CASE REPORT

Bouveret syndrome: A fatal diagnostic dilemma of gastric outlet obstruction

Fazıl Sağlam, M.D., Emre Sivrikoz, M.D., Ali Alemdar, M.D., Sedat Kamalı, M.D., Ufuk Arslan, M.D., Hakan Güven, M.D.

Department of General Surgery, Okmeydani Training and Research Hospital, Istanbul

ABSTRACT

The patient presented in this study was a 54-year-old woman complaining of nausea and vomiting, onset preceding four days, with no significant past medical history and an unremarkable surgical history. The patient was afebrile and hypertensive. Physical examination revealed a non-tender abdomen, and initial laboratory evaluation revealed elevated blood glucose level, ketonuria, leukocytosis, elevated C-reactive protein, gamma glutamyl transferase, lactate dehydrogenase, and total bilirubin. The patient was admitted to the internal medicine ward due to new onset of diabetes mellitus. Due to persistent nausea and vomiting, gastroscopy revealed a healed duodenal ulcer, and abdominal ultrasonography revealed cholelithiasis. The medical condition of the patient deteriorated further in the internal medicine ward, with impending hypotension, tachycardia, leukocytosis, and acute renal failure, and she was admitted to the intensive care unit due to septic shock. A computerized tomography was obtained, which revealed an impacted gallstone in the distal duodenum. The patient was taken to the operating room. The gallstone was encountered in proximal jejunum immediately distal to the ligament of Treitz. A longitudinal enterotomy was made, and the stone was extracted. Her drains were cleared on postoperative day 5, and gastrointestinal function returned to normal. Unfortunately, the patient developed an overwhelming sepsis due to bacteremia and fungemia, and died on post-operative day 19.

Key words: Bouveret syndrome; diabetes; gastric outlet obstruction.

INTRODUCTION

Bouveret syndrome was first described by Leon Bouveret in 1896.[1] The pathophysiology consists of a gallstone passing through a cholecystoduodenal fistula into the duodenal lumen, causing a gastric outlet obstruction. Prominent symptoms include abdominal pain, nausea and vomiting. Endoscopic findings suggestive of this syndrome include a dilated stomach containing digested food and a hard non-fleshy mass at the obstruction.[2] Rigler's triad was described to characterize Bouveret syndrome: pneumobilia, obstruction, ectopic gallstones.[3]

Address for correspondence: Fazıl Sağlam, M.D.
Okmeydanı Eğitim ve Araştırma Hastanesi, Genel Cerrahi Kliniği, Kaptan Paşa Mahallesi, Şişli, 34384 İstanbul, Turkey
Tel: +90 212 - 221 77 77 E-mail: fzlsaglam@yahoo.com

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CASE REPORT

The patient presented in this study was a 54-year-old woman with no significant past medical history and an unremarkable surgical history, complaining about nausea and vomiting, onset preceding 4 days. The patient was afebrile, and hypertensive (190/100 mmHg), and her physical examination revealed a non-tender abdomen. Her initial laboratory evaluation revealed elevated blood glucose level (379 mg/dl), ketonuria, leukocytosis (14.230 / mm), elevated C-reactive protein (50.2 mg/L), gamma glutamyl transferase (83 U/L), lactate dehydrogenase (831 U/L), and total bilirubin (1.68 mg/dl). She was admitted from the emergency department to the internal medicine ward due to the diagnosis of new onset diabetes mellitus.

Upon admission, intravenous insulin infusion was started and her blood glucose was regulated to upper-normal limits. The patient underwent routine evaluation for new onset of diabetes mellitus including echocardiography (left ventricular hypertrophy), ophthalmoscopy (unremarkable for retinopathy), and electromyography (unremarkable for polyneuropathy). Due to persistent nausea and vomiting, a gastroscopy was

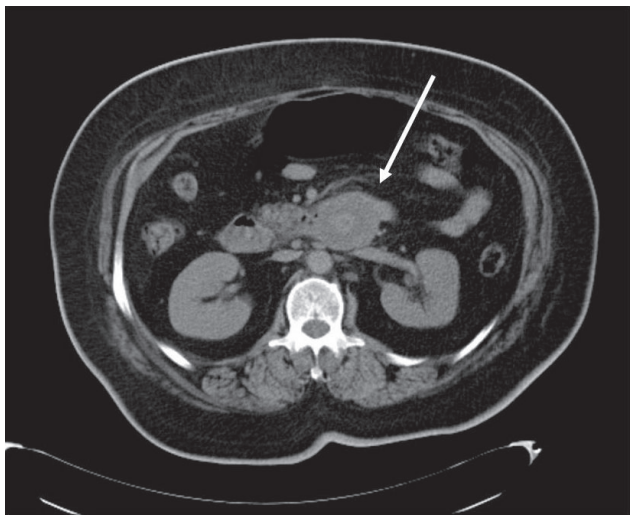

Figure 1. CT scan showing impacted gallstone.

