Karewsky syndrome secondary to cholecystoduodenal fistula. A case report

Axell Daniel Lugo Rodríguez M.D.
José de Jesús Zapata Rosales M.D.
Isabel Híjar Cabello M.D.
Hugo Armando Gomez Rios M.D.
Maricelda Hortencia Trujillo Murillo M.D.
Christopher Giovanni Vega Flores M.D.

Coahuila, Mexico

Background: Karewsky syndrome is a rare and frequently overlooked clinical variant of gallstone ileus, characterized by recurrent episodes of intermittent or subacute mechanical bowel obstruction resulting from the transient impaction and spontaneous migration of a gallstone through the intestinal lumen. Because of its nonspecific and fluctuating clinical presentation, diagnosis is often delayed, increasing the risk of complete intestinal obstruction and other potentially life-threatening complications. Gallstone ileus accounts for only 1–4% of all cases of mechanical bowel obstruction but remains an important cause of surgical emergencies in elderly patients with long-standing gallstone disease.

Keywords: Karewsky syndrome, gallbladder.

Case Report

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Gallstone ileus is an uncommon but well-recognized complication of long-standing cholelithiasis, resulting from the migration of one or more gallstones into the gastrointestinal tract through a biliary-enteric fistula. It accounts for approximately 1–4% of all cases of mechanical bowel obstruction and up to 25% of non-strangulated small bowel obstruction in patients older than 65 years. Although uncommon, gallstone ileus is associated with considerable morbidity and mortality because it typically affects elderly patients with multiple comorbidities, and its diagnosis is often delayed due to nonspecific clinical manifestations.

The formation of a biliary-enteric fistula is usually preceded by repeated episodes of acute or chronic cholecystitis, which promote chronic inflammation, adhesion formation, pressure necrosis, and eventual erosion of the gallbladder wall into the adjacent gastrointestinal tract. Cholecystoduodenal fistulas account for approximately 70–90% of cases, allowing large gallstones to enter the intestinal lumen. Most calculi pass spontaneously; however, stones larger than 2–2.5 cm may become impacted, most commonly in the distal ileum because of its relatively narrow lumen and less active peristalsis, resulting in mechanical intestinal obstruction.

Karewsky syndrome represents an unusual clinical variant of gallstone ileus characterized by recurrent episodes of intermittent or subacute bowel obstruction caused by temporary impaction and spontaneous migration of the gallstone before definitive intestinal obstruction develops. These recurrent and self-limited obstructive episodes often lead to diagnostic uncertainty and delay definitive treatment. Because of its rarity and nonspecific presentation, preoperative diagnosis remains challenging despite advances in abdominal imaging. Contrast-enhanced computed tomography has become the diagnostic modality of choice because of its high sensitivity and specificity, allowing identification of Rigler's triad, localization of the ectopic gallstone, and detection of the associated biliary-enteric fistula.

Given the rarity of Karewsky syndrome, published reports remain limited, and awareness among clinicians is low. We present the case of a 55-year-old woman with recurrent episodes of acute cholecystitis and intermittent subacute intestinal obstruction who was diagnosed with Karewsky syndrome secondary to gallstone ileus caused by a cholecystoduodenal fistula. The patient was successfully treated with enterolithotomy alone, highlighting the importance of maintaining a high index of suspicion in patients with recurrent obstructive symptoms and a history of gallstone disease to facilitate early diagnosis and timely surgical management.

Case report

We report the case of a 55-year-old woman with a medical history of type 2 diabetes mellitus and recurrent symptomatic cholelithiasis, including at least five previous episodes of acute cholecystitis managed conservatively, who presented to the emergency department with a 3-day history of abdominal pain, nausea, vomiting, progressive abdominal distension, obstipation, and recurrent episodes of intermittent

From the Department of General Surgery at , Instituto Mexicano del Seguro Social, Unidad Médica de Alta Especialidad H.E. No. 71 Torreón, Coahuila. Received on July 7, 2026, Accepted on July 11, 2026. Published on July 14, 2026.

