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Choledochoduodenal fistula: A rare cause of upper gastrointestinal bleeding in a child

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Informed Consent
The authors stated that the written consent was obtained from the parents of the patient presented with images in the study.

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Abstract

Spontaneous choledochoduodenal fistula (CDF) is a rare form of biliary enteric fistula. A child with CDF who had a motor vehicle collision as a pedestrian at the age of 2 years is presented in this article. Since the accident, recurrent abdominal pain and black-colored stools were noted thrice a year. As a 13- year-old, the patient was admitted with vomiting of blood and melena. He was hospitalized with gastrointestinal bleeding. Endoscopy is performed because of bleeding and a fistula was detected incidentally. Barium swallow series and magnetic resonance cholangiopancreatography showed a fistula tract. Endoscopic retrograde cholangiopancreatography (ERCP) confirmed the definitive diagnosis and guided treatment. In our case, we emphasize the importance of ERCP in facilitating the diagnosis of CDF. Barium swallow radiography detects the passage of barium to the biliary system in only half of the CDF patients diagnosed via ERCP. In summary, we reported the youngest case of CDF with a large fistula orifice managed by endoscopic sphincterotomy.

Keywords: Children, Choledochoduodenal fistula, Endoscopy, Endoscopic retrograde cholangiopancreatography (ERCP)

Introduction

The first biliary fistula was described in 1654 by Bartholini, and it was observed between the gallbladder and intestine. Various types of biliary fistulas have since been described between the biliary and respiratory systems, skin, and vessels. Most biliary fistulas originate from the gallbladder and end up in the digestive tract. Spontaneous biliary fistulas between the common bile duct and the duodenum are rare [1]. All of the reported choledochoduodenal fistula (CDF) cases consisted of adult patients except one patient as 15 years old [2 - 4].

Herein, we report a rare form of biliary fistula. To our knowledge, this case of CDF of a 13-year-old is the youngest reported case.

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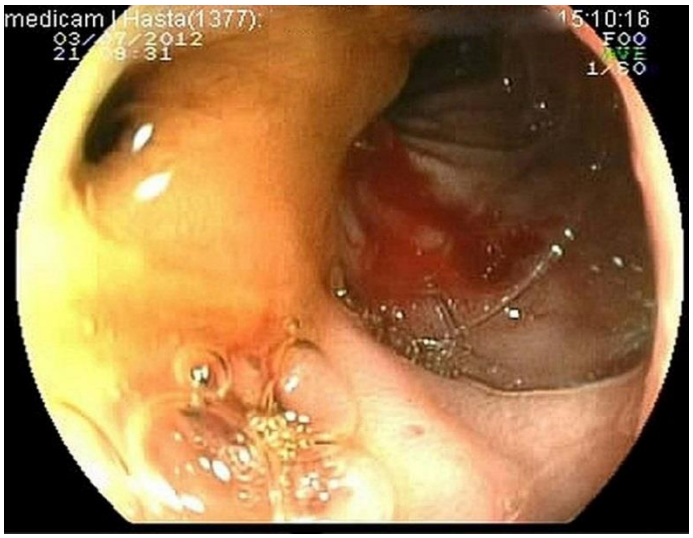
Case presentation

A thirteen-year-old male was admitted with the complaint of blood vomiting. The blood had an appearance similar to that of ground coffee. He had been suffering from epigastric pain for one month. The patient had a motor vehicle collision as a pedestrian when he was two-years old. Two months after the accident, abdominal pain and black-colored stools were noted and these recurred two to three times a year. The patient appeared ill and pale, without jaundice or fever. Abdominal examination revealed epigastric tenderness without muscular defence, and melena was present. Other physical examination findings were unremarkable. Laboratory analysis revealed anemia, with hemoglobin and hematocrit levels as 10 g/dL [normal: 13- 14.5] and 30 % [normal: 36-43], respectively. Liver function tests, serum amylase levels, and coagulation test results were within normal ranges. Neither an abdominal X-ray nor abdominal ultrasonography demonstrated any pathologies. The patient was hospitalized with a diagnosis of gastrointestinal bleeding and intravenous fluids were commenced. The informed consent was taken from the parents.

