

## CASE REPORT

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# Biliary dyskinesia an undiagnosed cause of abdominal pain: Case report

Victor R Medina, Jhony A Delgado, Alexandra S Valencia,  
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## ABSTRACT

**Introduction:** Biliary dyskinesia is a functional gallbladder disorder of rare presentation, the diagnostic suspicion is very important, the standard confirmatory test is a hepatobiliary iminodiacetic acid (HIDA) scan (scintigraphy) and standard treatment is cholecystectomy. **Case Report:** A 37-year-old female who had a surgical record of appendectomy, two caesarean sections, salpingoclastm, and cystectomy for ovarian cyst presented a 48-hour episodic postprandial abdominal pain that reached 10/10 intensity on visual analoguescale(VAS)scale. The pain was located in the right hypochondrium and dorsal region, it was accompanied by nausea. On physical examination, patient presented positive Murphy's sign. Furthermore, laboratory tests showed no leukocytosis nor neutrophilia. In addition, pancreatic enzymes, kidney, and liver function ranged within normal. Gallbladder presented normal volume as measured by ultrasound. In addition, thin walls without endoluminal lesions were found, intra- and extrahepatic bile ducts without dilation were detected. Due to the persistence of pain and the lack of association with lithiasis etiology, a HIDA scan was performed, which reported normal filling of the gallbladder, with a 27.22% ejection fraction, whereby a diagnosis of biliary

dyskinesia was determined. **Conclusion:** Laparoscopic cholecystectomy was performed, finding a gallbladder with signs of inflammation according to the pathology report. Afterward, the patient was discharged without complications. The diagnosis is made by exclusion of other pathologies and it is confirmed by a HIDA scan. Cholecystectomy is proven to be the treatment of choice, also aiding the diagnosis of biliary dyskinesia with relief of symptoms in most patients.

**Keywords:** Biliary dyskinesia, Ejection fraction, HIDA (hepatobiliary iminodiacetic acid), Laparoscopic cholecystectomy

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## INTRODUCTION

Biliary dyskinesia is defined as a functional/motile disorder of the gallbladder or Oddi sphincter with multiple potential underlying causes [1]. Additionally, it can be related to other disorders, such as irritable bowel syndrome, gastroesophageal reflux disease, chronic constipation, colonic inertia, and gastroparesis [2]. Functional disorder of the gallbladder or Oddi sphincter has increased the cholecystectomy rate from 5% to 25%, although its prevalence is unknown [1]. The diagnosis has evolved based on the Rome criteria, whose last transition has been from the Rome III to IV criteria [3]. A HIDA scan showing an ejection fraction of less than 40% of the

gallbladder confirms the diagnosis. Although it is a rare entity and the diagnosis is made by exclusion, several studies indicate that it can be successfully treated with cholecystectomy.

In absence of the gallbladder, biliary dyskinesia is more difficult to treat and it requires a specialized center with the ability to perform an Oddi sphincter manometry. The success of any invasive treatment for biliary dyskinesia is defined as the absence of biliary pain for more than 12 months after the intervention [2].

## CASE REPORT

The patient is a 37-year-old female who works as a merchandizer. She is Catholic, her blood group is A positive, and is allergic to metoclopramide. Moreover, she does not have clinical record. However, she has a surgical history of appendectomy 17 years ago. Furthermore, she had two caesarean sections, 14 and 10 years ago, salpingooclast 7 years ago, and cystectomy for hemorrhagic ovarian cyst 4 years ago. The patient presented a 48-hour clinical onset characterized by abdominal pain located in the right hypochondrium and radiating to the dorsal region that corresponds to 10/10 intensity on VAS scale. Pain was accompanied by nausea that did not lead to vomiting. Apparently, it could be attributed to fatty food ingestion. Then, the patient received analgesia. However, the pain persisted in a 6/10 intensity on VAS scale.

On physical examination, vital signs were stable, she was in pain; scars due to abdominal surgery were seen, hydro-air sounds presented normal frequency and intensity. By palpation, the abdomen was found soft and depressible. In addition, it was painful in the right hypochondrium, with a positive Murphy's sign and no signs of peritoneal irritation.

