**CASE REPORTS**

**GALL-STONE ILEUS – OWN PATIENTS AND LITERATURE REVIEW**

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Cholecystolithiasis occurs in 10% of the US and Western European population, and its rare (0.3-0.5%) but a serious complication is gall-stone ileus (Bernard’s syndrome) (1). This constitutes 1-4% of mechanical obstructions of the small intestine, and in patients over 65 years of age it causes non-strangulation small intestine obstructions in as much as 25% of cases (2-6).

Epidemiology

The estimated morbidity in the population is 0.9/100,000/year (1), however women are affected more frequently (the women/men ratio is 4:1 and the proportion is the same as in uncomplicated cholelithiasis) (7).

The oldest patient with gall-stone ileus was described by Japanese authors after she underwent successful surgery at the age of 91 years (8). Apart from the age, female gander and large stones resulting from long-standing cholecystolithiasis (often with recurrent episodes of biliary colic), cholecystitis or mechanical jaundice (9), the risk factors include diabetes melitius and obesity (10).

Pathogenesis

The most common cause of obstruction is a gall-stone migrating through a cholecystoenteric fistula to the gastrointestinal lumen. According to literature reports, internal biliary fistulas constitute 2% of bile duct diseases (11) and they accompany cholecystolithiasis in 90% of cases (12).

The fistula is often a complication of endoscopic retrograde cholangio-pancreatography (ERCP), and the stone causing the obstruction may get to the duodenum after sphincterotomy without fistula formation (13, 14). Furthermore, the fistula may be caused by penetrating gastric ulcer, cancer of the stomach, gall-bladder, bile ducts, duodenum or pancreas, as well as trauma or Crohn’s disease (15, 16). It is worth emphasizing that prior cholecystec-
Gall-stone ileus – own patients and literature review

Complaints associated with cholecystolithiasis preceding the development of the fistula occur in 50-70% of patients (1, 18), however in the remaining patients (approx. 1/3) it is the first manifestation of the disease (19). The stone migrating through the gastrointestinal tract may cause symptoms of incomplete obstruction (4, 10) (so-called “ball valve effect”) (20).

The limit size of a stone that will be obstructed in the gastrointestinal tract is 2-2.5 cm (7, 8, 9), however, the described stones sometimes reach impressive sizes: 17 cm – Turner (21), 10 cm – Nakao (8), 9 cm – Iancu (3), 7 cm – Tucker (22). Smaller stones even in 80-90% of cases are spontaneously eliminated by vomiting or by natural pathway without clinical implications (3, 4, 17, 23), although there is a report of spontaneous elimination of a stone 5 cm in diameter (24).

The most common location of a stone causing obstruction is the terminal ileum 50-75% (50-90%) (9, 18). A more proximal obturation is less common – the obstruction of the initial section of the ileum and jejunum occurs in 20-40% of cases, of the stomach up to 15%, and duodenal obstruction (Bouveret’s syndrome) occurs in 1-3% of all gall-stone ileus cases. The obstruction may develop in the colon in 3-25% of cases (3, 9, 10, 18).

Diagnostics

Non-typical symptoms, usually the patients’ advanced age (thus, a wider range of comorbidities), and diagnostics taking into account neoplastic diseases (8) may prolong the diagnostics and delay the final diagnosis.

The symptoms may be present for several hours (6 hours) to even 2-3 weeks (15-22 days) prior to hospital admission (6, 19, 25).

Although hospital diagnostics causes further, even 2-week (14 days) delay of surgical treatment (19), correct preoperative diagnosis is made only in 50-60% of cases (25).

Plain abdominal X-ray

The details observed by Rigler are known as the classic triad: 1) pneumobilia, 2) small intestine distension (small intestine obstruction), 3) ectopic radiopaque gall-stone.

The presence of 2 of the 3 abovementioned symptoms indicates gall-stone ileus (18, 25). Other symptoms that may be added to this classic triad include: change in the location of a previously observed stone (5, 9) or visible double fluid level in the epigastrium representing the level of fluid in the gall-bladder and in the dilated stomach (9) (according to Balthazar, the fluid levels may correspond to the gall-bladder and duodenum) (26).

In literature reports, the presence of Rigler triad in plain abdominal X-ray widely ranges from 9 to even 87% (19, 27). If obstruction symptoms are not predominant, oral contrast administration may be of great diagnostic value as it informs of anatomic structures (fistula, duodenal diverticulum, non-radioopaque stone) and topographic relations of the diagnosed pathology (8, 9).