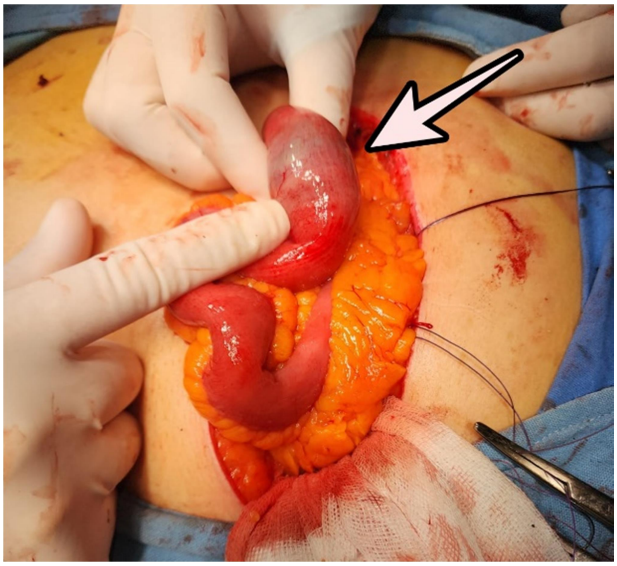

Figure 1. Gallstone impacted in the distal ileum, producing complete luminal obstruction

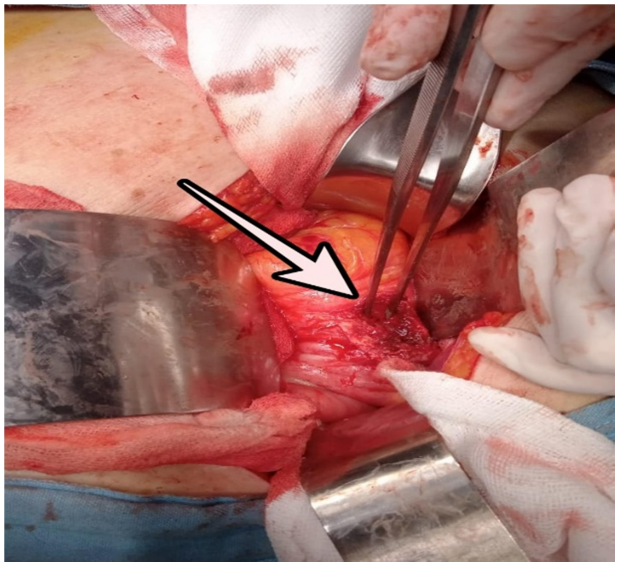

Figure 2. Intraoperative exploration demonstrated a cholecystoduodenal fistula

subacute intestinal obstruction. Physical examination revealed signs of mechanical bowel obstruction, including abdominal distension, diffuse tenderness on palpation, and decreased bowel sounds. Laboratory investigations demonstrated leukocytosis with neutrophilia, consistent with an underlying inflammatory process.

Contrast-enhanced computed tomography of the abdomen and pelvis revealed pneumobilia, dilated small bowel loops with air-fluid levels, and a 5-cm ectopic gallstone impacted within the distal ileum, findings consistent with gallstone ileus. Given the patient's history of recurrent transient obstructive episodes and chronic gallstone disease, Karewsky syndrome was strongly suspected.

The patient underwent emergency exploratory laparotomy, which confirmed a 5-cm gallstone impacted in the distal ileum, producing complete luminal obstruction(Figure 1). Intraoperative exploration also demonstrated a cholecystoduodenal fistula (Figure 2) without evidence of bowel ischemia or perforation. A simple enterolithotomy was successfully performed, allowing complete extraction of the obstructing gallstone and restoration of intestinal continuity. (Figure 3) Cholecystectomy and fistula repair were deferred because of the patient's clinical condition and the increased operative risk associated with a one-stage procedure.

The postoperative course was uneventful. Bowel function gradually returned, oral intake was initiated without complications, and the patient was discharged in good clinical condition on postoperative day 10. At outpatient follow-up, she remained asymptomatic, with no evidence of recurrent bowel obstruction or other postoperative complications.

Discussion

Gallstone ileus is an uncommon but potentially life-threatening complication of long-standing cholelithiasis resulting from the passage of one or more gallstones into the gastrointestinal tract through a spontaneous biliary-enteric fistula. Although it accounts for only 1–4% of all cases of mechanical bowel obstruction, its incidence increases considerably in elderly patients, representing up to 25% of non-strangulated small bowel obstruction in individuals older than 65 years (1–3). Delayed diagnosis remains one of the principal determinants of morbidity and mortality because the clinical presentation is often nonspecific and may mimic other causes of intestinal obstruction. Consequently, maintaining a high index of suspicion is essential, particularly in patients with recurrent biliary symptoms and intermittent obstructive episodes.

