INTRODUCTION

Xanthogranulomatous cholecystitis (XGC) is a rare type of chronic inflammation of the gallbladder, characterized by focal or diffuse destructive inflammatory responses. Although it is a benign condition, its destructive course may lead to more aggressive outcomes of the gallbladder, such as local infiltration, fistula, stricture, and perforation as compared with other gallbladder inflammations. There are reports about XGC accompanied by cholecystoenteric fistula. However, XGC accompanied by more than one cholecystoenteric fistula is rare. We report a case of a 54-year-old man with gastric outlet obstruction arising from XGC, accompanied by cholecystoduodenal fistula and cholecystocolonic fistula, but without impacted gallstones.

Xanthogranulomatous cholecystitis (XGC) is a rare type of chronic inflammation of the gallbladder characterized by focal or diffuse destructive inflammatory responses. Although it is a benign condition, its destructive course may lead to more aggressive outcomes of the gallbladder as compared with other gallbladder inflammations; these include local infiltration, fistula, stricture, and perforation.1-5

According to a review of literatures, there are reports about XGC accompanied by cholecystoenteric fistula.6 To our knowledge, however, there are no reports about a synchronous formation of two cholecystoenteric fistulas in patients with gastric outlet obstruction (GOO) arising from XGC.

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GOO, arising from XGC, accompanied by cholecystoduodenal fisula and cholecystocolonic fistula, but without impacted gallstones. Here, we report our case with a review of literatures.

**CASE**

A 54-year-old man visited the emergency department with a 4-day-history of hiccups and abdominal bloating. The patient had a past 7-year-history of taking aneurismal neck clipping and left frontotemporoparietal craniectomy for spontaneous subarachnoid hemorrhages due to aneurysmal rupture of the left internal carotid artery.

On admission, the patient was alert. The patient’s vital signs were as follows: blood pressure 100/65 mmHg, heart rate 90 times/minute, respiratory rate 18 breaths/minute, body temperature 36.9 ℃, and PO₂ 99%. The patient had abdominal distension, but showed no other notable find-

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![Fig. 1. (A-C) Enhanced computed tomography of the abdomen showed a deformity of duodenal bulb with suspicious cholecystoduodenal fistula (long arrow) and cholecystocolonic fistula (short arrow) at the hepatic flexure of the colon. (D) Enhanced computed tomography of the abdomen showed a diffuse wall thickening of gallbladder with suspicious gallstones and gastric distension.](image-url)
Gastric Outlet Obstruction arising from Xanthogranulomatous Cholecystitis accompanied by Both Cholecystoduodenal and Cholecystocolonic Fistulas

Findings, such as abdominal tenderness. On complete blood counts, the patient showed a white blood cell count of 15,200/mm$^3$, hemoglobin 12.5 g/dL, and platelet counts of 356,000/mm$^3$. On serum biochemistry, the patient showed 14 IU/L of aspartate aminotransferase, 20 IU/L of alanine aminotransferase, a total bilirubin of 0.5 mg/dL, 223 IU/L of alkaline phosphatase, 48 IU/L of r-glutamyl transpeptidase and 8.57 mg/dL of c-reactive protein. Serum carcinoembryonic antigen was 2.21 ng/mL and carbohydrate antigen 19-9 was 6.69 U/mL. On abdominal computed tomography (CT) scans, the patient had severe gastric distension and diffused gallbladder wall thickening, accompanied by findings that were suggestive of gallstones. The patient also had suspected findings of cholecystoduodenal fistula with duodenal bulb deformity and cholecystocolonic fistula (Fig. 1). On esophagogastroduodenoscopy, the patient had circumferential mucosal break and mucosal friability, extending from the esophagogastric junction to the upper esophagus (Fig. 2A), accompanied by the residual presence of dirty food debris in the stomach. The patient also had endoscopic findings of

![Figure 2](image_url)