Abdominal ultrasound examination

Despite the difference in the quality of ultrasound scanners, as early as in the 1990s authors reported high effectiveness of stone visualisation even when obstruction symptoms were present (28, 29).

The lack of visibility of the gall-bladder in ultrasound examination in patients who did not undergo cholecystectomy and present with symptoms of obstruction may be a clue to suspect gall-stone ileus and to look for a stone in a different part of the gastrointestinal tract (especially in patients with earlier diagnosis of cholecystolithiasis or medical history suggesting calculosis) (29).

On the other hand, examination confirming the presence of cholecystolithiasis does not exclude earlier evacuation of other stone which currently causes obstruction. Moreover, when the gall-bladder is contracted and invisible, a large stone in the duodenum may be confused with an orthotropic stone (30). However, ultrasound-confirmed pneumobilia may provide a certain clue (18).

Abdominal ultrasound is undoubtedly of significant importance in diagnosing abdominal complaints in older patients who have the symptoms for a long time and do not manifest complete gastrointestinal obstruction (28).
An experienced ultrasonographer may show a cholecystoenteric fistula, as well as a stone impacted in the intestine (25). The sensitivity of the ultrasound examination in combination with plain abdominal X-ray reaches 74% (25), and even 96% if probable diagnoses are also taken into account (27).

Computed tomography (CT)

Computed tomography appears to be of greatest value in diagnostics of gall-stone ileus as it allows to properly localize an ectopic gall-stone in the intestinal lumen. However, also this diagnostic method may sometimes fail to provide a clear result (7). In their prospective study, Yu et al. prove high sensitivity and specificity of CT of 93% and 100%, respectively. Diagnostic accuracy in gall-stone ileus was 99% (19). Additional supply of oral contrast helps to identify the cause of obstruction (19). It is also obvious that when obstruction symptoms are developed, the administration of oral contrast is contraindicated. However, intravenous contrast may be helpful in the identification of oedema, inflammation and small intestinal ischaemia (19). A potential “flaw” of the CT is that 15-25% of gall-stones are non-radiopaque and their radiation absorption coefficient is the same as of bile or other fluids (30).

Magnetic resonance imaging (MRI)

In case of vomiting, sensitization to contrast agent as well as stones invisible in CT, MRI may be very useful, especially because it may visualize not only stones but also the fistula (9, 30).

Gastrointestinal endoscopy

Diagnostic endoscopy may be useful in the diagnosis of gall-stone ileus, provided that the stone is impacted in the stomach, bulb or descending part of the duodenum, in the large intestine or on the Bauhin’s valve. In 1976 Grove for the first time described a gastroscopically diagnosed case of pyloric occlusion caused by a gall-stone (12).

On the other hand, ERCP is the key tool in diagnosis of choledochoduodenal fistulas (12).

Treatment

In case of developed gall-stone ileus, surgery is the management of choice. Although one standard procedure has not been developed to date, three main strategies are usually adopted (7). The first and the most common of them is selective enterolithotomy (removal of the gall-stone by incision of the intestine) without secondary cholecystectomy and removal of fistula – this approach is supported by reports stating that 80-90% of stones remaining after primary enterolithotomy are spontaneously eliminated without any symptoms, and the majority of cholecystoenteric fistulas close spontaneously in the absence of residual calculosis (25). The second strategy is selective enterolithotomy with elective cholecystectomy and fistula removal. The third method is concurrent enterolithotomy, cholecystectomy and fistulotomy. Concurrent performance of the operations is justified by lower risk of gall-stone ileus recurrence, recurrent episodes of cholangitis and cholecystitis, as well as lower risk of gall-bladder cancer in patients whose fistula was not removed (31). Such procedure turns out to be necessary in case of gall-bladder gangrene or perforation in the area of biliary fistula (accompanied or not by peritonitis).

Surgical treatment may be also performed using laparoscopy or laparoscopy-assisted method (32). Moreover, the literature describes endoscopic treatment methods: endoscopic extraction of the stone (if impacted in the stomach or duodenum) or – with variable effect – endoscopic lithotripsy using a laser lithotripter (Holmium Yag Laser & Lithotripter System) or electrohydraulic lithotripter (33, 34). There are also reports of endoscopic extraction of stones from the duodenum to the stomach, from which they were removed by mini-laparotomy (35).

One of treatment methods quoted by different authors is ESWL – extracorporeal shock wave lithotripsy. The purpose of this non-invasive method usually used in urology is to break the stone, which then may be eliminated spontaneously, although sometimes it is necessary to endoscopically remove the fragmented stone (36, 37). Unfortunately, this method often requires several sessions before the stone is crushed, and shows poor effectiveness in obese patients and in the presence of
large amounts of intestinal gases (in practice it excludes the method in case of developed obstruction) (36, 37, 38).

