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Case Report
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Bouveret syndrome: evaluation with multidetector computed tomography and contrast-enhanced magnetic resonance cholangiopancreatography

Bouveret sendromu: Çok kesitli bilgisayarlı tomografi ve kontrastlı manyetik rezonans kolanjiyografi bulguları

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Cholecystenteric fistula is one of the rarest complications of biliary lithiasis, with a frequency of less than 1%. Bouveret syndrome is a gastric outlet obstruction produced by gallstone(s) located in the distal stomach or proximal duodenum. The route of gallstone migration to the bowel is most commonly via a cholecystoduodenal fistula; however, fistulization of the stomach is a rarer variation. Early diagnosis of this situation is crucial to reduce morbidity and mortality. In this report, we present a patient with cholecystogastric fistula and Bouveret syndrome. To our knowledge, there is no published paper in the literature related to the diagnosis of Bouveret syndrome with multidetector computed tomography (MDCT) (64 detectors) and/or contrast-enhanced magnetic resonance cholangiopancreatography (CE-MRCP). Our aim was to discuss the efficacy of MDCT and CE-MRCP in the detection and evaluation of cholecystenteric fistulas. We showed the exact localization and relation of biliary stones and the fistula by MDCT and CE-MRCP. We also evaluated the biliary system with CE-MRCP physiologically. In conclusion, when biliary lithiasis and ileus are detected in plain radiography, the first-line diagnostic tool should be MDCT. In complicated cases or when biliary obstruction is suspected, CE-MRCP can give important morphological and physiological information regarding the whole abdomen and biliary system.

Key Words: Bile ducts; Bouveret syndrome; fistula; gastric outlet obstruction; gallbladder; magnetic resonance cholangiography; multidetector row computed tomography; stone.

Kolesistoenterik fistüller, safra taşlarının %1'den daha az bir sıklıkta görünen komplikasyonlarındandır. Bouveret sendromu, mide çıkımında safra taşı/taşları tarafından oluşturulan tıkanıklık sonucu gelişir. Bu taşlar sıklıkla kolesistoenterik veya nadiren de kolesistogastrik fistül aracılığı ile mide çıkımını veya proksimal duedenumu tıkarlar. Bouveret sendromunun erken tanı ve tedavisi, morbidite ve mortalitenin azaltılması için gereklidir. Bu olgu sunumunda, kolesistoenterik fistüllü ve Bouveret sendromlu bir hasta sunuldu. Bildiğimiz kadarıyla, çokkesitli bilgisayarlı tomografi (ÇKBT) (64 dedektörlü) ve kontrastlı manyetik rezonans (MR) kolanjiyopankreatografi (MRKP) ile Bouveret sendromu tanısı hakkında çalışma literatürde bulunmamaktadır. Amacımız kolesistoenterik fistüllerin saptanması ve değerlendirilmesinde ÇKBT ve kontrastlı MRKP etkinliğini tartışmaktır. ÇKBT ve kontrastlı MRKP ile safra yollarını fizyolojik olarak inceleyebildik ve biliyoenterik fistülü net olarak gösterdik. Ayrıca kontrastlı MRKP ile fizyolojik olarak safra sistemini değerlendirdik. Sonuç olarak, bu tür olgularda ÇKBT, röntgen ve ultrasondan sonraki ilk tercih olmalıdır. Komplike veya safra tıkanıklığı şüphesi olan olgularda kontrastlı MRKP, tüm karın ve safra sistemi ile ilgili önemli morfolojik ve fizyolojik bilgileri verebilir.

Anahtar Sözcükler: Safra yolları; Bouveret sendromu; fistül; mide çıkım obstrüksiyonu; safra kesesi; manyetik rezonans kolonanjiyografi; çok kesitli bilgisayarlı tomografi; safra taşı.

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Gallstone-induced ileus is a rare complication of cholelithiasis, and accounts for 1-3% of all cases with bowel obstruction.[1-3] Bouveret syndrome is a gastric outlet obstruction produced by gallstone(s) located in the distal stomach or proximal duodenum.[1] The route of gallstone migration to the bowel is most commonly via a cholecystoduodenal fistula; however, fistulization of the stomach is an infrequent variant.[2]

In this report, we present a patient with cholecystogastric fistula and Bouveret syndrome. We show the exact localization of biliary stones and the relation of the biliary stones and the fistula by multidetector computed tomography (MDCT) and contrast-enhanced magnetic resonance cholangiopancreatography (CE-MRCP). We also evaluated the biliary system with CE-MRCP physiologically. To our knowledge, there has been no previous report in the literature about a patient with Bouveret syndrome that was definitively diagnosed with MDCT (64 detectors) or CE-MRCP. Our aim was to discuss the efficacy of MDCT and CE-MRCP in the detection and evaluation of cholecystenteric fistulas.

