colon. Black arrows indicate the fistula

steatorrhea electrolyte disturbances, and malabsorption [8, 9]. Fecal vomiting is rarely reported in patients with DCF [10]. Even in our case, despite the colonic contents were observed in the duodenum, he never had fecal vomiting. The pylorus prevents the contents of the duodenum from flowing back into the stomach [10].

Chronic abdominal pain, prolonged diarrhea, and frequent nausea/vomit are also symptoms of pediatric FGIDs, which are common diseases of children especially in school-age children. The prevalence of FGIDs in school-age children of 8–15 years of age is estimated to be 2.4–38% [11–15]. Therefore, most pediatricians may tend to diagnose FGIDs when children present with abdominal symptoms. In our case, the abdominal symptoms worsened at the end of the summer vacation and the repeated remission and exacerbation of the patient's condition suggested that it was worsened due to stress at school. We, therefore, considered that he was suffering from FGID, which led to a delay in the diagnosis of DCF. It is difficult for children to undergo endoscopy. Thus, most pediatricians struggle to decide the indications for endoscopic examination. However, physicians should not hesitate to perform upper GI contrast and/or endoscopic examinations in cases with alert signs, such as prolonged vomiting and body weight loss [16].

An upper GI contrast study is reported to be the most reliable examination for diagnosing DCF, with sensitivity of 85–90% [17, 18]. However, in our case, the fistula was not detected by an upper GI contrast study, rather it was detected by EGD. The position of the fistula or the intestinal pressure gradient between the colon and duodenum might have been associated with the false-negative result. No effervescent agent was used during the upper GI contrast study. The use of an effervescent agent might have caused the internal pressure of the stomach and duodenum to increase, leading to the detection of the fistula. EGD was useful in the detection of the fistula in this case. Generally, endoscopic examinations are also useful for identifying the cause of fistula (e.g., Crohn's disease or colon cancer).

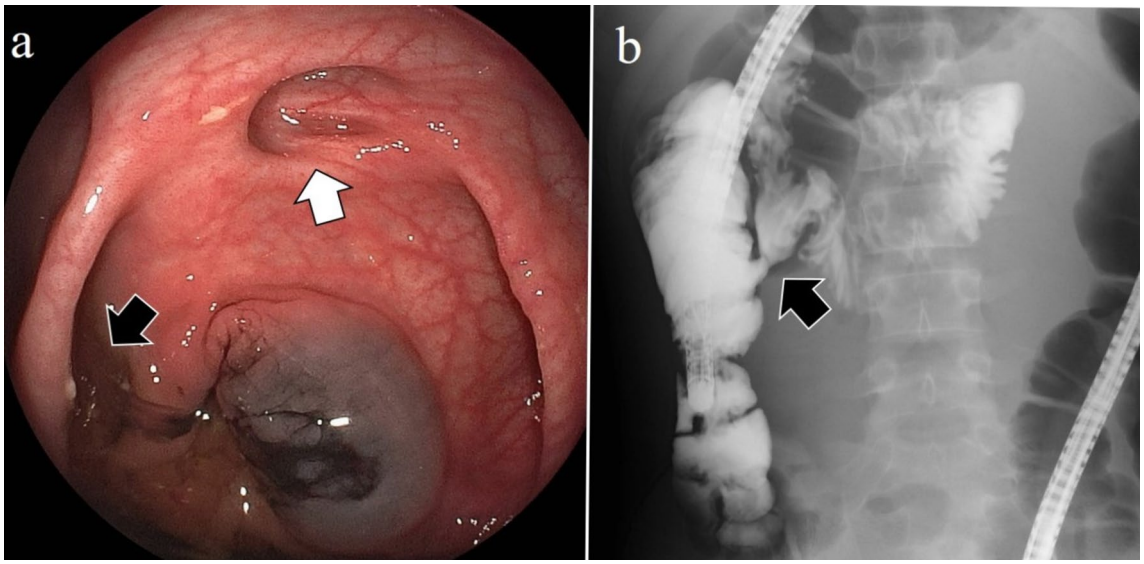

Fig. 3 Colonoscopy and endoscopic fistulography. **a** The colonoscopy findings. Colonoscopy revealed a diverticulum in the ascending colon near the fistula. **b** Fistulography through colonoscopy enhanced the duodenum. Black arrows indicate the fistula and the white arrow indicates the diverticulum

The frequency of causal factors of benign DCF were reported to be duodenal ulcer (24%), Crohn's disease (23%), duodenal diverticulum (11%), and idiopathic/spontaneous (11%) [10]. There are only five reports of DCF caused by colonic diverticulum [10]. In our case, in addition to *H. pylori* infection, the patient had a colonic diverticulum located near the fistula. One of the causes of his fistula was perforation of the colonic diverticulum due to diverticulitis. Since the diverticulum was located near the fistula, it is possible that he had another diverticulum and that its diverticulitis induced the fistula. The intraoperative finding that the diverticulum near the fistula was firmly adhered to the fistula wall was one of the findings that strongly suggesting a relationship between the fistula and the diverticulum. A perforated duodenal ulcer associated with *H. pylori* infection was another possible cause of the fistula. However, duodenal ulcers caused by *H. pylor*i infection typically occur in the bulb [19]. On the other hand, duodenal ulcers occurring in the second portion are usually induced by vasculitis, such as IgA vasculitis—especially in the children. Thus, we considered that the possibility of a relationship between *H. pylori* infection and the patient's fistula was low. He had never presented with symptoms of colonic diverticulitis or IgA vasculitis. He had loose stools and had never presented a formed stool since infancy. However, his stool was normally formed after the treatment of DCF. Furthermore, the shape of the fistula was smooth and round. For these reasons, we hypothesize that the fistula had existed for a long time and that it might have had a congenital origin. Sharma et al. reported a suspected case of congenital DCF [5].

Treatment of DCF depends on the underlying pathology. Surgical treatment is the initial treatment for the management of DCF [20]. Partial colectomy with en bloc excision of the fistula and partial duodenectomy is recommended [10]. OTSC was reported to be effective for the management of gastrointestinal fistula [21–23], and the long-term success rate of closing the fistula with OTSC was reported to be around 80% (11/14) [22]. OTSC allows the closing of defects by grasping much larger amounts of tissue with a higher compression force than conventional clips [22, 24]. On the other hand, Mizrahi et al. reported that it was less successful in lower GI defects, particularly for the treatment of chronic fistula [25]. They reported that the success rate in patients with lower GI fistula was 0% (0/9). Although endoscopic fistula closure with an OTSC also did not go well in our DCF case, it was useful in confirming not only the cause of patient's GI symptoms but also the improving effect of fistula closure on the symptoms. Therefore, it may be a valuable choice for closing GI fistulae, including DCF before surgical treatment because of its lower invasiveness, especially for children.

In conclusion, a patient with a DCF can present with symptoms and screening examination findings that mimic FGID. An upper GI contrast study and/or EGD examination should be actively attempted in cases with alert signs, such as prolonged vomiting and body weight loss.

Compliance with ethical standards

Conflict of interest All the authors declare no conflicts of interest in association with the present study.

Human rights All the procedures followed were performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from the patient and his mother for inclusion in this study.

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