The development of gallstone ileus is closely related to recurrent inflammatory episodes involving the gallbladder. Chronic inflammation promotes adhesion formation between the gallbladder and adjacent bowel, followed by pressure necrosis and erosion of the gallbladder wall, ultimately resulting in the formation of a biliary-enteric fistula (2,4). Cholecystoduodenal fistulas account for approximately 70–90% of these communications and represent the most common route through which large gallstones enter the intestinal lumen (5). Stones measuring more than 2–2.5 cm are unlikely to pass spontaneously and frequently become impacted, most commonly within the distal ileum because of its relatively narrow lumen and reduced peristaltic activity (3,6). In our patient, the presence of a 5-cm gallstone impacted in the distal ileum, together with an intraoperatively confirmed cholecystoduodenal fistula,

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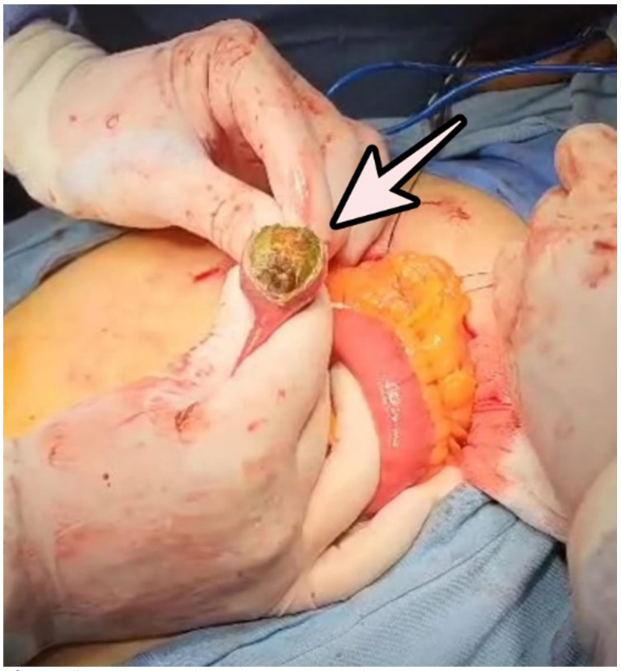

Figure 3. Intraoperative simple enterolithotomy allowing complete extraction of the obstructing gallstone

was entirely consistent with the pathophysiological mechanisms described in previous reports.

A distinctive feature of the present case is the diagnosis of Karewsky syndrome, an exceptionally rare clinical variant of gallstone ileus characterized by recurrent episodes of intermittent or subacute bowel obstruction preceding complete intestinal occlusion (7,8). These transient obstructive episodes result from the temporary impaction and spontaneous migration of the gallstone through the intestinal lumen, a phenomenon commonly referred to as the "tumbling phenomenon." Because symptoms frequently resolve spontaneously, patients often undergo repeated medical evaluations before definitive diagnosis is established, contributing to delays in surgical management (7). In retrospect, our patient's history of recurrent subacute bowel obstruction represented a key diagnostic clue and, when considered together with her previous episodes of acute cholecystitis, strongly supported the diagnosis of Karewsky syndrome.

Another noteworthy aspect of this case is the patient's age. Gallstone ileus predominantly affects elderly women, with a mean reported age between 70 and 80 years (1,2). In contrast, our patient was only 55 years old, demonstrating that this condition should not be excluded solely on the basis of age. Rather than chronological age alone, prolonged symptomatic cholelithiasis and recurrent inflammatory episodes appear to play a more decisive role in the development of biliary-enteric fistulas and subsequent gallstone migration. The history of at least five previous episodes of acute cholecystitis in our patient likely accelerated this process and emphasizes the importance of considering definitive cholecystectomy in patients with recurrent biliary disease to prevent severe complications such as gallstone ileus.

