

Hyperkinetic Biliary Dyskinesia in Adults: Cholecystectomy as a Treatment Option

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Background	Two female Caucasian patients, 69 years old and 41 years old, presented with six and twelve-month histories, respectively, of postprandial right upper quadrant and epigastric pain and nausea.
Summary	Two female Caucasian patients, 69 years old and 41 years old, presented with a several month history of postprandial right upper quadrant and epigastric pain and nausea. On abdominal ultrasound, neither patient had evidence of biliary pathology, including cholelithiasis or choledocholithiasis. Their liver function laboratory tests were normal. On further workup of their abdominal pain, they each underwent a hepatobiliary iminodiacetic acid (HIDA) scan to determine if there was evidence of biliary dyskinesia. Both patients were found to have elevated gallbladder ejection fractions (GBEF) of 97 percent and 98 percent, respectively, on HIDA scan. Both patients underwent elective minimally invasive cholecystectomies without complication. On final pathology, neither patient had evidence of gallbladder abnormalities. Both patients were followed for six months postoperatively and reported significant improvement of symptoms clinically. Although cholecystectomy has been shown to improve symptoms of biliary colic in patients with hypokinetic biliary dyskinesia (GBEF \leq 35 percent), there are no reports of cholecystectomy as a treatment for hyperkinetic biliary dyskinesia (GBEF \geq 80 percent) in adults. The resolution of symptoms of post prandial right upper quadrant and epigastric pain in these patients highlights the need for further investigation of hyperkinetic biliary dyskinesia as a potential cause acalculous biliary colic.
Conclusion	Acalculous biliary colic has been associated with several functional disorders including sphincter of Oddi dysfunction and hypokinetic biliary dyskinesia. We present two cases of resolved symptomatic biliary colic in patients with hyperkinetic biliary dyskinesia. Though not currently the standard of care, these cases highlight the need for continued investigation of hyperkinetic biliary dyskinesia with elevated GBEF as a potential indication for elective cholecystectomy.
Keywords	Hyperkinetic biliary dyskinesia, acalculous biliary colic

DISCLOSURE:

Dr. Higgins has disclosed the following conflicts of interest:

- Speaker for W.L. Gore & Associates
- Proctor for Intuitive Surgical

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Case Description

Cholelithiasis and biliary dyskinesia are the two main causes of biliary colic. Biliary dyskinesia is typically hypokinetic, defined as a gallbladder ejection fraction (GBEF) ≤ 35 percent on hepatobiliary iminodiacetic acid (HIDA) scan. In adults with hypokinetic biliary dyskinesia, cholecystectomy has been shown to improve biliary colic symptoms.¹ Typically hypokinesia develops in middle age females due to stress and autonomic dysfunction that interferes with contraction of the gallbladder, causing delayed emptying and contraction. Hyperkinetic biliary dyskinesia with a GBEF ≥ 80 percent is less recognized as a source of biliary colic.² There is literature that supports the benefit of cholecystectomy for hyperkinetic GBEF in children and adolescents, but not adults. We hypothesized that a similar mechanism could be contributing to hyperkinetic gallbladder by causing upregulation and increased sensitization to autonomic signals causing gallbladder hyper-contraction. Due to the lack of literature regarding hyperkinetic biliary dyskinesia as a possible etiology of acalculous biliary colic, cholecystectomy is not currently the standard of care. We aim to highlight two adult patients with hyperkinetic biliary dyskinesia and their symptom improvement after cholecystectomy. Both patients were followed for six months postoperatively.

The first patient is a 69-year-old Caucasian female with a six-month history of post-prandial right upper quadrant and epigastric pain and nausea. There was no evidence of cholelithiasis on abdominal ultrasound and HIDA scan showed an elevated GBEF of 97 percent. Due to the lack of literature supporting cholecystectomy for hyperkinetic biliary colic in adults, there was a discussion with the patient that her symptoms may not benefit from surgery. The patient wanted to proceed and underwent an uncomplicated laparoscopic cholecystectomy. The surgical pathology report showed no gallbladder abnormality. At two weeks and two months, the patient reported complete resolution of her preoperative symptoms.

The second patient is a 41-year-old Caucasian female with a one-year history of post-prandial right upper quadrant abdominal pain and nausea. She had a history of pancreatitis of unknown etiology that had resolved. An abdominal ultrasound demonstrated no evidence of cholelithiasis and a HIDA scan showed an elevated GBEF of 98 percent. The patient underwent an uncomplicated robotic-assisted cholecystectomy. The surgical pathology showed no gallbladder abnormality. The patient reported resolution of her symptoms post-operatively.

Discussion

The evaluation for biliary colic begins with a transabdominal ultrasound to evaluate for gallstones, gallbladder wall thickening, and biliary sludge. If these studies are negative, as seen in our patients, cholelithiasis can be ruled out as the etiology of the pain. Additional etiologies of right upper quadrant and epigastric abdominal pain are then ruled out (ischemic heart disease and peptic ulcer disease). Acalculous biliary disease can be further investigated by evaluating GBEF via HIDA scan. Functional gallbladder dysfunction has been associated with biliary-type pain. Patients with hypokinetic GBEF ≤ 35 percent have been shown to benefit symptomatically from cholecystectomy.¹ However, in patients with normal or hyperkinetic GBEFs, the etiology for acalculous biliary pain is less clear. Sphincter of Oddi dysfunction is another reported functional cause of acalculous biliary pain with a normal GBEF. In several studies, endoscopic retrograde cholangiopancreatography (ERCP) with sphincterotomy was evaluated as a treatment for patients with elevated sphincter pressure. Although ERCP with sphincterotomy can alleviate pain, the cohort of patients in these studies included those with refractory pain after cholecystectomy.³ These findings suggest that sphincter of Oddi dysfunction should also be investigated in patients with persistent biliary-type pain that is refractory to cholecystectomy.

The role of hyperkinetic GBEF and its contribution to acalculous biliary colic in adults is unclear. In the pediatric literature, there is evidence to support cholecystectomy for hyperkinetic GBEF. A retrospective review of 12 patients under 18 years old who underwent cholecystectomy for hyperkinetic GBEF had cholecystitis on final pathology. Hyperkinetic GBEF was defined as ≥ 80 percent. The patients were contacted post-operatively, with a mean follow-up of 16 months, and had relief of their preoperative biliary colic symptoms.⁴ In another retrospective study, 310 patients under 21 years old who underwent a HIDA scan were analyzed. GBEF, whether it be hypokinetic or hyperkinetic, was not found to correlate with microscopic gallbladder pathology.² Therefore, hyperkinetic biliary dyskinesia has been considered as an indication for surgery in pediatric acalculous biliary colic patients. This treatment paradigm has not yet been discussed in the adult literature.

Conclusion

This case report identifies two adult patients who demonstrated symptomatic improvement after cholecystectomy in the setting of hyperkinetic GBEF. As previously identified in pediatric patients, this warrants further investigation of hyperkinetic biliary dyskinesia as a source of acalculous biliary colic in adults.

Lessons Learned

Hyperkinetic biliary dyskinesia, defined as a GBEF ≥ 80 percent, may be associated with symptoms of biliary colic in adult patients. Cholecystectomy can be considered as treatment option for these patients, although definitive improvement in symptoms is still uncertain.

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