undertaken, and a healed duodenal ulcer was encountered. An abdominal ultrasonography revealed cholelithiasis, and an elective cholecystectomy was planned. However, her medical condition deteriorated further in the internal medicine ward, with impending hypotension (80/40 mmHg), tachycardia (140/min), leukocytosis (19.000/mm), and her creatinine level increased to 3.62 mg/dl (initially normal). She was admitted to the intensive care unit (ICU) due to septic shock, and intravenous dopamine infusion was started.

Upon admission to the ICU, a computerized tomography (CT) was obtained, which revealed an impacted gallstone in the distal duodenum (Fig. 1). General surgery was consulted for the first time, and decision for an operative intervention was made due to her unstable medical condition associated with a proximal gastrointestinal obstruction. After stabilizing her vitals in the ICU, the patient was taken to the operating room. Following a median laparotomy, the ligament of Treitz was exposed and the gallstone was encountered in proximal jejunum immediately distal to the ligament of Treitz (Fig. 2a). A longitudinal enterotomy was made and the stone was extracted (Fig. 2b). Enterotomy was closed transversely. The patient was taken back to the ICU. Her drains were cleared on postoperative day 5, and gastrointestinal function returned to normal. Unfortunately, she developed an overwhelming sepsis due to bacteremia and fungemia, and died on post-operative day 19.

DISCUSSION

Gallstone ileus is encountered in 0.3-0.4% of all cholelithiasis cases, with a preponderance of advanced age. Terminal ileum is the most affected (50-90%) site for impaction, which results in a mechanical small bowel obstruction.[4] Less commonly, a proximal obstruction at jejunum and ileum may occur (20-40%), which essentially presents as a gastric outlet obstruction. Obstructive jaundice may accompany the clinical picture depending on the level of obstruction close to the ampulla of Vater.

Given the lack of substantial expertise on such a rare disease, there is controversy regarding the management of gallbladder at the time of surgery. Surgery should be carried out in a goal directed fashion, where the mainstay of treatment is relief of the offending obstruction.[5] Cholecystectomy with or without fistula repair should carefully be weighed against a medically deprived patient requiring a rapid alleviation of obstruction rather than an expeditious surgery. The present

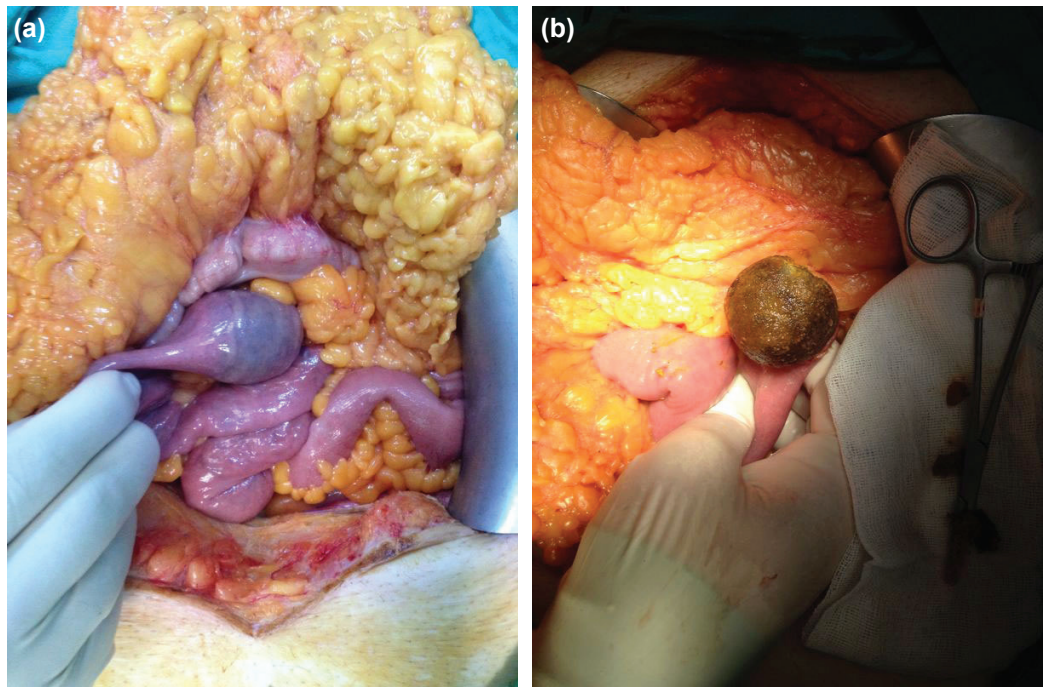

Figure 2. (a) Exposure of impacted gallstone. **(b)** Enterotomy and gallstone extraction.