In laboratory tests, neither leukocytosis (8210 cells/mm<sup>3</sup>) nor neutrophilia (62.6%) was detected; pancreatic enzymes (amylase 55 U/L, lipase 60 U/L), kidney (creatinine 0.60 mg/dL), and liver function (total bilirubin 1.35 mg/dL, direct bilirubin 0.36 mg/dL, alkaline phosphatase (ALP) 96 U/L, and gamma-glutamyl transpeptidase (GGT) 67 U/L) ranged within normal. The ultrasound showed a thin-walled gallbladder, its volume was normal, it did not have endoluminal lesions, intra- and extrahepatic bile ducts were not dilated and the common hepatic duct measured 4 mm (Figure 1).

Due to persistence of pain without biliary pathology identified by ultrasound, biliary dyskinesia was suspected and a HIDA scan was requested, which reported normal filling of the gallbladder during 20 minutes, reaching maximum filling at 60 minutes. In fact, it showed significant retention of the radiolabeling on subsequent images. Furthermore, the ejection fraction corresponded to 27.22% (Figure 2). Therefore, she was diagnosed with biliary dyskinesia, thereby a laparoscopic cholecystectomy was performed. Then, a thin-walled tense gallbladder was found, it measured 7 × 4 × 3 cm, with clear bile, without

gallstones and not dilated bile duct. Consequently, it was an uncomplicated procedure. During the hospital stay, intravenous hydration with ringer lactate solution, analgesia based on ketorolac and acetaminophen, and gastric protector with omeprazole were intravenously administered.

At 48-hour post-operation phase, the patient was hemodynamically stable, with adequate pain management and diet tolerance, without signs of systemic inflammatory response, and she was discharged for post-surgical control in an outpatient clinic. The pathology report showed a gallbladder with signs of inflammation (Figure 3).

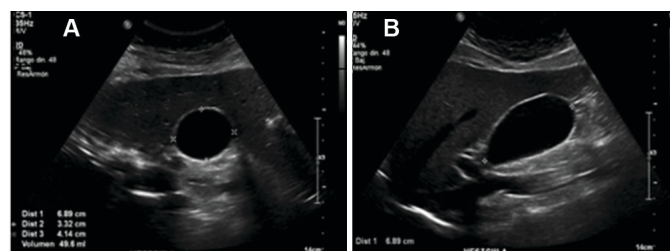


Figure 1: (A) Transversal vision. (B) Longitudinal view of gallbladder with thin walls and without pathology inside it.

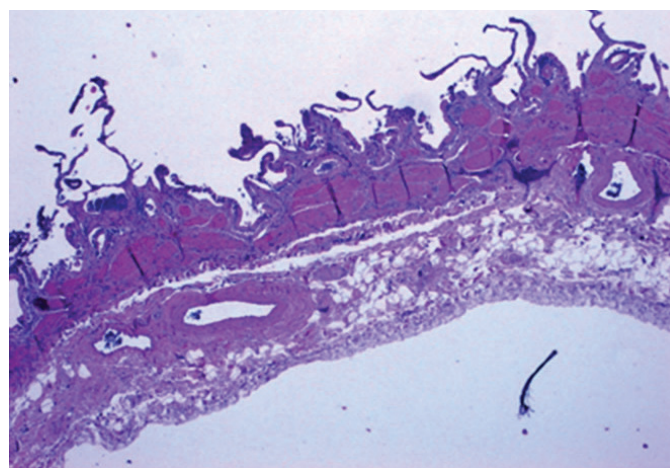


Figure 2: Histopathology: Cholecystitis with epithelial hyperplasia, fibrosis, and muscular hypertrophy (staining with hematoxylin & eosin, 10×).

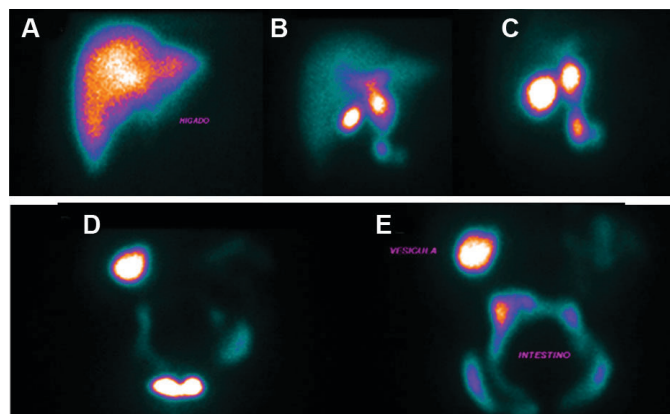


Figure 3: HIDA scan, static images. (A) 10 minutes, (B) 30 minutes, (C) 60 minutes, (D) 2 hours 30 minutes, and (E) 4 hours 30 minutes with an ejection fraction of 27.22%.