**CASE REPORTS**

In 2011-2013 (27 months) in our Department of Surgery we operated 5 patients with gall-stone ileus, which is 4.6% of the total number of 108 patients operated during that period due to obstruction (excluding incarcerated hernias).

Finally, all patients underwent laparotomy with enterolithotomy and stone evacuation. One patient additionally had gall-bladder and cholecystoduodenal fistula removed. In most cases, the postoperative course was uncomplicated and treatment results were in each case satisfactory.

Some significant components of the medical history and additional tests are presented in tab. 1.

Case 1. The patient admitted to the Department as an emergency due to waves of intra-abdominal pain and vomiting with chyme. The complaints were accompanied by coprostasis and flatus. Plain abdominal X-ray showed single, not very clear levels of fluids in the right epigastrium. Abdominal USG showed heavy gastric stasis and insignificant dilation of the small intestinal loop in the epigastrium. The examination failed to visualise the gall-bladder.

Due to an unclear clinical picture, the patient underwent abdominal and pelvic CT scan with intravenous administration of a contrast agent. The examination showed non-dilated bile ducts with signs of aerobilia. An oval structure, 30 mm in diameter, forming a target sign was visualised about 15 cm from the duodenojejunal ligament (fig. 1). The radiologist describing the examination diagnosed intestinal intussusception.

The patient was operated – she underwent laparotomy and a stone of about 35 mm in diameter, which caused the obstruction, was found about 40 cm from the ligament of Treitz. The intestine above the stone was incised longitudinally on the antimesenteric side, the stone was extracted and the intestine was sutured transversely using a double-layer interrupted technique. A fistula was located between the gall-bladder and duodenal bulb. Due to purulent discharge from the fistula area, cholecystectomy was made and the fistula was removed. The opening in the duodenum was stitched with interrupted sutures and Heineke-Mikulicz pyloroplasty was performed. Complications were not reported in the perioperative period.

Case 2. The patient admitted to the department due to abdominal pain accompanied by vomiting with gastric contents after each attempt to drink.

A hernia bulge 5 cm in diameter, irreducible, quite hard but painless on pressure was found in the umbilicus during the physical examination.

Plain abdominal X-ray made in the sitting position showed few levels of fluids that might

<table>
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<td>Fluid level in the small intestine in X-ray examination</td>
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F – female, M – male, n.t. – not tested

Fig. 1. Abdominal CT. The stone is marked with an arrow
have corresponded to mechanical ileus, and linear radiolucencies in the view of the liver suggesting aerobilia (fig. 2) (features of aerobilia were found only during repeat postoperative analysis of the X-ray image).

The patient was operated after a short preparation. The umbilical hernia turned out not to be the reason of the obstruction. After extending the laparotomy, the stone obstructing the small intestine was located about 80 cm from the ligament of Treitz. A transverse enterotomy was performed to evacuate a gallstone 3 cm in diameter. The obstruction was relieved by the incision, then the intestine was sutured transversely using a double-layer, interrupted technique.

The gall-bladder was not palpable on examination and tissues in the area of the hepatic hilus and hepatoduodenal ligament were soft (many adhesions which were not dissected). The postoperative course was uncomplicated.

Case 3. The patient was seen in the admission room due to coprostasis for several days and an episode of vomiting. Plain abdominal X-ray did not show signs of obstruction. The patient received enema which was effective and improved her well-being. The patient was discharged from the hospital with the recommendation to see the doctor if the problems recur.

The patient reported to the hospital again after five days due to abdominal pain accompanied by vomiting, coprostasis and retention of gases. Plain abdominal X-ray showed low levels of fluid in the right umbilical region (corresponding with small intestine obstruction). Abdominal CT did not reveal the cause of the obstruction.

The patient was operated – an impacted oval stone causing the obstruction was found about one metre from the Bauhin’s valve. The intestine was incised longitudinally after the obstruction, and the concrement sized 2 x 3 cm was removed (fig. 3).

The obstruction was relieved by incision, then the intestine was sutured transversely using a double-layer interrupted technique.

Minor suppuration of the wound and transient incident of gastrointestinal subobstruction were observed in the postoperative course. In a transitional period, the patient required parenteral feeding for several days.

Case 4. The patient was admitted to the Department due to abdominal pain, for two days accompanied by vomiting with retained gastric content. Plain abdominal X-ray showed several levels of fluids in the small intestine in the left umbilical region.

