

Gallstone Ileus of the Sigmoid Colon: A Rare Complication of Gallbladder Calculosis Typical of Advanced Age. Report of a Case

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Abstract

Sigmoid colon gallstone ileus is a mechanical large bowel obstruction caused by a biliary stone which migrates into the colon through a cholecysto-enteric fistula and gets impacted in the sigma. It is a very rare complication of gallbladder calculosis in the general population, but its frequency increases with age, with elderly people being the most frequently affected. Diagnosis is often delayed because of the prevalent symptom aspecificity, and the choice of the optimal surgical treatment is still debated, especially as the elderly have increased operative risk also due to multiple comorbidities. The most frequently employed procedures are: the enterolithotomy alone, aimed at just removing the obstacle and solving the occlusion; the one-stage procedure, where enterolithotomy, cholecystectomy and fistula repair are performed in the same operation, and the two-stage procedure, where enterolithotomy is performed first and cholecystectomy at a later stage. We report the case of a sigmoid colon gallstone ileus in an 81 year-old female patient with multiple comorbidities, who was treated with enterolithotomy alone-with residual gallbladder stone-to minimize the operating time and risk. A successful outcome of the intervention performed in our case is reported at a 1-year follow-up, the patient was in a good clinical status at the medical checks performed and had experienced no recurrences of ileus and no new pain symptoms from the gallbladder. The case and its outcome are discussed in the light of the background literature on pathogenesis, symptomatology, diagnosis and treatment of sigmoid colon gallstone ileus.

Keywords: Elderly people; Sigmoid colon gallstone ileus; Surgery; Operative risk

Introduction

Gallstone ileus is a very rare complication (0.3%-0.5%) of gallbladder calculosis caused by the passage of a stone through a gallbladder-enteric fistula [1,2]. It occurs preferentially in the female sex, with a 4:1 women/men ratio, and individuals of advanced age, the mean age of affected patients varying from 74-80 years (age range 60-84 years) [3-8]. The first description of gallstone ileus was provided by the Danish physician Erasmus Bartholin in 1654 [in 9], in a case of gallbladder perforation into the jejunum. In 1890, Courvoisier [10] published the first review on the topic, reporting 131 cases. In 1914, von Wagner [11] reported a total of 334 documented cases at that time and in 1955, in a review of gallbladder ileus, Deckoff [12] stated that the total number of cases in the literature at that date was approximately 555. More specifically, sigmoid colon gallstone ileus, due to a gallbladder-colon fistula, represents 2%-8% of all cases of gallstone ileus [13]; its rarity is confirmed by Anselme [14], publishing a case report on the topic, who ranked it as 57th in the international literature. Because of this rarity, the surgical approach is to date not yet standardized, with different interventions being performed and variable outcomes in terms of survival, complications and recurrences in the cases reported so far [15-19]. On the other hand, with the progressive aging of the population and the increase in the frequency of biliary calculosis, the choice of the most appropriate surgical

treatment is becoming of crucial importance in this condition, especially considering that the preferentially affected patients are elderly, in most cases with multiple comorbidities and a critical general health status [7,15,20]. We report here a case of sigmoid colon gallstone ileus in an elderly female patient in whom a minimally invasive surgical procedure was successful in terms of both survival and lack of recurrences. Based on these results, we aimed at critically discussing the different available surgical approaches to gallstone ileus of the sigmoid colon in the light of the previously reported studies in the literature.

Case Report

An 81-year-old female patient was hospitalized (Medical Unit) because she had been complaining for 10 days of alvus closed to faeces but not gas, abdominal distension and pain in the lowest abdominal quadrants. She had a clinical history of ischemic cerebral stroke 9 years previously, involving left hemiparesis followed by partial functional recovery, hysterectomy for ovarian cancer 6 years before, followed by pelvic irradiation, gastritis, hypercholesterolemia and hypertension. She was under current medical treatment with acetylsalicylic acid (100 mg/day), atorvastatin (20 mg/day), ramipril, (5 mg/day) and lansoprazole (30 mg/day).

At admission, abdominal examination revealed abdominal distension and tenderness of the lowest quadrants at both superficial and deep palpation; arterial blood pressure (PA): 160/80, Heart Rate (HR): 72/min and no fever. Hematochemical analyses showed:

erythrocyte sedimentation rate (ESR) 86 mm/h, prothrombin time (PT): 72%, I.N.R: 1.12, white cell count $11.140/\text{mm}^3$ (neutrophils 73.9%), hemoglobin 11.9 g/dl and platelet count $207 \times 10^3/\text{ul}$. Plain abdominal radiography showed marked distension of the intestinal anse, mainly at ileal level, with multiple hydro-aerial levels. Ultrasound imaging, though partially impaired by the intestinal meteorism, showed a gallbladder lumen completely occupied by lithiasic material. Plain abdominal X-rays were repeated the following day, with results similar to those of the first examination; a subsequent abdominal CT-scan without contrast medium documented: a 3.1 cm round hypodense, semisolid formation within the distal descending colon, with neat, regular walls, partially calcified (phytobezoar?); modest gaseous colon distension through the cecum and several fecal residues; a gross lithiasic formation (about 3.3 cm) within the gallbladder, which appeared sclero-atrophic; mild pneumobilia in the left biliary duct. The patient was meanwhile being treated with oral laxatives, enemas, antibiotics, analgesics on demand, and intravenous nutritional support during hospitalization, with partial attenuation of symptoms. A further abdominal plain X-rays performed on the fourth day documented a reduction of the hydro-aerial levels detected at the first examination and persistence of moderate colon distension. Medical therapy was continued, but symptoms exacerbated again over the following days and therefore a colonoscopy was performed on the 11th day, showing a suspected phytobezoar occluding the sigmoid lumen; repeated attempts to break, move or retrieve it failed.

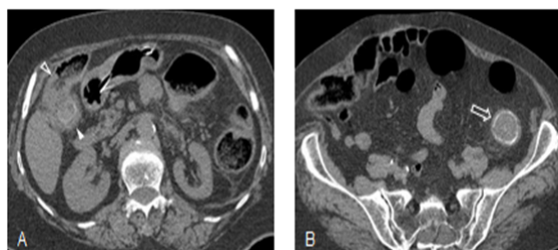


Figure 1: CT scan of the abdomen. Axial sections of the CT scan show the fistula (A) between the gallbladder (full arrowhead) and the right colon (empty arrowhead) as well as the intracolonic gallstone (B) (arrow). The gallbladder lumen contains a residual gallstone (full arrowhead in A).

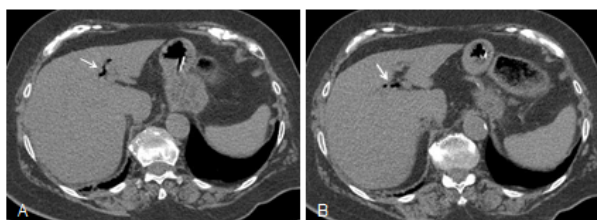


Figure 2: CT scan of the abdomen. Axial sections of the CT scan show air within left (A) and right (B) hepatic ducts. The presence of pneumobilia is one of the signs indicative of gallstone ileus.

The patient was thus transferred to our Surgery Unit to undergo an urgent explorative laparotomy. An umbilico-pubic incision was performed on the previous surgical scar; upon opening the

peritoneum, dilation was observed of the small bowel anse, while palpation revealed the presence of a round, solid formation impacted at the colon-sigmoid junction. This was not mobile and could not be pushed distally towards the rectum with digital manoeuvres. A longitudinal 5 cm colotomy was therefore performed above this formation, at the level of an antimesenteric toenia; a huge biliary stone (about 4 cm in diameter) was seen; through manual squeezing this was maneuvered out of the colon incision and removed (Figure 4A and 4B).

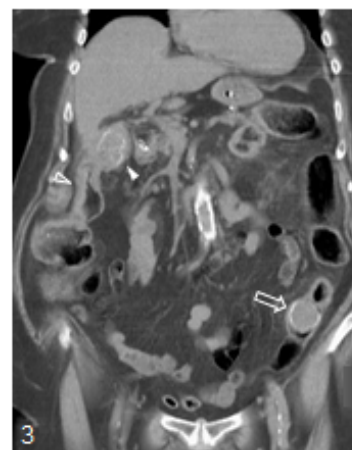


Figure 3: CT scan of the abdomen. Coronal reconstruction shows the fistula between the gallbladder (full arrowhead) and the right colon (empty arrowhead) as well as the intracolonic gallstone (arrow). The gallbladder lumen contains a residual gallstone (full arrowhead).

A longitudinal double-layer suture was then performed. The gallbladder-colon fistula was identified but not surgically treated in order to minimize the operating time and therefore the risk of post-operative complications (patient with an ASA III-IV anesthesiologic risk). The duration of the intervention was 95 min. The patient recovered well from surgery, experiencing no medical or surgical complications and no significant postoperative pain (max 4 cm on the Visual Analogue Scale-VAS). She was discharged from hospital on the 8th postoperative day in satisfying physical conditions. One year after surgery she was still alive and in a good clinical status at the medical checks performed. She experienced no recurrences of gallstone ileus and no new pain symptoms from the gallbladder.