CASE REPORT

An 88-year-old male patient, with no significant personal history, was admitted to the emergency department with the complaints of fever, nausea and pain in his right hypochondrium. Abdominal examination revealed sensitivity in the right upper quadrant and pain with palpation. Leukocytosis and elevated blood urea nitrogen, creatinine, transaminases, and alkaline phosphatase were detected in the laboratory tests. In plain radiography, multiple gallstones and biliary tree air were detected. In the ultrasound (US) examination, the liver size was at the upper limit of normal, and an enlarged gallbladder filled with gallstones was observed. There was another 4x4 cm stone inferomedial to the gallbladder, in the neighborhood of the head of the pancreas and duodenum. Optimal abdominal US examination could not be performed due to poor cooperation of the patient. Since heterogeneity was detected in the gallbladder wall and in the head of the pancreas and the choledochus could not be detected in US examination, non-contrast-enhanced (NCE) abdominal MDCT was done to clarify a possible gallbladder perforation, ileus and acute biliary pancreatitis.

The MDCT scans with 64 detectors confirmed the presence of a 4x4 cm gallstone in the gastric pylorus, causing gastric outlet obstruction (Bouveret syndrome) and dilatation. In addition, there were a few stones in the gastric lumen, the largest being about 1 cm (Fig. 1). Diameter of the ductus choledochus was 13 mm, and intrahepatic bile ducts were minimally dilated. In the MDCT, the gallbladder content was heterogeneous and the gallbladder had penetrated the gastric pylorus. There was no pericholecystic fluid or acute pancreatitis.

In the upper gastrointestinal endoscopy, a gallstone obstructing the gastric pylorus was observed, but attempts to move the stone were unsuccessful. NCE-MRCP was performed to evaluate the biliary system. NCE-MCRP images were immediately evaluated by an experienced radiologist. Penetration of the gallbladder to the gastric wall, multiple gastric and cholecystic gallstones and gastric dilatation were demonstrated clearly (Fig. 2). However, the choledochal