Endoscopy was performed after the restoration of intravascular volume. The endoscopy revealed a duodenal ulcer, measuring 1.5 cm, located on the posterior wall of the bulbus. The major duodenal papilla had a normal appearance and was located in the medial wall of the second part of the duodenum. There was a 1.5 cm fistula orifice proximal to the ampulla of Vater (Figure 1).

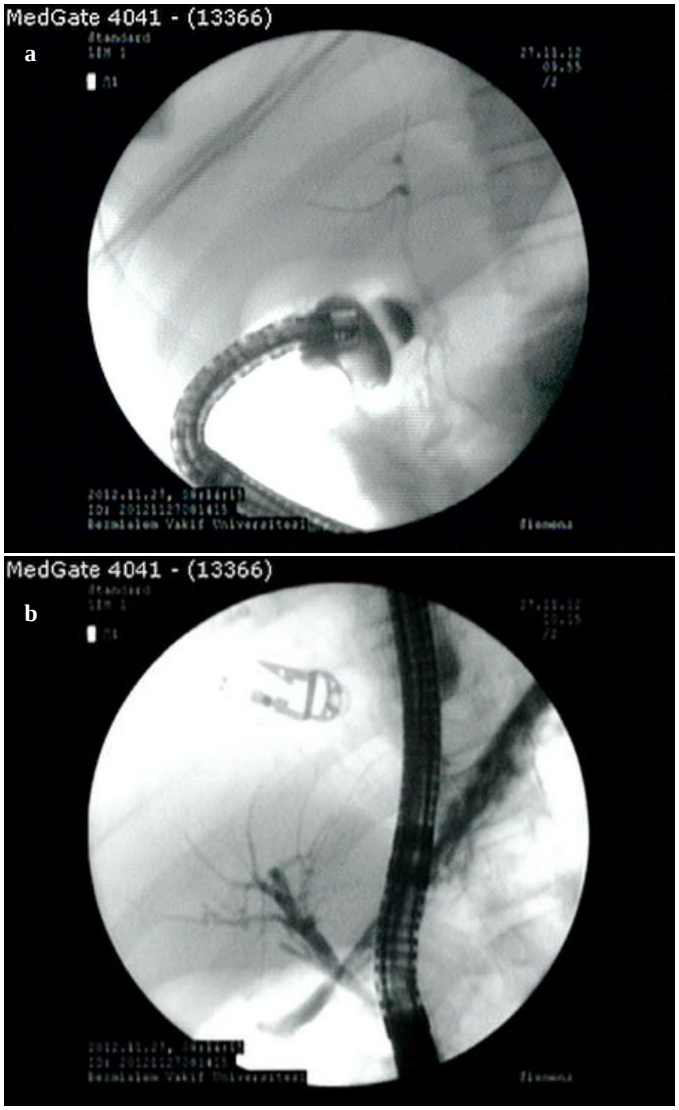
Figure 1: Endoscopic appearance of the fistula orifice



In this case, we were able to observe active bile flow to the duodenum. The upper gastrointestinal system barium swallow series showed a fistula orifice and ulcer niche in the duodenum, as well as barium reflux to the biliary system.

Magnetic resonance cholangiopancreatography (MRCP) also revealed a fistula between the duodenum and middle-to-distal common bile duct. Duodenal ulcer and gastroesophageal reflux were treated with three months of lansoprazole and domperidone. The symptoms subsequently resolved. Control endoscopy showed complete regression of the ulcer, but the fistula orifice size remained. Endoscopic retrograde cholangiopancreatography (ERCP) revealed that the biliary tree was filled through both the fistula and papilla of Vater (Figure 2a and 2b).