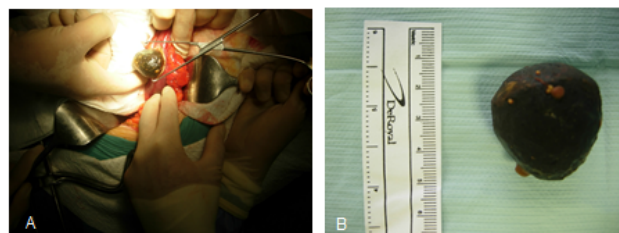


Figure 4: Enterolithotomy at sigmoid level: intraoperative view (A). Gallstone of about 4 cm. extracted from sigmoid colon (B).

Discussion

The reported case of gallstone ileus of the sigmoid colon in an emergency condition in an elderly patient shows that a surgical approach of enterolithotomy alone, which reduces the operative time to a minimum, is successful in terms of both survival and lack of complications and recurrences.

Gallstone ileus represents 1-4% of the causes of mechanical intestinal occlusion in the general population, but can reach values of over 25% as the cause of mechanical small bowel obstructions in patients over 65 years [2,7]. The stone migrates through a gallbladder-enteric fistula; the duodenum is the most frequently affected tract (68%-80%) [2,21], while the colon is involved much more rarely (4% on average) [12,17]. The impact can occur anywhere in the gastroenteric tract when the stone diameter is over 2.5-3 cm.

The formation of the fistulous connection would start after episodes of acute cholecystitis from obstruction of the cystic duct [22,23] which remain silent or misdiagnosed mostly in the elderly due to the reduced sensitivity to visceral pain with the aging process, as happened in our patient, who had never complained of biliary pain in the past [24-28]. In the absence of medical treatment, these episodes, if recurrent, would be followed by hydrops/empyema. With the pericholecystitis, the inflammatory involvement of the gallbladder serosa would produce adhesions with the serosa of the nearby organs, such as the duodenum or colon [12]. In the formation of the cholecysto-colonic fistula, almost always at the hepatic flexure, a fundamental role is played by stone size; most case reports of gallstone ileus in the sigmoid colon, in fact, refer a stone size of over 3 cm [14,29-33]. The weight of a big stone and the friction on the already inflamed wall of the gallbladder would promote local ischemia and erosion of the area; the further intervention of gangrenous processes would then favor ulceration of the adhered wall with creation of the fistula within the colic wall [34]. The fistula permits the resolution of a possible empyema, through the voiding of the puruloid content into the colic lumen, with consequent migration of the stone into the colon. After a trajectory along the colon, the stone impacts in the sigma, both because this is the narrowest tract of the colon, and because of post-diverticulitis stenosis or fibrosis secondary to previous operations on the uterus and ovaries or because of prior pelvic irradiation, as in our case.

The clinical presentation of gallstone ileus is almost always aspecific [35]. The symptoms can be initially vague and insidious, with a sub-occlusive clinical picture that can last for several days, delaying the diagnostic process, as happened in our patient. This is due to the fact that the stone tumbles down the colon and into the small bowel ileus, causing intermittent partial subocclusions and occlusions (the so-called tumbling phenomenon) with gas and feces which can be expelled for some time either spontaneously or with the use of enema, again as in our case. With the definitive impaction of the stone in the sigmoid colon, the occlusive symptoms prevail, with abdominal pain, nausea, alvus closed first to feces and then to gas, and vomiting. The high mortality reported in many case series of gallstone ileus (12%-18%) [7,15] has also been attributed to a delayed preoperative diagnosis, due to the strong limitations of plain abdominal X-rays (3%-12.5%) [4,36] in distinguishing promptly and clearly the Rigler's triad-ectopic stone, intestinal obstruction and pneumobilia [37]. In our case, in fact, the initial diagnosis was phytobezoar. The combination of ultrasounds and plain abdominal X-rays has significantly increased diagnostic sensitivity (74%) [36,38]; however, the radiologic gold standard for the preoperative diagnosis is represented by the CT scan, due to its very high sensitivity (93%) and specificity (100%) [39,40].

Furthermore, the CT slides are very often able to identify the fistulous connection, as in our case, an element which could be considered an additional criterion to the Rigler's triad in the diagnosis of gallstone ileus.

The final clinical picture of the gallstone ileus in general and sigmoid colon ileus in particular, is that of an acute abdomen, generally in very old patients with often serious concomitant diseases, and an often critical clinical situation with hemodynamic instability, requiring emergency surgery. The most controversial aspect of the surgical debate about gallstone ileus regards the choice of the most appropriate intervention [16]. The main current surgical procedures are: enterolithotomy alone (EA), whose principal objective is to just remove the obstacle and solve the occlusion; one-stage procedure (OSP), where enterolithotomy, cholecystectomy and fistula repair are performed in the same operation, and two-stage procedure (TSP), where enterolithotomy is performed first and cholecystectomy is performed later, an intervention reserved to patients remaining symptomatic after enterolithotomy alone [15]. Bowel resection is necessary in certain cases after enterolithotomy is performed.