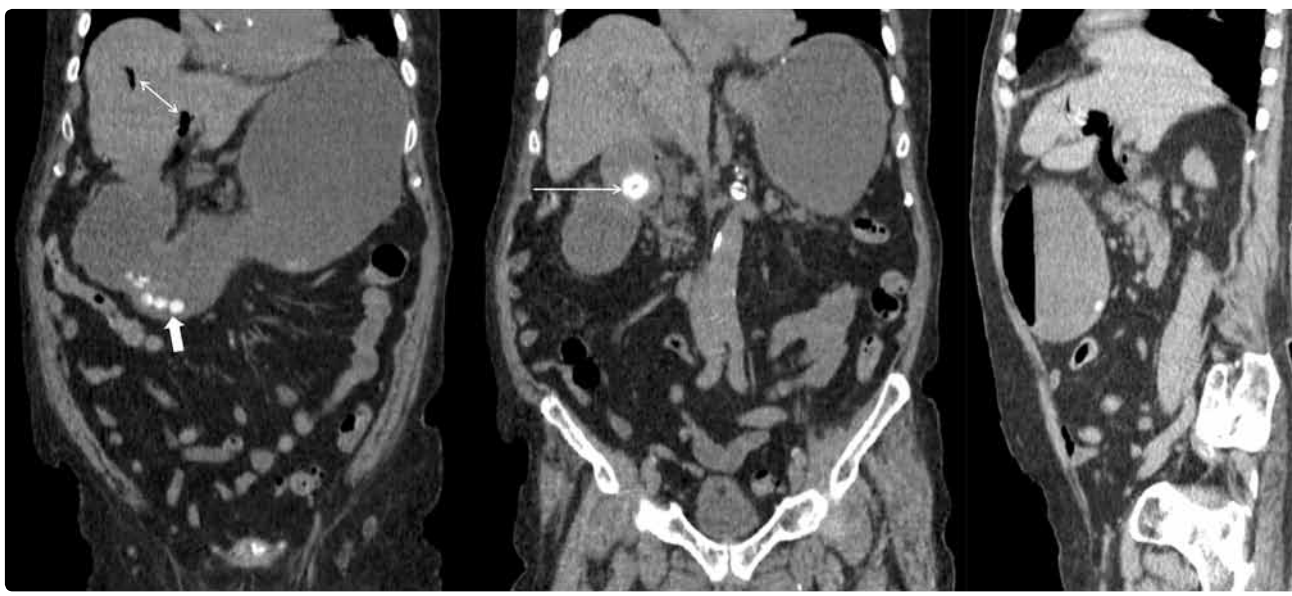

Fig. 1. Coronal (on right and center) and sagittal (on left) reformatted MDCT images of the patient. MDCT images demonstrate pneumobilia (double-headed arrow), gastric distention, and antral (thick arrow) and pyloric (long arrow) stones.

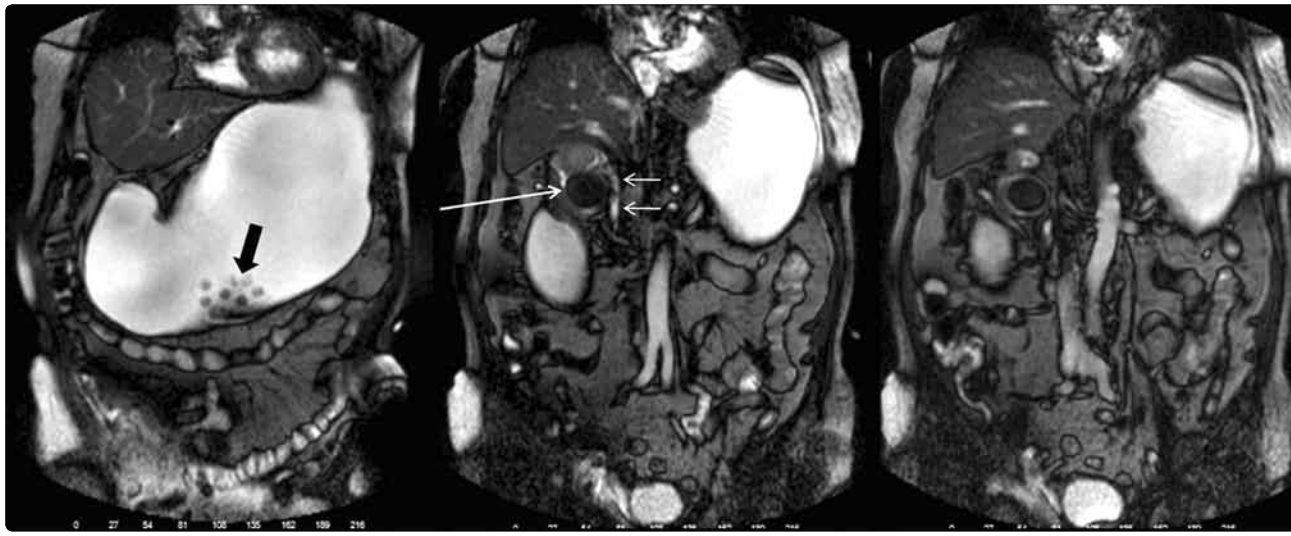

Fig. 2. Sequential coronal non-contrast-enhanced MRCP images show multiple gastric stones (thick arrow) and a large calculus impacted in the gastric pylorus (long arrow). There are several T2-weighted hypointensities in the ductus choledochus (short thin arrows).

lumen and obstruction in the biliary pathway could not be evaluated optimally. Thus, liver-specific contrast agent (0.1 ml/kg gadoxetic acid, Primovist, Schering, Germany) was given intravenously (IV). Images were taken at the 1st (portal phase), 20th (early-phase CE-MRCP) and 40th (late-phase MRCP) minutes after IV administration of gadoxetic acid. No obstruction was detected in the biliary pathways and no choledochal stone was detected in CE-MRCP (Fig. 3).

Surgical treatment was planned. During surgery, the gallbladder was found to be fistulized into the gastric wall, and a stone measuring 4x4 cm in diameter was detected in this region. Cholecystectomy together with antrectomy followed by gastrojejunostomy were performed. There was full correlation between the surgical and radiological findings. The patient died on the 5th postoperative day due to cardiovascular problems.

