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Biliary dyskinesia: a potentially unrecognized cause of abdominal pain in children

Received: 1 August 2003 / Accepted: 27 February 2004 / Published online: 18 August 2004
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Abstract Biliary dyskinesia is defined as symptomatic biliary colic without cholelithiasis, and is diagnosed during cholescintigraphy by assessing gallbladder emptying with cholecystokinin (CCK) stimulation. Unfortunately, gallbladder emptying is not routinely assessed during cholescintigraphy in pediatric patients. The purpose of this review is to assess the effectiveness of cholecystectomy in patients with chronic abdominal pain and delayed gallbladder emptying and to assess whether these findings correlate with the histologic evidence of chronic cholecystitis. We retrospectively reviewed the medical records of all patients ($n=16$) at our institution from October 1997 to August 2001 who underwent quantitative cholescintigraphy with CCK stimulation that demonstrated delayed gallbladder emptying ($<35\%$ at 60 min) and who subsequently underwent cholecystectomy. Laparoscopic cholecystectomy was performed in 16 patients with chronic abdominal pain. All 16 patients had delayed gallbladder emptying (mean ejection fraction: $15 \pm 8\%$, range: 3–32%). The mean age was 12 ± 2 years (range: 8–17 years). Presenting symptoms included abdominal pain (86%), fatty food intolerance (27%), emesis (13%), and diarrhea (13%). Mean duration of abdominal pain before operation was 11 ± 19 months (range: 2 weeks–6 years). One patient's symptoms persisted postoperatively, but abdominal pain resolved in all other patients. Histologic evidence of chronic cholecystitis was demonstrated in 86% of surgical specimens. Five patients underwent concurrent appendectomy, and all had normal appendiceal histol-

ogy. Our experience suggests that children with chronic abdominal pain and delayed gallbladder emptying on CCK-stimulated cholescintigraphy are likely to benefit from cholecystectomy and to have histologic evidence of chronic cholecystitis.

Keywords Biliary dyskinesia · Laparoscopic cholecystectomy · Cholescintigraphy · Abdominal pain

Introduction

Biliary dyskinesia is characterized by symptomatic biliary colic without evidence of cholelithiasis and is diagnosed by assessing gallbladder emptying with cholecystokinin (CCK) stimulation during cholescintigraphy. Although this clinical entity has been recognized in adults for more than 2 decades, it has only recently been identified in children [1–4]. For this reason, many children with chronic abdominal pain caused by biliary dyskinesia may be inappropriately diagnosed and managed. The purpose of this review is to assess the effectiveness of cholecystectomy as treatment for patients with chronic abdominal pain and delayed gallbladder emptying and to assess whether these findings correlate with the histologic evidence of chronic cholecystitis.

Methods

We retrospectively reviewed the medical records of all patients at our institution from October 1997 to August 2001 who underwent quantitative cholescintigraphy with CCK stimulation demonstrating delayed gallbladder emptying ($<35\%$ at 60 min) and who subsequently underwent cholecystectomy, and identified 16 patients ($n=16$). Seven patients underwent concurrent procedures: five appendectomies, one intraoperative cholangiogram, and one central venous line placement. During the study period, all children who underwent CCK-

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Table 1 Characteristics of patients undergoing lap cholecystectomy for biliary dyskinesia ($n = 16$)

	n (%)
Age (mean)	12 ± 2 years
Gender	
Male	5 (31)
Female	11 (69)
Symptoms	
Abdominal pain	14 (86)
Fatty food intolerance	4 (27)
Emesis	2 (13)
Diarrhea	2 (13)
Duration of symptoms (mos)	11 ± 19
Mean GB ejection fraction (%)	15 ± 8
Complications	0
Resolution of symptoms	15 (94)
Chronic cholecystitis	12 (86)

stimulated cholescintigraphy at our institution had their gallbladder ejection fraction measured using a standard technique that is described in detail elsewhere [5]. Data regarding symptom resolution postoperatively was obtained from clinic notes. This study was approved by the Institutional Research Review Committee at Sparrow Health System (Lansing, Michigan, USA) and the Institutional Review Board at Hurley Medical Center (Flint, Michigan, USA).

