The surgical treatment of a rare complication: gallstone ileus

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The authors describe 3 cases of gallstone ileus observed in two different hospitals and evaluate the current rarity of this complication of the biliary lithiasis and/or of cholangiocarcinomas. There were two cases of stones at the ileal level and one case in the left colon. In one case a single surgical intervention was carried out while in the other a two-step strategy was adopted. Notwithstanding the fact that the correct strategy to adopt is still controversial in literature, the authors discuss the single and double step strategies and propose that the second intervention of the 2-step strategy should not be performed in high risk surgical patients.

KEY WORDS: Biliary fistula, Enterotomy, Gallstone ileus, Rare complication.

Introduction

Gallstone ileus is a rare complication of the biliary lithiasis that is not often seen today thanks to the diffusion of ultrasound diagnostic techniques and the use of the "gold standard" of laparoscopic cholecystectomy, thus the incidence of this pathology has dramatically decreased over the last 20 years. It has been observed that the occasional occurrence of this rare disease persists in some rural areas due to cultural reasons or poor access to diagnostic technologies, leading to the possible development of this late complication of cholelithiasis [1,2]. The clinical presentation of gallstone ileus is often ambiguous and can have a notable diagnostic delay; the surgical treatment is still the object of controversy in literature. The aim of this work was to present three cases diagnosed and treated with the differentiated approach for surgery in two different hospitals.

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Case reports

CASE N. 1

C.P , a 75 year old female reported abdominal pain of the colic type localized at the right iliac fossae of about one month, with associated nausea and occasional temperature up to 39°C. Due to the increase in pain the patient was hospitalized in the Week-Surgery unit having had no bowel movement for about 7 days. At the work up, the patient presented a low positivity for arterial hypertension, diabetes, myocardial infarct, with dilational cardiopathy and moderate concentric hypertrophy of the left ventricle, poor bowel movement, and dehydrated. She presented an acute swollen abdomen, painful to deep palpation in the right quadrants. Accentuation of the ileocolic timpanism. Absence of peristaltic sounds. The hematic results showed notable Leucocytosis (18.000 GB, with neutrophilia 87%), Piastrinosis (455x 10^3/mmc), hyperfibrinogenemia (608 mg/dl), PCR 13.20 mg/dl. The x-ray of the abdomen showed the presence of air –water levels on the right side, in the right iliac fossae and in the hypogastric and mesogastric areas. In the pelvis other than numerous fecal residues the x-ray showed the presence of an oval opacity of about 5 cm diameter with calcified borders. The following day, a CT

Fig. 1: Case 2 - CT scan of the abdomen: Image showing stone in the ileum. Presence of gastric ectasis.

Fig. 2: Case 2 - Intraoperative diffused dilation of the ileal loops above the obstruction.

scan of the abdomen was carried out that showed air in the biliary tree, in particular in the left branches, air bubbles between the anterior margin of the head of the pancreas and the right flexure of the colon. The gallbladder was not visible. There was also a calcium dense roughly round formation of 33 mm diameter in the lumen of the sigmoid colon. There was also diverticulosis of the colon above all sigmoid, with diffused signs of peridiverticulitis. It was therefore decided to carry out emergency surgery.

Surgery: after having carried out an xifo-pubic laparotomy, the exploration of the peritoneal cavity showed notable dilation of the transverse colon and the caecum, suggesting the presence of an endoluminal obstacle firm to the touch in the sigmoid colon, with the presence of a flogistic tumour in the right colic flexure and a notable state of retraction of the omentum. Due to the general state of the patient it was decided to perform a transverse colotomy with the extraction of a lithiasic formation of 9 x 4 cm (that was moved backward to the transverse colon). A double-barrel colostomy was performed. Post-operatively the patient's blood pressure rose to 200/80, there was transitory respiratory insufficiency and an anterior-septal infarct that kept the patient in the intensive care unit. The patient was discharged in good condition on the 19th day post-operatively. At three months from the first surgical intervention colostomy closure was carried out but the patient refused to undergo definitive treatment of the biliary fistula and of the peridiverticulitis. At three years from surgery the patient reports only slight dyspepsia.

