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Case Report

Combined Laser and Mechanical Lithotripsy for Endoscopic Management of Bouveret's Syndrome: A Case Report

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Abstract

Bouveret's syndrome (BS) represents an exceedingly rare clinical entity characterized by gastric outlet obstruction induced by a gallstone passing through a cholecystoduodenal, cholecystogastric or choledochoduodenal fistula and impacting in the duodenum or pylorus. Endoscopy is the preferred first-line therapy. It has a favorable safety profile, but requires high level of expertise to achieve stone clearance.

We report here the case of a 75-year-old Caucasian male who presented with vomiting and upper gastrointestinal bleeding and was diagnosed with BS by upper endoscopy. A single session stone extraction utilizing mechanical lithotripsy and laser lithotripsy was executed. Complete clearance of the duodenum and gallbladder was achieved, extracting successfully all fragments larger than 5 mm.

The rarity of BS and its non-specific presentation makes it largely overlooked leading commonly to delayed diagnosis, which results in a high mortality rate of 12-30%. Computer tomography is the diagnostic method of choice. Upper endoscopy can establish the presence of obstruction, but fails to visualize a stone in up to 1/3 of the cases. Management is either endoscopic or surgical. Endoscopic treatment has a good safety profile (mortality 1.6% vs. 17.5-25% for surgery) but poor success rate – 43% vs. 94.1%. To achieve higher success rate, more advanced techniques are required such as mechanical or laser lithotripsy.

Laser lithotripsy is a feasible option for endoscopic treatment of BS, but the duration of the procedure might be an issue in frail patients.

Keywords

Bouveret's syndrome, cholecystitis, endoscopy, gallstone, laser lithotripsy, mechanical lithotripsy

Abbreviations	
BS: Bouveret's syndrome	**UE:** upper endoscopy
GOO: gastric outlet obstruction	**ML:** mechanical lithotripsy
GSD: gallstone disease	**RT:** Rigler's triad

INTRODUCTION

Bouveret's syndrome (BS) represents an exceedingly rare clinical entity characterized by gastric outlet obstruction (GOO) induced by gallstone passing through a cholecystoduodenal, cholecystogastric or choledochoduodenal fistula and impacting in the duodenum or pylorus. No more than 5% of the cases of small bowel obstruction occur as a complication of gall stone disease (GSD), of them barely 1-3% are due to Bouveret's syndrome.[1] Since the initial description of the disease in 1986, there are only 315 documented cases of BS (mainly in the form of case reports), this fact further underlining the rarity of the condition.[2,3] Treatment could be either endoscopic or surgical. Endoscopic treatment has a much better safety profile (1.6% mortality) compared to 12–30% for the surgical approach.[4] On the other hand, surgery is undeniably advantageous in terms of success rate - 94.1% vs. 43% for endoscopy.[5] Herein, we present our experience with a single case of BS, managed in a single session endoscopically with combined laser and mechanical lithotripsy.

CASE REPORT

A 75-year-old Caucasian male was admitted with a 24-hour history of dull pain in the epigastrium, bloating, nausea, persistent vomiting of 'coffee-like' content, and three bowel movements of liquid, tarry stools. A digital rectal examination was performed with signs of melena. An abdominal ultrasound examination failed to establish etiology. A non-contrast enhanced computer tomography established no data suggestive of BS (mild dilation of the stomach, but no pneumobilia or a stone in the duodenum) ***(Fig. 1)****.*

Within less than 24 hours of admission (as per the standard of care in Bulgaria), an upper endoscopy (UE) was performed (Olympus GIF-HQ190, Olympus, Hamburg, Germany). It was found that in the first portion of the duodenum, there was a large bile stone measuring about 4 cm in diameter. A 2-cm fistulous opening was visualized with a second stone of about 3 cm in diameter impacted in it. The mucosa in the duodenal bulb was severely inflamed with multiple bleeding erosions and superficial ulcerations induced by the mechanical friction of the calculus ***(Fig. 2)****.*

The stone in the duodenum was entrapped with a 30 mm loop (Captivator; Boston Scientific, Marlborough, Massachusetts, USA) and pulled through the pylorus into the stomach. A 5 cm Dormia basket (Trapezoid^{TM} RX; Boston Scientific, Marlborough, Massachusetts, USA) was carefully negotiated around the stone. It was attempted then to extract the stone in one piece through the mouth, but it proved impossible to pass it through the upper esophageal sphincter. It was decided that mechanical lithotripsy (ML) was the only viable option for stone clearance. The Dormia basket was cut at the handle and lithotripsy spiral (Endoflex, Hamburg, Germany) combined with lithotripsy handle type III (Enfolex, Hamburg, Germany) were used to carefully fragment the stone, with stone fragments subsequently being removed with a 4-cm Dormia basket (Trapezoid^{TM} RX; Boston Scientific, Marlborough, Massachusetts, USA).