DISCUSSION

Cholecystenteric fistula is one of the rarest complications of biliary lithiasis, with a frequency of less than 1%.[1-3] Most are cholecystoduodenal fistulas (in 60% of cases), but cholecystocolic (in 17%), cholecystogastric (in 5%), and choledochoduodenal (in 5%) fistulas have been described as well.[4] Large stones passing through the fistula may cause intestinal obstruction, and this condition can be located in the terminal ileum (60%), proximal ileum and jejunum (20-40%), colon, and less frequently, stomach or duodenum, as detected in our case.[3] Bouveret syndrome is a special form of gastric outlet obstruction produced by impaction of a gallstone in the distal stomach or proximal duodenum.[4] It was described by Leon Bouveret in 1896 with upper abdominal pain, fever and emesis in an elderly patient with a history of biliary pain.[1-4] The differential diagnosis of this syndrome includes gallstone ileus, perforated peptic ulcer disease, pancreatitis, and malignant fistula.[2] Size of the gallstone (2-8 cm), long history of biliary disease, repeated episodes of acute cholecystitis, female sex, and advanced age (over 60 years) have been described as risk factors for fistula formulation.[3] Early diagnosis of Bouveret syndrome or bilioenteric fistulas is important because the mortality rate has been reported to be as high as 30%, although it has decreased to 12% in recent years.[4] The high mortality may be related to advanced age, other comorbidities and complications of surgical intervention.[2]

Plain radiographs, upper gastrointestinal fluoroscopy, and/or US examination can be useful in the diagnosis of Bouveret syndrome.[4] However, these techniques are diagnostic in about 50% of cases.[5,6] Endoscopic imaging can confirm the diagnosis and may provide a therapeutic modality.[1] Since patients with Bouveret syndrome are usually elderly and in poor general condition, these tests are insufficient and cannot give optimal results. This situation can result in a delay in diagnosis. CT or MDCT generally establishes the diagnosis of Bouveret syndrome.[7] As we experienced in our case, the choledochus lumen cannot be evaluated clearly using CT or MDCT in approximately 25% of the cases. On the other hand, NCE-MRCP is a useful non-invasive tool for the evaluation of the whole biliary system and gastric/duodenal lumen. Also, NCE-MRCP clearly differentiates fluid from calculi, and can directly detect cholecystoduodenal fistula if there is enough fluid in the area.[4]

As we observed in our patient, when complicated or obstructive biliary pathology is suspected, CE-