In an effort to circumvent surgery, especially in the elderly where open procedures involve a high risk of morbidity and mortality, some authors have tried less invasive techniques, i.e., colonoscopy to access the gallstone and relieve the obstruction by fragmentation employing electrohydraulic lithotripsy (EHL) [41-43]. Though encouraging results have been reported, the applicability of these techniques remains limited, as a positive outcome is strongly dependent on the size and composition of the impacted gallstone as well as on the local expertise of the operators. Minimally invasive surgical procedures, i.e. laparoscopic, have also been attempted; here again, in spite of positive outcomes being reported, these are limited experiences and further future studies with a wider number of patients are needed for confirmation [44-47].

The surgical treatment approach to gallstone ileus remains controversial in the literature. Some authors are in favor of enterolithotomy alone; they prefer to solve only the occlusion during the emergency laparotomy, using this shorter and less complex surgical approach, and postponing the possible re-operation on the biliary tract (two-stage procedure) only in symptomatic patients [7,8,19, 38, 48-51]. Simple enterolithotomy has indeed long been associated with a lower mortality [7]. Other authors prefer the one-stage procedure, to solve the occlusion and intervene on the biliary tract in the same emergency sitting, claiming that this would avoid recurrences of the gallstone ileus, the occurrence of cholecystitis, cholangitis and the development of gallbladder cancer [50,52,53]. While there are pros and cons in both EA and OSP, a number of data show that the objections against enterolithotomy alone are not fully justified. Regarding recurrences with EA, in fact, although some report percentages of 17% in limited case series [2], the data on the amplest case series, such as those by Reisner and Cohen [7], report values of less than 5%, with less than 10% of the patients requiring re-intervention for biliary symptoms. Furthermore, the fistula closes spontaneously in most cases, once the stone has passed and the occlusion is solved [54], as verified in the course of re-operations for other reasons, or at autopsies of patients that had already undergone EA [7,12]. This is probably what happened also in our case, considering the lack of recurrences and of biliary symptoms one year after EA in our patient, although a residual stone was present in the gallbladder. Spontaneous closure of the fistula can be explained by the fact that the inflamed walls of the empiematous or gangrenous gallbladder become gradually fibrotic, embedding the

residual stone, the gallbladder becoming, in the end, atrophic and de-functionalized [12,55].

Only a few cases have been described of late complications associated with persisting fistulae, not treated during the first operation [12]. McQueeney [56] observed, instead, in the long-term follow-up of his casuistry, that two patients with a persisting cholecysto-enteric fistula were in good health and experienced no recurrences after 19 and 15 years, respectively. Furthermore, data from the literature report that 60% of the recurrence of gallstone ileus occurs within 30 days after surgery [57].

Regarding the possible development of gallbladder carcinoma, which, according to some authors, would be facilitated if the fistula is not closed during the first intervention, data from the literature are not homogeneous, and the high incidence reported by some authors (15% in the cases reviewed by Berliner and Burson) [52] is not confirmed by others (6.2% by Rodriguez-SanJuan et al., 4% by Day et al.) [50,58]. In any case, the possible benefits of the cholecystectomy and fistula repair at the same time as the enterolithotomy need to be weighed against the actual risks of this intervention, especially in the elderly, with compromised general conditions and often signs of peritonitis, when it is an absolute priority to solve the occlusion and save the patient's life in the shortest possible time [30,50,59]. Many authors believe that once the fistula is formed, and the stone has already passed into the intestine, during surgery one has to expect a gallbladder with a certain degree of inflammation, especially in the gangrenous form. They underline the risk of performing an anastomosis in these areas, since the inflammatory substrate could negatively impact on the sutures, mainly at colic level. The higher mortality rate of OSP vs EA reported by many could also be explained on this basis [7,48,50,60-62], though in recent years the mortality has decreased with both interventions, undoubtedly thanks to the more intensive preoperative preparation of the patient and the progress of anesthesiologic and resuscitation therapy [30-32,63,64].

The controversy between EA and OSP has not yet been solved; the rarity of gallstone ileus, and thus the difficulty in performing randomized controlled trials, is the main reason behind the still lacking standardization of the surgical approach to this entity. However, the bulk of data from the literature strongly suggests that the general condition of the patients at the time of the emergency surgery is the leading factor in the choice of the intervention. EA should be preferred in critical situations-multiple comorbidities as typically happens in the oldest patients, and, particularly, in the case of hemodynamic instability-when it is crucial to save the patient's life using the simplest and most rapid approach [7,19,38,48-51], while OSP should be chosen in selected cases with relatively stable general conditions [50,52,53]. With the progressive increase in life expectancy of the population, the age of the patient is a further criterion to be strongly taken into account. In conclusion, the critical evaluation of the available surgical approaches to gallstone ileus of the sigmoid colon here performed, suggests that, at present, enterolithotomy alone continues to provide good results for elderly patients in a critical general status, as in the case reported here.

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