It was impossible to negotiate the second stone with a snare or basket, so it was decided that laser lithotripsy (LL) might

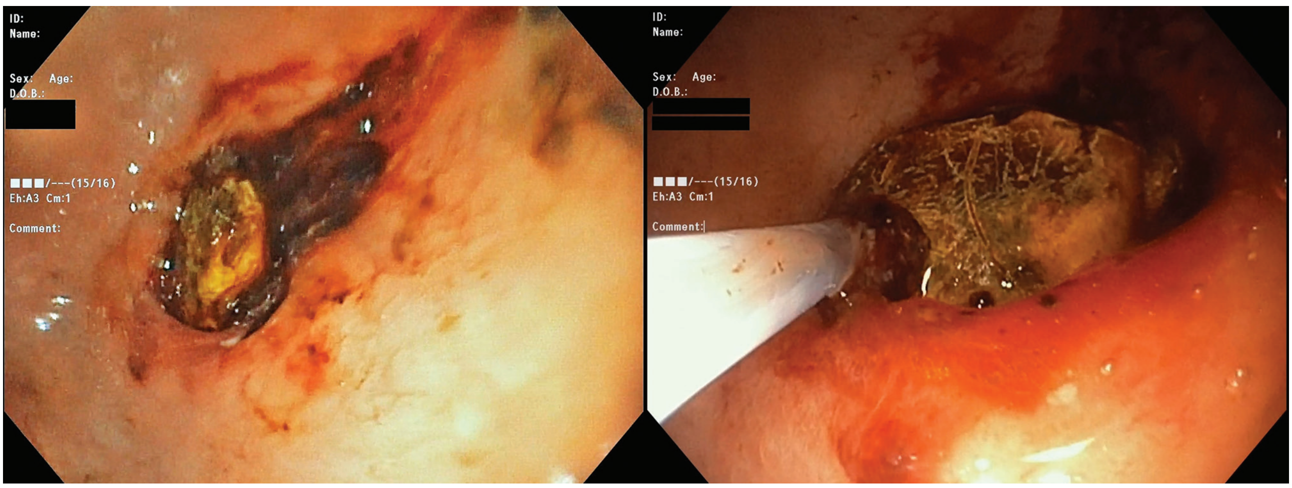

Figure 1. Gallstone impacted in duodenum and protruding from pylorus (left); Entrapment of the stone with a snare (right).

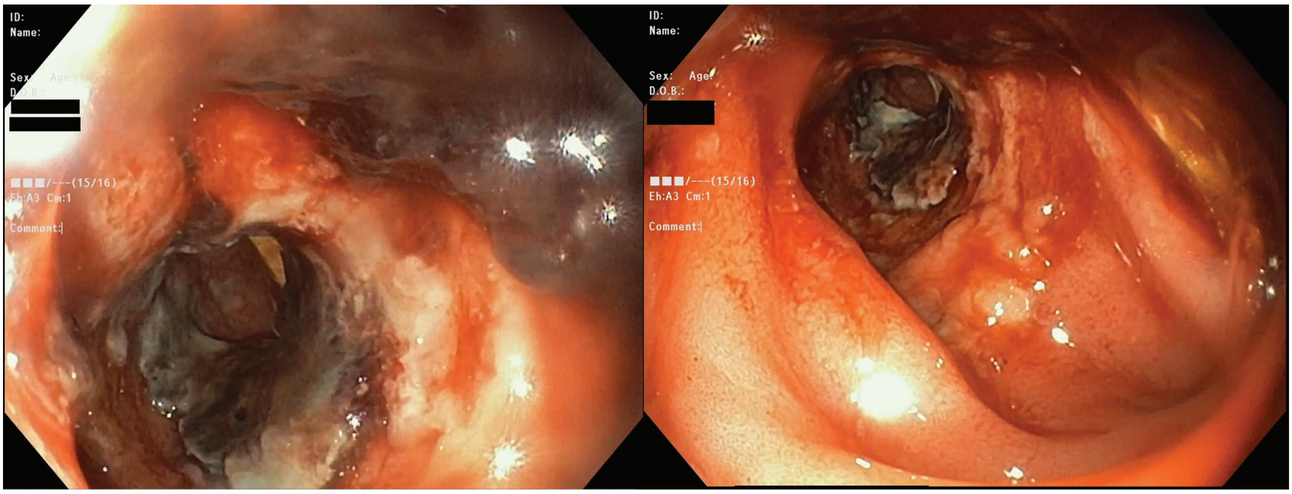

Figure 2. Two-cm cholecystoduodenal fistula.

represent a viable albeit lengthy therapeutic option. Using a laser fiber of 250 μm introduced through the gastroscope, laser lithotripsy of the stone was performed using a Karl Storz Calculase II Holmium laser system (Karl Storz, Tuttlingen, Germany) set at 1.4 J/12 Hz. Laser bursts of <5 s of duration were applied. Eventually the stone was sufficiently fragmented to achieve complete clearance of the gall bladder. The gall bladder was diligently inspected, confirming that there were no residual stones in it and a patent cystic duct was visualized. The duration of the entire procedure was about 210 min, with the laser lithotripsy session solely taking about 180 min ***(Fig. 3)****.*

Patient's recovery was uneventful. There were no adverse events (as described by ASGE lexicon).[6] The patient remains well and symptom-free for 8 months post-procedure.