Results

Laparoscopic cholecystectomy was performed in 16 patients with chronic abdominal pain who had preoperative cholescintigraphy. Each of the 16 patients demonstrated delayed gallbladder emptying (Table 1; mean ejection fraction: $15 \pm 8\%$, range: 3–32%). The mean age was 12 ± 2 years (range: 8–17 years). There were 11 girls (69%) and five boys (31%). Presenting symptoms included abdominal pain (86%), fatty food intolerance (27%), emesis (13%), and diarrhea (13%). Mean duration of abdominal pain before operation was 11 ± 19 months (range: 2 weeks–6 years). There were no conversions to open operation and no intraoperative or postoperative complications. One patient's symptoms persisted postoperatively, but abdominal pain resolved in all other patients. Histologic evidence of chronic cholecystitis was demonstrated in 86% of surgical specimens. All five patients who underwent concurrent appendectomy had normal appendiceal histology.

Table 2 Literature review of pediatric biliary dyskinesia case series

Study	No. of patients	Mean age (years)	Duration of symptoms (mos)	Symptom resolution (%)	Histologic chronic cholecystitis (%)
Tsakayannis et al. [2]	12	13	–	100	100
Gollin et al. [3]	29	13	11	79	72
Michail et al. [4]	63	12	14	72	80
Campbell et al. (current study)	16	12	11	94	86

Discussion

We have demonstrated that most children with chronic abdominal pain and no evidence of cholelithiasis who have delayed gallbladder emptying on CCK-stimulated cholescintigraphy have resolution of symptoms following laparoscopic cholecystectomy. We have also shown a correlation between these findings and histologic evidence of chronic cholecystitis.

Biliary dyskinesia has been a recognized clinical entity in adults for more than 2 decades, and laparoscopic cholecystectomy is now believed by many authors to be a highly effective treatment for this condition [6–10]. Two recent reviews of biliary tract disease in children and indications for pediatric cholecystectomy, however, fail to mention biliary dyskinesia [11–12]. Biliary dyskinesia (also called chronic acalculous cholecystitis) has only recently received attention in the pediatric surgical literature [2–4].

Our findings corroborate those of the three previously published case series on this subject in children. The patients with biliary dyskinesia described in these studies were between 5 and 18 years old, and the condition appears more commonly in females. The non-specific nature of the children's complaints described in these studies coupled with the absence of abnormalities on routine laboratory and radiologic tests usually delays diagnosis for nearly a year, and often for significantly longer.

For the five children who underwent concurrent appendectomy, it is possible that the appendectomy rather than the cholecystectomy, despite normal appendiceal histology, was responsible for the cessation of their symptoms. In the one patient who did not experience symptom relief following cholecystectomy, limited follow-up provided no alternative explanation for the etiology of the abdominal pain.

The small sample size of this study, along with the fact that it is a retrospective case series without a comparison group, limits the quality of the evidence that it provides [13]. Data provided by case series like this one and those cited in this study could be improved with more detailed and thorough postoperative follow-up. Future studies in which children with persistent abdominal pain of unclear etiology are identified early and evaluated prospectively will provide valuable insight into the natural history of biliary dyskinesia. Promising

computer models for improving the diagnostic accuracy of less common causes of abdominal pain in children are being developed [14].

Biliary dyskinesia is a rare cause of persistent abdominal pain in children and a diagnosis of exclusion. When routine laboratory and radiologic tests are inconclusive, CCK-stimulated cholescintigraphy should be considered. Children whose studies demonstrate abnormal gallbladder emptying should then be considered candidates for laparoscopic cholecystectomy.

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