The diagnosis of gallstone ileus remains challenging because neither clinical manifestations nor laboratory findings are specific. Most patients present with abdominal pain, nausea, vomiting, abdominal distension, and obstipation, while laboratory abnormalities are generally limited to leukocytosis, neutrophilia, dehydration, or electrolyte disturbances associated with bowel obstruction (2,5). Accordingly, imaging studies play a central role in establishing the diagnosis. Although plain abdominal radiography has historically been used as the initial imaging modality, its diagnostic sensitivity is limited because the complete Rigler triad—pneumobilia, intestinal obstruction, and an ectopic gallstone—is identified in only a minority of patients (12). Consequently, reliance on conventional radiography alone may contribute to delayed diagnosis.

Contrast-enhanced computed tomography is currently considered the diagnostic modality of choice because it provides excellent visualization of the site of obstruction, accurately identifies the ectopic gallstone, demonstrates pneumobilia, evaluates bowel viability, and frequently detects the underlying biliary-enteric fistula (5,9,10,14). Reported diagnostic accuracy approaches 90–99%, making CT indispensable for both diagnosis and preoperative planning (10,13). In the present case, CT successfully demonstrated pneumobilia, dilated small bowel loops, and a large ectopic gallstone lodged within the distal ileum, allowing a confident preoperative diagnosis of gallstone ileus. More importantly, correlation of these imaging findings with the patient's history of recurrent intermittent bowel obstruction enabled recognition of Karewsky syndrome before surgery, a diagnosis that is rarely established preoperatively.

Recognition of this uncommon syndrome has important clinical implications. Patients with recurrent episodes of bowel obstruction are frequently diagnosed with postoperative adhesions, inflammatory bowel disease, gastroenteritis, or functional gastrointestinal disorders, particularly when symptoms improve spontaneously (7,8). Failure to recognize the characteristic intermittent course may delay definitive treatment until complete obstruction, bowel ischemia, or perforation develops. Therefore, Karewsky syndrome should be included in the differential diagnosis of patients with recurrent obstructive symptoms, especially when there is a documented history of gallstone disease or repeated episodes of cholecystitis. Our case reinforces the importance of obtaining a thorough clinical history and integrating it with cross-sectional imaging findings to facilitate early diagnosis and timely surgical intervention.

Once gallstone ileus is diagnosed, surgical management remains the definitive treatment, as

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spontaneous resolution is uncommon in cases of complete obstruction. However, the optimal operative strategy continues to be debated, particularly regarding whether treatment should be limited to relief of obstruction or combined with definitive management of the biliary pathology. Three main surgical approaches have been described: simple enterolithotomy, one-stage surgery (enterolithotomy combined with cholecystectomy and fistula repair), and a staged approach. The choice of technique should be individualized based on patient condition, comorbidities, degree of inflammation, and intraoperative findings (1–3).

Simple enterolithotomy is currently the most widely accepted approach and is considered the treatment of choice in the majority of patients. This procedure focuses exclusively on relieving the intestinal obstruction by removing the impacted gallstone, thereby minimizing operative time and physiological stress. This is particularly relevant because most patients with gallstone ileus are elderly and frequently present with multiple comorbidities. Several retrospective studies and meta-analyses have demonstrated that enterolithotomy alone is associated with lower perioperative morbidity and mortality compared with more extensive procedures, while still achieving satisfactory long-term outcomes (2,4,5). In the present case, although the patient was relatively younger than the typical population, the presence of acute obstruction and inflammatory adhesions justified a limited surgical approach.

In contrast, one-stage surgery aims to definitively treat both the intestinal obstruction and the underlying biliary disease by performing enterolithotomy, cholecystectomy, and fistula closure in a single operation. The theoretical advantage of this approach is the prevention of recurrent gallstone ileus, recurrent cholecystitis, cholangitis, and potential gallbladder carcinoma. However, this procedure is technically demanding due to dense inflammatory adhesions in the hepatoduodenal region and significantly increases operative time and complexity. Consequently, multiple studies have reported higher rates of postoperative complications, including bile duct injury, intra-abdominal abscess, biliary leakage, and cardiopulmonary complications, especially in high-risk patients (3,6,7).