DISCUSSION

GSD is becoming more frequent, with a prevalence of roughly 7.1% in Western countries.[7] Despite that, the overall complication rate is around 1% with BS accounting for merely 1-3% of those cases. Only 1-3% of those cases are due to a stone impacted in the pylorus/duodenum thus causing BS. The rarity of the condition combined with the patients' characteristics result in a high mortality rate of 12-30%.[2]

In the current paper, clinical symptoms included vomiting, hematemesis, and melena. This presentation is consistent with the symptoms described in literature - nausea/vomiting (86%), abdominal pain (71%), hematemesis (15%), unintentional weight loss (14%), anorexia (13%), constipation (9%), melena (6%), early satiety (3%), and dysphagia (2%).[8]

The diagnosis of BS requires multidisciplinary approach to be set. Rigler's triad (RT) is considered to be the most specific finding in BS. RT includes the presence of dilated stomach, pneumobilia (Gotta-Mentschler sign) and ectopic stone in the duodenum/pylorus. Unfortunately, such combination of symptoms is encountered in merely 50% of patients with BS.[9]

CT is the most commonly utilized diagnostic technique. Regrettably, in about 20% of the cases biliary stones are isoattenuating and thus invisible on CT.[2] UE is another diagnostic option. While almost universally capable of

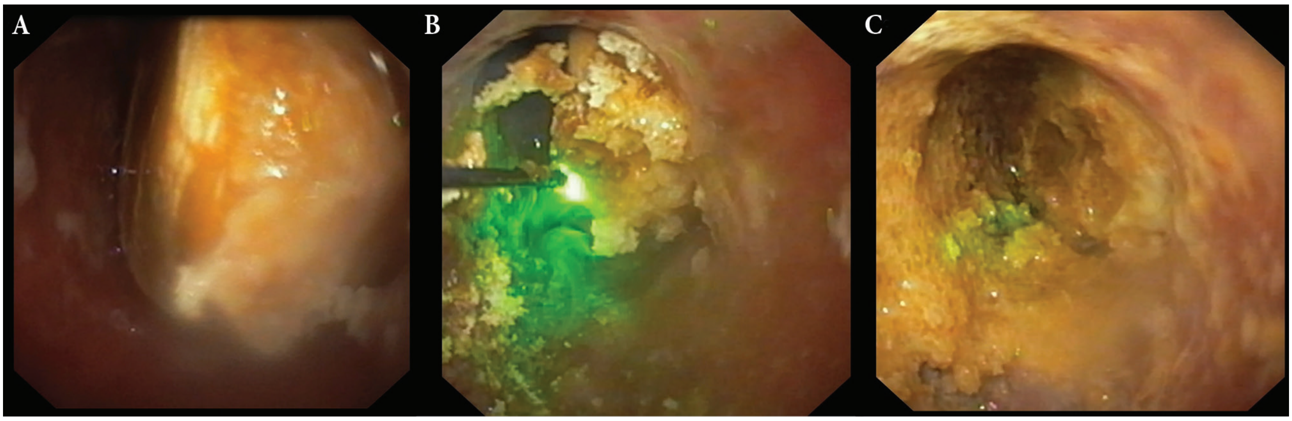

Figure 3. Laser lithotripsy of a gallstone impacted in the cholecystoduodenal fistula: **A**. Gallstone prior lithotripsy; **B**. Partially fragmented stone; **C**. Most of the stone is pulverized.

detecting the presence of GOO, in approximately 1/3 of the patients, it fails to establish the etiology, namely, to visualize a stone.[8]

UE successfully set the diagnosis in our case. Since it allows for both diagnosis and treatment, we might speculate that UE should assume a leading role in the multimodal management of BS patients.

Management options in BS consist of endoscopic and surgical treatment. Currently endoscopy is regarded as the first-line therapeutic approach in BS. The rationale for choosing endoscopy derives from the relative safety of the procedure compared to surgery (mortality and morbidity rates of 1.6% for endoscopy vs. 17.3%-25% for surgery, p=0.003).[4,10]

Endoscopic options in BS include either stone extraction using a snare, net or basket, or initial lithotripsy with subsequent extraction of the stone fragments. Lithotripsy techniques include mechanical, laser, electrohydraulic or extracorporeal shockwave lithotripsy. Simple extraction of the stone without fragmentation is a viable option in stones smaller than 2.5 cm, while for larger stones, lithotripsy is usually required as an adjunct technique.