The patient was admitted to the Department where he had a stomach tube placed. This revealed about 1,500 ml of stasis. Due to an unclear nature and the lack of clinical symptoms resolution, gastroscopy was performed the next day. It showed hiatus hernia and gastritis with quite serious cholestasis. Abdominal CT showed an oval lesion (63 x 36 mm) of heterogeneous, soft-tissue density, contacting the small intestine loop at the level of the upper edge of the left iliac wing – the radiologist interpreted the image as intussusception of the small intestine, GIST or foreign body (fig. 4).
Laparotomy was found to be necessary. When the peritoneal cavity was opened, it was found that ileum loops were dilated, and that there was an obstacle 80 cm from the ligament of Treitz. A longitudinal enterotomy was made above the obstruction and 2 x 3 cm gall-stone was removed. The incised intestine was closed transversely. The gall-bladder was difficult to examine, free from stones. The postoperative period was uncomplicated.

Case 5. The patient admitted to the Department of Surgery due to abdominal pain accompanied by vomiting with gastric contents, retention of gases and coprostasis. Plain abdominal and chest X-ray did not show significant deviations. Physical examination showed moderately marked peritoneal symptoms. The abdominal ultrasound examination showed dilated loops of the small intestine and serious gastric stasis. The gall-bladder was not visualised. Due to unclear symptoms, a decision was made to perform laparotomy. The stone obstructing the small intestinal lumen was located at half its length. The intestine was incised longitudinally above the stone and the stone 3 cm in diameter was removed. Then the intestine was closed transversely (fig. 5).

The gall-bladder was completely sealed by the omentum, no examinable stones were found. The postoperative course was uncomplicated.

**SUMMARY**

The mean age of the operated subjects was 80.2 years (76-84) and the mean hospitalization time was 11.8 days (7-24). Two men and three women were operated. Two of the patients had positive history of cholecystolithiasis (not operated to date). One patient was previously diagnosed with glucose intolerance, the remaining patients did not have diabetes mellitus.

Preoperative examination failed to determine the exact cause of the obstruction in any of the patients, and the correct diagnosis was made only intraoperatively. Despite good visualisation of stones in the CT, the image was misinterpreted by radiologists in two cases, and in one case (case 3) the stone was not described at all, although it was easily found in repeat postoperative analysis of the CT image (fig. 6).

A common feature of all ultrasound and CT scans was gastric stasis (fig. 7) and lack of ultrasound visualization of the gall-bladder. In one case (case 1), postoperative analysis of CT scans allowed to visualize a cholecystoduodenal fistula (fig. 8).

Although plain abdominal X-ray turned out to be poorly specific and not very useful in preoperative determination of obstruction cause, it was very helpful in surgical decision-making.

The common feature of the vast majority of cases is the image of serious obstruction (gastric stasis) and lack of gall-bladder visualization in both CT and ultrasound examination. Apart from elevated inflammatory parameters, laboratory tests did not show other common abnormalities.

**CONCLUSIONS**

Gall-stone ileus is still too rarely diagnosed preoperatively. It is not possible to indicate
laboratory parameters typical of the described disease and physical examination is completely non-specific.

CT scan appears to be the most sensitive imaging examination, although despite good visualization of the stone, the image is rarely correctly interpreted by radiologists. The fact of diagnosing small intestinal intussusception is also puzzling, as gall-stone ileus occurs with a similar rate (intestinal intussusception in adults 1-5% vs gall-stone ileus 1-4%) (39, 40). Plain abdominal X-ray still remains the basic examination at the admission room, although apart from visualising non-typical levels of fluids in the small intestine, it does not facilitate making an exact diagnosis. Subtle aerobilia signs were detectable only during accurate postoperative analysis of X-ray images and the Rigler triad was not present in any case.

Despite the lack of accurate preoperative diagnosis, the decision to make laparotomy seems to be justified and should be made as early as possible – longer diagnostic process does not lead to accurate diagnosis but only causes an increase in obstruction symptoms and deterioration of the patient’s condition.

Selective enterolithotomy is an effective treatment method especially in elderly patients as it allows to shorten surgery duration, limit the injury and brings good therapeutic effects. Concurrent enterolithotomy, cholecystectomy and fistulotomy are useful in case of fistula perforation or in patients with no medical history, in good clinical condition, relatively young (long expected survival time).

Bernard’s syndrome should be taken into consideration in differential diagnosis especially in older women with a history of biliary colic, presenting signs of obstruction or incomplete obstruction of the gastrointestinal tract.

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