**Fig. 2.** Esophagogastroduodenoscopy showed (A) grade LA-D erosive esophagitis at the esophagogastric junction, (B, C, D) mucosal edema surrounding prepyloric antrum, and extraluminal narrowing of the duodenal bulb, with no presence of ulcerations.
mucosal edema surrounding the prepyloric antrum and extraluminal narrowing of the duodenal bulb, without the presence of an ulcer (Fig. 2B-D). Nevertheless, the scope barely passed through the bulb. Due to a lack of findings indicative of an ulcer, Helicobacter pylori infection test was not preformed. The patient received conservative treatment regimens using an intravenous injection of fluid, and antibiotics, during fasting. Thus, the patient was tentatively diagnosed with XGC accompanied by cholecystoduodenal fistula and cholecystocolonic fistula. Moreover, the patient was presumed to have GOO, possibly due to a partial obstruction that may be associated with an inflammation arising from XGC. However, it could not be completely ruled out the possibility of gallbladder cancer occurred as a result of the gallbladder wall thickening on CT scans. We consulted a general surgeon, according to whom the patient was suspected of having XGC, rather than malignancy. On day 2, per request from the general surgeon, a percutaneous transhepatic gallbladder drainage (PTGBD) was preformed. A tubogram showed duodenal and colonic filling. This was suggestive of cholecystoduodenal fistula in the duodenal bulb and cholecystocolonic fistula in the hepatic flexure of the colon (Fig. 3). After receiving PTGBD and conservative managements, including an intravenous injection of metoclopramide and proton pump inhibitor, the patient’s hiccups and abdominal distension was resolved. On day 16, the patient underwent exploratory laparotomy. Intraoperatively, the patient had edematous gallbladder with diffused wall thickening, accompanied by an adhesion to the adjacent omentum, the first portion of duodenum and the proximal transverse colon. The patient underwent cholecystectomy and primary closure of cholecystoduodenal fistula and cholecystocolonic fistula. On histopathologic examinations, the patient was diagnosed with XGC, accompanied by hemorrhagic necrosis and fistula tract formations (Fig. 4). On day 28, the patient was discharged without complications.
DISCUSSION

XGC is an uncommon variant of chronic cholecystitis, and it is characterized by focal or diffused destructive inflammatory findings, showing a variability in acute and chronic inflammatory cells, lipid-laden macrophages, and fibrosis during the later stages.2,3 Clinically, it shows no typical symptoms and signs, when compared with other inflammatory gallbladder diseases except for the frequent occurrence of complications, such as adhesions to adjacent tissues and organs, and the formation of internal fistula between the gallbladder and the adjacent viscera.2,3 Its clinical presentations are similar to those of acute or chronic cholecystitis; these include symptoms in the right hypochondrial region, radiating pain in the shoulder and back, nausea, vomiting, and/or fever.5 In this case, the patient presented with hiccups and abdominal bloating. It is important to note that these findings are not indicative of XGC. Most cases of hiccups show a benign, self-limited and time-limited course. Although rare, clinicians should consider the possibility of serious underlying diseases in patients with a persistent hiccup lasting for more than 24 hours.7,8 Hiccups have been reported to be an atypical symptom of gastro-esophageal reflux disease (GERD).9 Its prevalence is estimated at 4.5-9.5% in patients with GERD.9 In this case, the patient had cholecystoduodenal fistula with a deformed bulb, leading to partial obstruction on CT scans. A subsequent esophagogastro-duodenoscopy showed a few linear mucosal denudations at the esophagogastric junction extending to the upper esophagus and extraluminal narrowing of the duodenal bulb, without the presence of ulcerations. We assume that the patient presented with GOO, and severe gastric reflux esophagitis, due to a partial obstruction of the duodenal bulb arising from XGC accompanied by cholecystoduodenal fistula.

Cholecystoenteric fistula is a rare complication of biliary lithiasis that is defined as, a spontaneous tract with bile flow between an inflamed gallbladder and one or more adjacent structures.10 Its incidence has been estimated at 0.9-3.2% in patients with biliary disease.10,11 The fistula can be cholecystoduodenal (69-70%), cholecystocolonic (14%) and cholecystogastric (6%).13 Although patients with XGC are vulnerable to cholecystoenteric fistula, at a reported incidence of 12.1%,5 there are few reports that it is accompanied by synchronous complications, such as cholecystoenteric fistulas. There are also reports about gallbladder-common bile duct fistula accompanied by cholecystocolonic fistula.5 To our knowledge, there are no reports about a synchronous occurrence of cholecystoduodenal fistula and cholecystocolonic fistula in a patient with XGC. There are numerous articles about cholecystoenteric fistula associated with gallstones. In this case, the patient had no definite evidence of impacted gallstones. However, the patient did have gallstones accompanied by both cholecystoduodenal fistula and cholecystocolonic fistula.

There is a possibility that XGC might progress to coincident carcinoma. Once diagnosed with XGC, patients with XGC should undergo a cholecystectomy, and excision of adjacent xanthogranulomatous tissue and any fistulas.2 In this case, however, the patient had no malignancy upon histopathological examinations.

We experienced a rare case of GOO arising from XGC, accompanied by cholecystoduodenal fistula and cholecystocolonic fistula. To our knowledge, this has not been described in the literature. Here, we report our case with a review of literatures.
Conflicts of Interest

The author has no conflict to disclose.

REFERENCES