Case N. 2

C.C., a 79-year-old female referred to our department from the neurological clinic suffering from senile dementia, arrived after seven days of hospitalization. She had temporal spatial disorientation and sensorial obnubilation. During the seven days in hospital, she presented constipation with poor bowel movement, with the last bowel movement 3 days before. During surgical work-up, the patient complained of intense pain of about three day in all the quadrants of the abdomen. Under examination, her abdomen was found to be distended and acute (tender and hyperextended at superficial palpation), with intense pain at deep palpation in all the quadrants of the abdomen, in particular in the epigastrium and right hypocondrium. Accentuation of the ileo-colic tympanism. Slow peristalsis. The laboratory examinations carried out when she was transferred to our unit showed: The abdominal x-ray revealed no air-water levels or air bubbles under the diaphragm, but only a slight gastric and ileal distension. The CT scan of the abdomen showed: gallbladder with thickened walls, air bubbles and defused dishomogeneity of the pericholecystic fat, not dissociable from duodenal-C, which showed an increase of the lumen with internal lithiasic concretions, irregular lithiasic concretions of 38x27 mm localized in a loop of the small intestine, and loops of the small intestine dilated extending to the left paramedian area (Fig. 1). From the CT scan a diagnosis of gallstone ileus was made and the patient was sent to surgery: after carrying out the xifo-pubic laparotomy, and the exploration of the abdominal cavity, a flogistic tumour of the right hypocondrium was found associated with the liver and duodenal-pancreatic complex, with a notable state of jejunal and ileal loop distension (Fig. 2), to the point where the stone was palpable through the intestinal wall. Longitudinal enterotomy was carried out with the extraction of the stone and resolution of the ileus (Fig. 3-4-5), with interrupted stitches transverse enterorraphy. The post-operative course was regular even if the patient was discharged at the 10th day post-operatively due to a delay

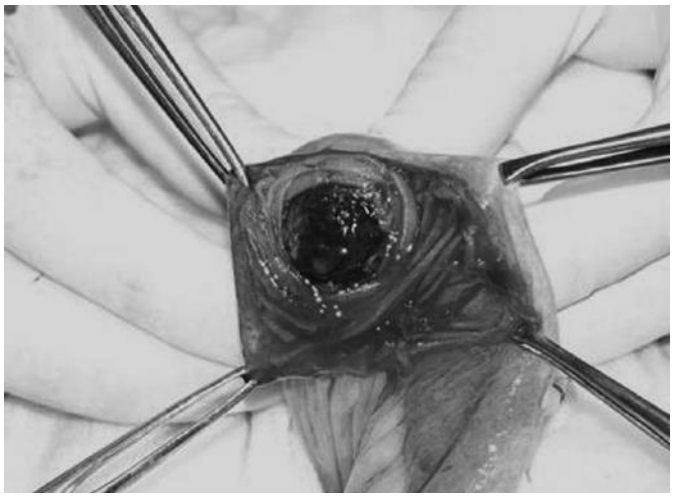

Fig. 3-4: Case 2 - Enterotomy with stone extraction.

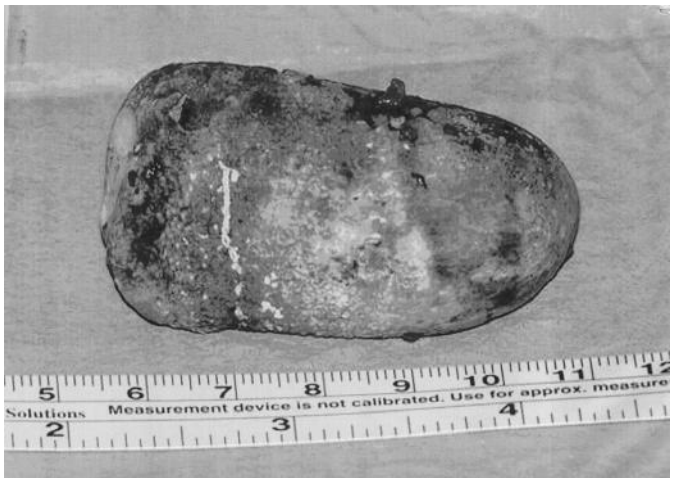

Fig. 5: Case 2 - Stone 5x3 cm.

in the re-establishment of the coagulative parameters. At the clinical and echographic observations carried out after one year from surgery the patient was asymptomatic.

CASE N. 3

R.G., a 75-year-old female was hospitalized in the section of general surgery of the University Hospital of Cagliari. At admission the patient complained of intense pain startied 20 days before that then decreased. The patient had no bowel movement and abdomen was notably stretched but not painful. The clinical work- up showed a clinical history of cholelithiasis with numerous colic episodes. Abdominal x-ray showed the presence of numerous ileal air-water levels and the diagnosis was intestinal occlusion (at the level of the small intestine). The echographic exam showed a chronic lithiasic cholecystopathy with the suspected presence of a fistula. An emergency explorative laparotomy was performed, that showed a gallstone ileus caused by a stone at about 1 m from the ileo-cecal valve. The fistula was a cholecystoduodenal fistula (between the infundibulum of the gallbladder and the 2nd duodenal portion). It was decided to perform a cholecystectomy, with papillo-sphincteroplasty, closure of the fistula and sinking of the duodenal stump with Ileal resection and termino-terminal anastomosis (single surgical intervention). The stone was cylindrical and measured about 4 cm in length and about 2.5 cm in diameter. Moreover, inspecting the biliary tree 2 more stones were found in the biliary tract (that were removed) and two in the gallbladder. An intraoperatory trans-papillar retrograde cholangiography was carried out with negative results for the presence of further concretions. Dual abdominal drainage and nasal-gastric tube were inserted. The postoperative course was uneventful, and the patient was discharged after 15 days.