Figure 2: a: Endoscopic retrograde cholangiopancreatography. Biliary tree filling through the fistula, b: Endoscopic retrograde cholangiopancreatography. Biliary tree filling through the papilla of Vater



We were also concerned about the future consequences of bile ponding in a segment of the common bile duct, especially in the segment between the fistula and papilla of Vater. Sphincterotomy was performed both to provide the union from the papilla extending to the fistula and to divert the bile flow distally, because bile reflux from the large fistula orifice may have caused recurrent ulcers and cholangitis. The patients follow-up was done periodically after the procedure, there were no recurrent bleeding or any symptoms related with the fistula.

Discussion

Choledochoduodenal fistula cases constitute less than 5 % of all biliary fistulas especially before 1980 in literature [1]. With frequent use of ERCP, published cases of CDF were also increased. In 1997, Yamashita et al. [3] reported that 62 % of biliary fistulas in their study were CDF cases. Endoscopic retrograde cholangiopancreatography facilitated the diagnosis of CDFs. Barium swallow detects the passage of barium to the biliary system in only half of the CDF patients diagnosed via ERCP [3]. Air in the biliary system is another indirect fistula sign that can be detected with X-ray or computed tomography, but it is only seen in 25%–40% of patients with CDF [4, 5]. In our patient, the abdominal X-ray was normal, but endoscopy revealed a fistula orifice, and barium swallow series and MRCP

confirmed the fistula tract. Finally, ERCP provided a definite diagnosis.

Endoscopic retrograde cholangiopancreatography is useful for diagnosing CDF but may be complicated by pancreatitis, cholangitis, hemorrhage, and perforation, particularly after sphincterotomy. Our patient did not have any complaints after ERCP. Choledochoduodenal fistulas are classified according to the opening level of the fistula to the common bile duct as either peripapillary or proximal. The proximal fistulas are detected less frequently and are thought to be caused by the perforation of ulcers located in the bulbus, to the common bile duct [6]. Peripapillary fistulas originate from the distal 2 cm of CBD. Spontaneous peripapillary fistulas are associated with biliary stones in more than 95% of patients [5, 7]. They are supposed to be formed by common bile duct stones fistulizing to the duodenum due to associated infection and pressure. In addition to biliary stones, papillary cancers [8] or blunt abdominal trauma [2] may result in spontaneous peripapillary fistulas. Our patient had no history of biliary surgery or intervention, and we did not detect any biliary stones. The fistula orifice and ulcer did not overlap in terms of localization. Complaints in our patient started after a traffic accident. Chao et al. [2] reported a similar CDF case due to blunt abdominal trauma. The trauma resulted in papillary edema, biliary flow obstruction, pancreatitis, and fistula in a 15-year-old patient. The mean age of patients with CDF is reported as 60 years in previous studies [3-8].

Here, we present the youngest case. Symptoms of CDF are usually associated with underlying diseases. In proximal fistulas, ulcer symptoms [6] are common, whereas biliary stones and cholangitis symptoms [5, 7] are predominant in peripapillary fistulas. Studies concerning peripapillary fistulas [3-5] reported biliary and pancreatic complaints. In our case, we performed an endoscopy because of bleeding, and a fistula was detected incidentally. Unlike the major duodenal papilla, the fistula orifice does not have a sphincter, so the intestinal flora may reflux into the biliary system. Li et al. [4] reported that cholangitis attacks become more frequent when the size of the fistula orifice is increased. In a third of patients, food particles were present in the biliary system if the fistula orifice was larger than 1 cm [4]. Our patient did not have symptoms of cholangitis on admission. His course after the sphincterotomy procedure was uneventful.

Conclusion

In summary, we reported the youngest case of CDF resulting from a duodenal ulcer that was managed by endoscopic sphincterotomy. Because of high suspicion, radiologic procedures were followed for the definitive diagnosis, although the abdominal X-ray was normal. Barium swallow series and MRCP showed fistula tract. Finally, ERCP provided a definite diagnosis. Endoscopic retrograde cholangiopancreatography is a useful diagnostic procedure for CDF.

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