## DISCUSSION

Biliary dyskinesia is a functional gastrointestinal disorder that can cause significant abdominal pain. In addition, functional biliary disorders include gallbladder pain without gallstones, biliary dysmotility, sphincter of Oddi dysfunction, ampullary stenosis, and postcholecystectomy syndrome [2].

Biliary dyskinesia should be considered a diagnosis by exclusion and it is a rare disorder [1]. The incidence outside the United States is 25 cases per million compared to 85 cases per million in this country, particularly in the south east. In the United States, 20% of cholecystectomies in adults and 60% in pediatrics are performed for a functional gallbladder disorder [2, 4]. However, there is no consensus on diagnostic criteria for biliary dyskinesia in children since the symptoms overlap with those of functional dyspepsia [5].

Rome IV criteria for gallbladder functional disorder must include the following:

1. Biliary pain.
2. Absence of vesicular stones or other structural pathologies.

Support criteria

1. Low ejection fraction in gallbladder scintigraphy.
2. Normal liver enzymes, conjugated bilirubin, amylase, and lipase.

As a rare entity, the clinical onset is characterized by pain in the upper right quadrant, generally 30 minutes after food intake. Sometimes, it presents with nausea, vomiting, bloating and diarrhea. Nevertheless, some are also asymptomatic [1]. Differential diagnosis should be taken into account (Table 1), laboratory and imaging tests should focus on excluding the most common diagnosis, such as malignancy, gallbladder microlithiasis, choledocholithiasis, peptic ulcer disease, chronic pancreatitis, and musculoskeletal syndromes [2].

Indeed, laboratory tests should include liver and pancreatic function (serum aspartate aminotransferase, alanine aminotransferase, bilirubin, amylase, and lipase) [2].

Table 1: Differential diagnosis of pain in the upper right quadrant or of biliary dyskinesia

### Syndromes that simulate biliary pain: lithiasis cholecystitis/cholangitis/choledocholithiasis/tumorous biliary pathology

Irritable bowel syndrome

Peptic ulcer disease

Chronic constipation

Gastroesophageal reflux disease

Cirrhosis/terminal liver disease

Coronary artery disease

Costochondritis/musculoskeletal disorder

Differential diagnosis (pain in the upper right quadrant)

Moreover, abdominal ultrasound should be the first diagnostic test for biliary pain as computed tomography required to assess nonspecific acute abdominal pain does not adequately assess the gallbladder nor bile duct. Finally, magnetic resonance imaging must be requested in cases of biliary lithiasis suspicion or tumorous obstruction [2]. Hepatobiliary iminodiacetic acid scan is very useful, indirectly evaluating the contraction capacity of the gallbladder when stimulated with intravenous cholecystokinin or a fatty food. The results are expressed in the percentage of radiolabeling excreted by the gallbladder, being less than 40% of the excretion a positive result of biliary dyskinesia [6].

## Non-surgical treatment

This modality has not been widely studied; although, there are observational studies that indicate that symptoms can decrease over time and that invasive procedures are not the treatment of biliary dyskinesia due to its functional component in the Oddi sphincter [7]. Calcium channel blockers such as nifedipine have been evaluated with some potential improvement in pain, but with headache and tachycardia as side effects [8].

## Cholecystectomy

Cholecystectomy is considered the standard treatment for biliary dyskinesia which has had an increase of this pathology resolution in the United States [6]. In absence of the gallbladder, invasive procedures such as endoscopic retrograde cholangiopancreatography with sphincterotomy or injection of botulinum toxin in the Oddi sphincter are more controversial.

Ponsky et al. identified five studies including 274 patients, 98% of patients had symptomatic relief with surgery [9]. Mahid et al. identified 10 studies including 462 patients, no stones on ultrasound examination and a lower gallbladder ejection fraction were seen. Then, these patients benefited from cholecystectomy versus observation [10].