Discussion

The first case of gallstone ileus was described by Bartolini in 1654, however, a complete description was only given by Coirvoisier in 1890, when this author reported 131 cases undergoing surgery with a mortality of 44% [5,6]. In 1896 Bouveret described a higher variant with early vomiting, due to wedging of the stone at the high duodenal level just under the pyloric sphincter. Today gallstone ileus is considered a rare complication of the biliary lithiasis (only 0.4% of the patients are affected by cholelithiasis)[8,9], and, furthermore, it seems to be responsible for about 25% of the occlusions of the small intestine in the absence of strangulation in the over 60s [10]. An association between gallstone ileus and Caroli's disease [11], or with Mirizzi's syndrome [12] has also been

documented; the latter association induced Csendes ed al. to formulate a pathogenetic theory unifying the two pathological conditions, and consequently creating a new classification of Mirizzi's syndrome [1,13,14]. However, these epidemiological data are from about 20 years ago, and it is presumable that they are an over estimation of the current situation, given the diffusion over the last 20 years of early ultrasonic diagnosis of gallstones, and consequently of the preventive surgical treatment due to the the laparoscopic "gold-standard". This clinical condition is found more frequently in females (85%), with a female/male ratio of 6:1. The average age of presentation of the clinical symptomatology is from 66 to 73 years. The pathogenetic mechanism is that of the formation of an adherence between the gallbladder and the duodenum or colon and then of a well described cholecysto-duodenal or cholecysto-colic fistula at the diagnostic image level known as perforation of the 3rd type according to Niemeier[15]. However, only 6-14% of the bilio-enteric fistulas would lead to an ileus; in fact, it seems that the majority of stones are eliminated in the feces or with vomiting and only in 10-15% of the cases do they have a diameter sufficient to cause intestinal occlusion. In about 4% of the cases stones are multiple [17]. The site where the progression of the stone in the intestinal lumen stops is variable. This is based on: the site of the fistula formation (cholecysto-duodenal, cholecysto-colic), the dimensions of the stone, the motility of the intestinal tract (ileal occlusions), the presence of stenosis and/or concomitant pathologies (stenosis from colic diverticulosis would frequently stop the stone at the level of the sigmoid colon in the cholecysto-colic fistulas). The condition of gallstone ileus in the presence of a double cholecyst-duodenal-colic fistula is extremely rare (to date, only seven cases) [18,19,20]. Clinically, the ambiguous characteristic of gallstone ileus is the absence for a long time of symptomatology, as well as episodes of cholecystitis (about 50% of patients do not have a known history of previous gallstones). This can be explained by the development of a chronic flogosis, clinically silent until the sudden migration of the stone and the beginning of symptomatology. Today, diagnosis is generally made after a scan [21] that can sometimes give pathognomonic indications (Triad of Rigler: 1.occlusions of the small intestine, 2.biliary air, 3.ectopic gallstones) [22], to which we can add the fourth sign of Balthazar et al. (The presence of air in the gallstones and two air-water levels in the gallstones and duodenal bulb) [23]. From the surgical point of view, there is still some controversy about the timing of surgery and which surgical strategy is the most suitable. It is possible to carry out surgery in a single phase (resolution of the obstruction and cholecystectomy with eventual repair of the cholecysto-duodenal or cholecysto-colic fistula); or surgery in two phases; the first to resolve the obstruction (enterotomy and removal of the stone and/or intestinal resection), followed by a second-look with repair of the cholecysto-duodenal or cholecysto-colic fistula. The advantages of single phase surgery are obviously linked to the definitive character of the surgical treatment, even if there has been demonstrated a notable post-operative morbidity and mortality (7-25%) [3,10,24]. On the contrary, the two-step surgery has the advantage of initially resolving the intestinal occlusion, and putting off the surgical repair of the cholecysto-duodenal fistula to the second step with morbidity and mortality notably inferior if surgery is carried out by election and not in emergency. The problem of the choice of therapeutic strategy, in our opinion, is conditioned by the convictions and orientation of Surgical School, age and general condition of the patient. While in our third case, the 20 days of painful symptomatology in the right side, ileus had started for some hours and thus the general conditions of the patient were not bad and allowed single step surgery; in the other two cases described, the patients came to our unit after many days from the start of the initially incomplete occlusive symptomatology. Consequently, the coagulative problems, the state of general dehydration, the hydro-electrolitic imbalance and the co-morbidity of both patients (above all from the cardiological point of view), forced us in both cases to adopt the most rapid and safest solution, with the least post operative morbidity and mortality, leaving the eventual necessity of fistula repair to an eventual second-look. In these two cases, the clinical and echographic observations in a variable period from 1 to 3 years did not indicate the necessity to operate the biliary lesion. However, in literature, there are three reasons that would sustain the necessity for fistula repair in a second step: occlusion relapses, recurrent angiocolitis from previous infections, and the risk of developing cholangiocarcinoma that is about 15 times higher in patients with cholecysto-enteric fistula with respect to the general population. It must be noted that ileus relapse can be avoided by a careful intestinal washing above the obstruction (as we did in the second case), more so with a colostomy (as in our first case), while angiocolitis is an outcome that is inconsistent with these fistulas. If we consider that only 14% of the biliary-enteric fistulas lead to ileus, the remaining 86% are not diagnosed and remain clearly silent, indicating an over-estimation of the frequency of the episodes of angiocolitis as demonstrated by the autoptic data from the paper by Hildebrandt et al. [25]. This finding would not justify surgery *tout court*. As concerns the risk of cholangiocarcinoma, this conviction comes from the papers published in the 1960s by Berliner et al. and Bossart et al, who showed that in patients with duodenal-cholecystic fistulas from biliary ileum the incidence of cholangiocarcinoma was 15 times higher than in the subjects operated only once (15% vs 1%)[26,27]. These observations have never been repeated or confirmed; recently, for example, 2% of cases of cholangiocarcinoma have been reported in patients with cholecysto-colic fistula[28]. On the other hand, there have been descriptions of cases of