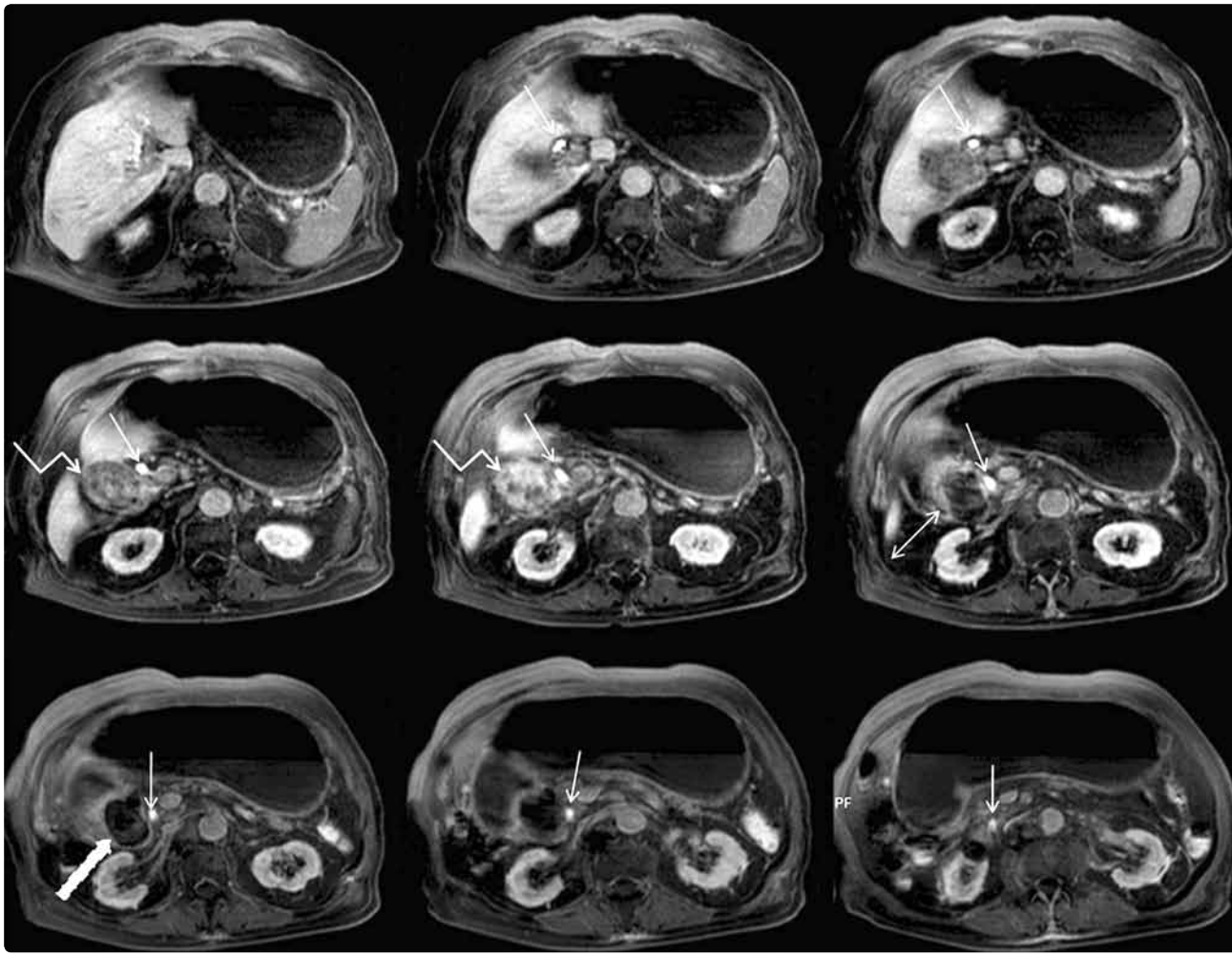

Fig. 3. Sequential axial contrast-enhanced MRCP images of the patient. There was no obstruction in the biliary system, the choledochus lumen is open, and there was no stone in the lumen (thin arrows). There were multiple stones and heterogeneous appearance in the gallbladder lumen (curved arrows). In the neighborhood of the inferior wall of the gallbladder, there was contrast-material enhancement and heterogeneous appearance (double-headed arrow). A calculus impacted in the gastric pylorus was seen next to this area (thick arrow).

MRCP can be useful for evaluating whether or not there is intrahepatic or extrahepatic obstruction. CE-MRCP can be used for detection of pericholecystic inflammation and evaluation of the gallbladder, stomach and bowel walls.[8,9] In addition, in patients with renal insufficiency or impaired renal functions, use of gadoxetic acid is more convenient when compared with other MR contrast agents, because 50% of gadoxetic acid is eliminated by the liver. CE-MRCP is useful in the detection of other complications of cholelithiasis (such as gallbladder perforation) as well.[8]

Open surgery, endoscopic removal and laparoscopic enterolithotomy have all been attempted for stone removal.[5] Although endoscopic removal is less invasive, it fails when the obstructing gallstone is very large. Fragmentation of calculi with endoscopic graspers can remove the blockage of the distal small bowel.[7] The ideal treatment is to relieve the obstruction by removing the offending gallstone, to close the fistula and prevent recurrences, and this can be achieved by open surgery.[3] In patients who are poor surgical candidates secondary to concomitant illnesses and advanced age, surgery is not preferred. If surgery is performed, enterolithotomy alone may be sufficient in such patients and a subsequent cholecystectomy may not be required.[2] In our patient, upper endoscopy was performed first. A stone obstructing the gastric pylorus and a fistula between the antrum and gallbladder were observed. All endoscopic attempts to move the stone were unsuccessful. Hence, surgery was the only opportunity for relieving the obstruction and preventing cholangitis, as well as to rule out gallbladder cancer. A semi-elective operation was performed, after completing fluid and electrolyte resuscitation and the obligatory consultations.

In conclusion, Bouveret syndrome is a very rare

variant of gallstone ileus. Early diagnosis is crucial to reduce morbidity and mortality. Therefore, when biliary lithiasis and ileus are detected in plain radiography, the first-line diagnostic tool should be MDCT. NCE-MRCP is useful for the morphological evaluation of the biliary system, but it cannot give physiological information. In complicated cases or when biliary obstruction is suspected, CE-MRCP can give important morphological and physiological information regarding the whole abdomen and biliary system.

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