Cholecystectomy studies are very promising with a cure rate between 80% and 100%. However, they can leave up to 20% of patients with persistent biliary pain due to a concomitant functional disorder in the Oddi sphincter [4, 11].

Patients with normokinesis and biliary dyskinesia diagnosed with HIDA scan are clearly benefited by cholecystectomy [10]. In pediatric patients, symptom relief after cholecystectomy for biliary dyskinesia is inconsistent [12].

In Ecuador, it constitutes one of the very rare or underdiagnosed pathologies. Therefore, prevalence is not reported. The young female patient presented could be associated with the description of a high intensity biliary pain, which interrupts daily activities and forces to seek medical evaluation at an emergency room, meeting one of the criteria of Rome IV for dyskinesia vesicular.

Regarding laboratory tests, leukocytosis and neutrophilia were not evident in this patient, and the other tests remained normal, which could be related to two more Rome IV criteria for biliary dyskinesia.

Finally, in the HIDA scan, the patient showed an ejection fraction of 27.22%, which is related to the last of the Rome IV criteria for biliary dyskinesia.

Laparoscopic cholecystectomy was considered the standard treatment for biliary dyskinesia in this patient, and it was performed without complication. After the procedure, the patient had good pain control, diet tolerance, and had no signs of systemic inflammatory response. Consequently, she was discharged in a good condition after 48 hours of hospitalization.

## CONCLUSION

Biliary dyskinesia is defined as a functional or motility disorder of gallbladder or the Oddi sphincter. It is an uncommon etiology of abdominal pain which needs to be considered in the evaluation of abdomen pain when investigations are normal. Therefore, Rome IV criteria for functional gallbladder disorder should include biliary pain and absence of gallstones or other structural pathologies. Furthermore, as support criteria, low ejection fraction in gallbladder scintigraphy and normal liver enzymes, conjugated bilirubin, amylase, and lipase. A decrease in the gallbladder ejection fraction (<40%) is considered an objective measurement of functional gallbladder disorder obtained by HIDA. Finally, cholecystectomy is considered the standard treatment for biliary dyskinesia.

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## Author Contributions

Victor R Medina – Conception of the work, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Jhony A Delgado – Conception of the work, Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Alexandra S Valencia – Conception of the work, Acquisition of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Diego A Díaz – Design of the work, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Stalin R Ordoñez – Conception of the work, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Paola A Palacios – Conception of the work, Analysis of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the

version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

**Guarantor of Submission**

The corresponding author is the guarantor of submission.

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**Consent Statement**

Written informed consent was obtained from the patient for publication of this article.

**Conflict of Interest**

Authors declare no conflict of interest.

**Data Availability**

All relevant data are within the paper and its Supporting Information files.

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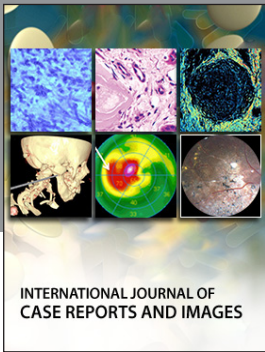
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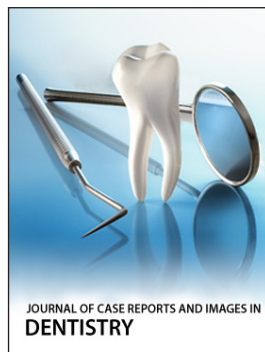
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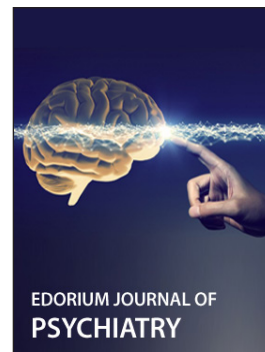
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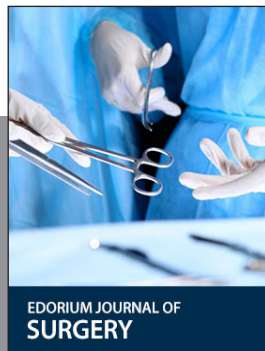
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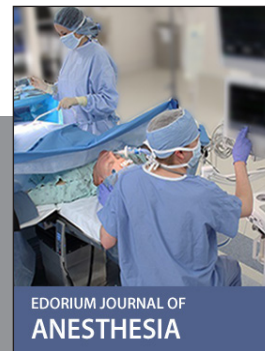
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