case emphasized the need for correct diagnosis in a timely fashion. Since the patient, cohort harboring this syndrome, rapidly reached the extremis due to excessive electrolyte imbalance and possible obstructive jaundice, the diagnosis depended upon meticulous suspicion. It should also be noted that the surgical procedure is not skill demanding, but early intervention is the only hope for cure. In addition, there is evidence in the literature documenting spontaneous closure of fistulae following endoscopic gallstone retrieval.[6]

As reported in this unfortunate case, surgery was delayed due to diagnostic confusion, and an irreversible cascade not amenable to surgical intervention developed. Nausea and vomiting comprised the cardinal presenting symptoms of the patient, and a gastro-duodenoscopy was performed revealing a healed duodenal ulcer during her work-up in the internal medicine ward. The third and fourth portions of the duodenum were not routinely visualized during gastro-duodenoscopy. Since the lesion/stone was not within the reach, an endoscopic treatment was not deemed feasible during the surgical consultation. Moreover, the overall medical condition of the patient was unstable, which was a clear indication for urgent laparotomy. The patient survived the operation, yet surgery could not arrest the development of overwhelming sepsis which led to the patient's demise. Bouveret Syndrome is a rare entity, which should be kept in mind when evaluating a gastric outlet obstruction associated with chronic cholelithiasis, especially when gastro-duodenoscopy reveals normal findings.

Conflict of interest: None declared.

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OLGU SUNUMU - *ÖZET*

Bouveret sendromu: Mide çıkış yolu obstrüksiyonunda ölümcül bir tanısal ikilem

Dr. Fazıl Sağlam, Dr. Emre Sivrikoz, Dr. Ali Alemdar, Dr. Sedat Kamalı, Dr. Ufuk Arslan, Dr. Hakan Güven

Okmeydanı Eğitim ve Araştırma Hastanesi, Genel Cerrahi Kliniği, İstanbul

Bilinen ek hastalığı ve geçirilmiş ameliyat öyküsü olmayan 54 yaşında kadın hasta dört gündür devam eden bulantı ve kusma yakınmasıyla acil servise başvurdu. Fizik muayenede karında hassasiyet saptanmadı. Başvurusunda yapılan laboratuvar incelemelerinde yüksek kan şekeri, ketonüri, lökositoz, yüksek C-reaktif protein, gama-glutamil-transferaz, laktat dehidrogenaz ve total bilirubin saptandı. Hasta yeni başlangıç diabetes mellitus tanısıyla dahiliye servisine yatırıldı ve rutin incelemelerine başlandı. Devam eden bulantı ve kusma nedeniyle yapılan gastroskopide iyileşmiş duodenal ülser saptandı. Karın ultrasonografisinde kolelityazis saptandı. Dahiliye servisinde incelemeleri devam etmekteyken hastanın tıbbi durumu giderek kötüleşti. Hipotansiyon, taşikardi, lökositoz ve akut böbrek yetersizliği gelişen hasta septik şok tanısıyla yoğun bakım ünitesine alınarak intravenöz dopamin infüzyonu başlandı. Çekilen bilgisayarlı tomografide distal duodenumda sıkışmış görünümde bir safra taşı saptandı. YBU'da vital bulguları stabilize edilen hasta ameliyata alındı. Laparotomiyi takiben Treitz ligamanı distalinde proksimal jejunumda safra taşı görüldü. Longitudinal enterotomi ile taş çıkarıldı. Hasta tekrar YBU'ya alındı. Ameliyat sonrası beşinci günde drenleri alınan hastanın gastrointestinal fonksiyonları normale döndü. Takiplerinde bakteriyemi ve fungemiye sekonder sepsis gelişen hasta ameliyat sonrası 19. günde kaybedildi.

Anahtar sözcükler: Bouveret sendromu; diyabet; mide çıkış yolu obstrüksiyonu.

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