The staged approach represents an intermediate strategy in which enterolithotomy is performed initially, followed by delayed cholecystectomy and fistula repair in selected patients. However, evidence suggests that spontaneous closure of biliary-enteric fistulas may occur in a significant proportion of cases after relief of obstruction, particularly cholecystoduodenal fistulas. Moreover, the risk of recurrent gallstone ileus after enterolithotomy alone remains relatively low, reported in most series between 2% and 8% (4,6,8). For this reason, routine delayed biliary surgery is no longer universally recommended and is generally reserved for patients with persistent biliary symptoms or recurrent complications.

The decision to perform simple enterolithotomy in our patient was based on both intraoperative and clinical considerations. Despite the presence of a cholecystoduodenal fistula, the surrounding inflammatory changes and adhesions made biliary dissection potentially hazardous. Attempting definitive fistula repair in this context would have significantly increased the risk of iatrogenic injury and prolonged operative time without clear short-term benefit. Given that the primary life-threatening issue was mechanical bowel obstruction, a focused procedure was considered the safest and most evidence-based option.

Postoperative outcomes in gallstone ileus depend largely on early diagnosis and timely surgical intervention. Delayed treatment has been associated with increased rates of bowel ischemia, perforation, sepsis, and mortality. In contrast, patients treated promptly with enterolithotomy generally experience favorable outcomes, with rapid recovery of bowel function and low recurrence rates (2,5,7). Our patient followed this expected clinical course, with uncomplicated recovery, early resumption of oral intake, and no evidence of recurrent obstruction during follow-up.

Minimally invasive surgery has also been increasingly explored in the management of gallstone ileus. Laparoscopic enterolithotomy has been described as a feasible alternative in selected cases, offering potential benefits such as reduced postoperative pain, shorter hospital stay, and faster recovery. However, its application remains limited due to technical challenges, including bowel distension, difficulty in handling inflamed tissues, and the complexity of intracorporeal enterotomy closure. Additionally, conversion rates remain significant, and open surgery continues to be the preferred approach in emergency settings, particularly when large impacted gallstones or unclear bowel viability are present (6,8,9,15).

Another important surgical principle emphasized in the literature is the need for complete inspection of the small bowel during surgery. Multiple synchronous gallstones may be present in up to 10–15% of cases, and failure to detect them may result in persistent or recurrent obstruction requiring reoperation. Therefore, systematic exploration from the ligament of Treitz to the ileocecal valve is recommended as part of the standard operative procedure in gallstone ileus (4,7,10).

From a clinical perspective, the present case reinforces several important considerations. First,

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Karewsky syndrome should be suspected in patients with recurrent episodes of subacute bowel obstruction, particularly when there is a background of chronic gallstone disease and repeated biliary inflammation. Second, computed tomography plays a central role not only in confirming gallstone ileus but also in suggesting the intermittent obstructive pattern characteristic of this syndrome. Third, early surgical intervention remains crucial to prevent progression to complete obstruction and its associated complications. Finally, simple enterolithotomy remains an effective and safe treatment strategy in most cases and should be considered the first-line surgical approach in appropriately selected patients.

Conclusion

Karewsky syndrome represents a rare and underrecognized clinical variant of gallstone ileus characterized by recurrent episodes of intermittent or subacute small bowel obstruction due to transient migration of a gallstone through a biliary-enteric fistula. Its nonspecific and fluctuating clinical presentation frequently delays diagnosis, making it a challenging entity in emergency surgical practice.

A high index of suspicion is essential in patients with a history of recurrent cholecystitis and repeated episodes of intestinal subobstruction, even in relatively younger individuals. Contrast-enhanced computed tomography remains the most valuable diagnostic tool, allowing accurate identification of pneumobilia, ectopic gallstones, and the level of obstruction, thus facilitating timely surgical planning. Surgical management should be individualized, but simple enterolithotomy remains a safe and effective first-line approach in most cases, particularly in the setting of acute obstruction and inflammatory changes, as it minimizes operative risk while achieving definitive relief of symptoms.

This case highlights the importance of early recognition of this rare syndrome and reinforces that prompt diagnosis and appropriate surgical intervention are key factors in achieving favorable outcomes.

Conflicts of interests

The authors declare no conflicts of interests.

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Axell Daniel Lugo Rodríguez
Department of General Surgery
Instituto Mexicano del Seguro Social
Unidad Médica de Alta Especialidad H.E. No. 71
Torreón, Coahuila

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