gallstone ileus caused by a migration of the stone to the duodenum by a neoplastic fistula from cholangiocarcinoma (6% of the cases of gallstone ileus) [29,30]. Therefore, there is a doubt that these cases could, really, be cholangiocarcinomas with a sudden gallstone ileus and not intraoperative diagnosed due to the natural tendency of the surgeon to resolve the flogistic tumour of the biliary tract later choosing the strategy of two steps and in the meanwhile the pre-existing cholangiocarcinoma becomes manifest [31]. It should be noted that even if we had programmed the second step of surgery in two of our cases, there is the question, considering the epidemiological data cited, if it was really necessary, given the absolute absence of symptomatology in both cases at one and three years from the first surgery. In fact, only in 5% of cases is there a relapse within the first six months and only 10% need surgery for cholecystitis[10]. Finally, there seems to be evidence of the spontaneous closure of the fistula, according to some reports up to 50% of the patients who undergo only enterolithotomy, compared to the rate of cholangitis of only 11% [3]. It is our opinion that in older patients at high surgical risk (that constitute a high percentage of the patients with gallstone ileus, as in two of our cases), after six months from surgery for the resolution of the occlusive symptomatology, in the absence of relapse and/or angiocolitis, it is necessary to carefully evaluate the necessity of a second-look. These patients should undergo duodenal plastic surgery, extensive colic resection or reconstructive surgery of the biliary tract (this is rather difficult as there is the frequent association with Mirizzi's syndrome, above all of the IV type)[14,32] with the inevitable morbidity and mortality not comparable with the low-risk of recovery from the billiary disease (5% of cases).

Conclusions

Gallstone ileus is today a very rare complication of the lithiasic disease of the biliary tree. The choice of surgery needs a holistic vision of the problems of the patient, a balancing of costs and benefits for the single surgery strategy (certainly resolutive), or the necessity to wait and see with surgery carried out in two steps. It is the opinion of the authors that in very old patients, with high surgical risk, asymptomatic after the first surgical step of removal of the occlusive obstacle, as well as at a follow-up of at least one year, in selected cases, the second surgical step is not necessary, maintaining the patients under clinical and echographic monitoring.

Riassunto

Gli Autori descrivono 3 casi di ileo biliare, osservati in due strutture chirurgiche differenti e valutano l'attuale rarità di tale complicanza della litiasi biliare e/o dei colangiocarcinomi. Trattavasi rispettivamente di due casi con arresto del calcolo a livello ileale ed uno a livello del colon sinistro. In un caso è stata adottata la strategia in un tempo, mentre negli altri due ci si è orientati per un trattamento in due tempi. Tuttavia per motivazioni diverse nei due casi con strategia in due tempi non è stato eseguito il secondo tempo di chiusura della fistola biliare. Nonostante sia ancora oggetto di controversia in letteratura quale delle due strategie sia la più idonea da adottare, gli Autori discutono della tattica chirurgica in uno o due tempi esponendo vantaggi e svantaggi di ambedue gli approcci sulla base delle evidenze scientifiche della letteratura, prospettando l'opportunità di un'astensione dal 2° tempo nei soggetti anziani, asintomatici con un elevato rischio operatorio e proponendo per essi un follow-up clinico ed ecografico.

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