Upon literature review, we established a total of eight cases reporting the usage of LL for management of BS.[11-17] This data explains the lack of clear guidelines on the application of LL in BS.

In the current article, we present a case of BS managed endoscopically using two lithotripsy techniques - ML and LL. To the best of our knowledge, this is the first report of two endoscopic lithotripsy techniques utilized in conjunction, specifically mechanical lithotripsy of a biliary stone held in the esophagus. Our experience suggests that biliary stones larger than the 2.5-cm threshold reported in literature (4 cm in our case), even if successfully negotiated into the stomach, should be fragmented prior to extraction to minimize the risk of esophageal trauma.

LL was utilized to fragment the second stone which was impossible to engage with a snare or basket. In our previous reports on LL of stones in the common bile (CBD) or cystic duct, we suggest that a 250-μm laser fiber ensures adequate power distribution while preserving excellent scope maneuverability. We advocate that higher power output settings might be used in BS cases (1.4 J/12 Hz) compared to common bile duct stones (0.8-1 J/10 Hz), mainly in relation to the considerably larger stone size. All stone fragments larger than 5 mm were extracted, which is postulated to be a major goal in order to avoid more distal gall-stone ileus.

The chief limitation of the endoscopic treatment is that adequate visualization of the obstructing stone is possible in only 60% of patients with BS. In our case, we found another factor which is worth considering – time. The total duration of the procedure was 210 minutes, which might be a major issue in patients with poor general condition and multiple comorbidities.

CONCLUSIONS

BS is a rare condition, which requires high level of expertise to manage. Endoscopy is a preferred first-line treatment. Stones larger than 2.5 cm should be fragmented prior to an attempt for extraction through the esophagus. LL is a safe and feasible option for BS patients, but it is time-consuming, which should be accounted for in the decision-making process.

Author contributions

Conceptualization: B.H. and M.D.; methodology: D.S. and E.T.; investigation: B.H. and E.N.; resources: D.D. and K.D.; data curation: K.K.; writing - original draft preparation: B.H.; writing - review and editing: S.V.; visualization: G.K. and P.U.; supervision: M.D. and S.V.; project administration: D.S. and E.T.; funding acquisition: D.D.

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Informed Consent Statement

Informed consent was obtained from the subject involved in the study. Written informed consent has been obtained from the patient to publish this paper.

Data Availability Statement

Data are available on request due to privacy restrictions.

Conflicts of Interest

The authors declare no conflict of interest.

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Комбинированная лазерная и механическая литотрипсия для эндоскопического лечения синдрома Бувере: отчёт о клиническом случае

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Резюме

Синдром Бувере (СБ) представляет собой чрезвычайно редкую клиническую нозологию, характеризующуюся обструкцией выходного отверстия желудка, вызванной прохождением жёлчного камня через холецистодуоденальный, холецистогастральный или холедоходуоденальный свищ и попаданием в двенадцатиперстную кишку или пилорус. Эндоскопия является предпочтительной терапией первой линии. Она имеет благоприятный профиль безопасности, но требует высокого уровня знаний для достижения очищения от камней.

Мы сообщаем здесь о случае 75-летнего мужчины европеоидной расы, у которого была рвота и кровотечение из верхних отделов желудочно-кишечного тракта, и которому был поставлен диагноз СБ с помощью верхней эндоскопии. Было выполнено односеансовое извлечение камня с использованием механической литотрипсии и лазерной литотрипсии. Было достигнуто полное очищение двенадцатиперстной кишки и жёлчного пузыря, успешно извлечены все фрагменты размером более 5 мм.

Редкость СБ и его неспецифическое проявление делают его в значительной степени упускаемым из виду, что обычно приводит к запоздалой диагностике, что приводит к высокому уровню смертности в рамках 12–30%. Компьютерная томография является диагностическим методом выбора. Верхняя эндоскопия может установить наличие обструкции, но не может визуализировать камень в 1/3 случаев. Лечение либо эндоскопическое, либо хирургическое. Эндоскопическое лечение имеет хороший профиль безопасности (смертность 1.6% против 17.5-25% при хирургическом вмешательстве), но низкий уровень успеха – 43% против 94.1%. Для достижения более высокого уровня успеха требуются более продвинутые методы, такие как механическая или лазерная литотрипсия.

Лазерная литотрипсия является возможным вариантом эндоскопического лечения СБ, но продолжительность процедуры может быть проблемой для ослабленных пациентов.

Ключевые слова

Синдром Бувере, холецистит, эндоскопия, жёлчные камни, лазерная литотрипсия, механическая литотрипсия