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JOURNAL OF PEDIATRIC SURGERY, PHILADELPHIA, v. 47, n. 11, supl. 1, Part 4, pp. 2033-2038, NOV, 2012
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Management of gallstone disease in children: a new protocol based on the experience of a single center

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Received 25 February 2012; revised 16 June 2012; accepted 19 June 2012

Key words: Cholelithiasis; Choledocholithiasis; Laparoscopic cholecystectomy; Sickle-cell anemia; Cholangiography; Endoscopic retrograde cholangiopancreatography

Abstract

Background/purpose: Gallstones and cholelithiasis are being increasingly diagnosed in children owing to the widespread use of ultrasonography. The treatment of choice is cholecystectomy, and routine intraoperative cholangiography is recommended to explore the common bile duct. The objectives of this study were to describe our experience with the management of gallstone disease in childhood over the last 18 years and to propose an algorithm to guide the approach to cholelithiasis in children based on clinical and ultrasonographic findings.

Methods: The data for this study were obtained by reviewing the records of all patients with gallstone disease treated between January 1994 and October 2011. The patients were divided into the following 5 groups based on their symptoms: group 1, asymptomatic; group 2, nonbiliary obstructive symptoms; group 3, acute cholecystitis symptoms; group 4, a history of biliary obstructive symptoms that were completely resolved by the time of surgery; and group 5, ongoing biliary obstructive symptoms. Patients were treated according to an algorithm based on their clinical, ultrasonographic, and endoscopic retrograde cholangiopancreatography (ERCP) findings.

Results: A total of 223 patients were diagnosed with cholelithiasis, and comorbidities were present in 177 patients (79.3%). The most common comorbidities were hemolytic disorders in 139 patients (62.3%) and previous bariatric surgery in 16 (7.1%). Although symptoms were present in 134 patients (60.0%), cholecystectomy was performed for all patients with cholelithiasis, even if they were asymptomatic; the surgery was laparoscopic in 204 patients and open in 19. Fifty-six patients (25.1%) presented with complications as the first sign of cholelithiasis (eg, pancreatitis, choledocolithiasis, or acute calculous cholecystitis). Intraoperative cholangiography was indicated in 15 children, and it was positive in only 1 (0.4%) for whom ERCP was necessary to extract the stone after a laparoscopic cholecystectomy (LC). Preoperative ERCP was performed in 11 patients to extract the stones, and a hepaticojejunostomy was indicated in 2 patients. There were no injuries to the hepatic artery or common bile duct in our series.

Conclusions: Based on our experience, we can propose an algorithm to guide the approach to cholelithiasis in the pediatric population. The final conclusion is that LC results in limited postoperative
Although gallstones have traditionally been considered to be much less common in children than in adults, gallstone disease has increasingly been diagnosed in the pediatric population, mainly owing to the widespread use of ultrasonography [1].

In the past, gallbladder disease in children was usually observed primarily in patients with comorbid conditions such as hemolytic disorders, parenteral nutrition dependence, or cystic fibrosis [2,3]. However, the cholecystectomy rate in children without the diagnosis of hemolytic anemia has doubled in United States in recent years [4]. Obesity is a known risk factor for gallbladder disease, and the increase in the incidence of pediatric gallbladder disease parallels the rise in childhood obesity [5].

The history of gallstones in adults has shown that the majority (>80.0%) are incidentally detected as asymptomatic gallstones, and indeed, the majority (>80.0%) remain asymptomatic on long-term follow-up. However, if adult patients develop complications (eg, pancreatitis, acute cholecystitis, or choledocolithiasis), these complications are usually preceded by biliary colic pain [6]. In the pediatric population, the picture is not so clear; hence, the treatment remains controversial.

Another point of discussion is that, in the past, routine intraoperative cholangiography (IOC) was recommended during a cholecystectomy because a common bile duct (CBD) exploration was indicated. This situation, however, is different in the era of laparoscopic cholecystectomy (LC) and even more because of pediatric endoscopic retrograde cholangiopancreatography (ERCP). Laparoscopic IOC and CBD exploration, although feasible, are not easy to perform, require expertise, and may be time consuming [7-9]. Furthermore, this procedure may lead to the conversion of an LC into an open and unnecessary CBD exploration, considering the 20.0% to 25.0% false-positive rate of IOC [10] and the possibility of a stone migrating spontaneously through the papilla and into the duodenum.

The objective of this study was to describe our experience with the management of gallstone disease of childhood over the last 18 years, with particular attention given to the associated comorbidities, clinical presentation, postoperative outcomes, surgical techniques, and our experience with IOC. In addition, we propose an algorithm to guide the approach to cholelithiasis in children based on clinical and ultrasonographic findings.

1. Patients and methods

The data for this study were obtained by reviewing the records of all patients with gallstone disease treated between January 1994 and October 2011 at the Pediatric Surgery Division of the Child Institute of the University of Sao Paulo School of Medicine. This retrospective study protocol was approved by the ethical committee of the institution.
The information collected for each patient included age, sex, associated diseases, presenting symptoms, and symptom duration. Based on the symptoms, we divided the patients into the following 5 groups:

- Group 1: asymptomatic;
- Group 2: nonbiliary obstructive symptoms (eg, recurrent pain, abdominal discomfort, nausea related to fatty meals);
- Group 3: acute cholecystitis symptoms (eg, fever, vomiting, leukocytosis, acute right upper quadrant pain);
- Group 4: a history of biliary obstructive symptoms that were completely resolved at the time of surgery;
- Group 5: ongoing biliary obstructive symptoms (eg, jaundice, acholia, choluria, acute pancreatitis).

Ultrasonographic data included number of gallstones and presence or absence of choledocolithiasis. Based on their clinical, ultrasonographic, and ERCP data, patients were treated following the algorithm in Fig. 1.

The technique used for LC was similar to that described in the literature. Four 10- or 5-mm ports were used in combination with a 30° angled laparoscope at the umbilicus. The abdominal cavity was insufflated with carbon dioxide to a pressure of 10 to 12 mm Hg. A Harmonic Scalpel (Ethicon, Cincinnati, OH) or an electrocautery hook was used for dissection. One technical detail of note is that all of the dissections were performed very close to the gallbladder with the aim of avoiding any damage to the CBD or to the hepatic artery.

We also analyzed the following additional variables: whether the patients had ERCP done before or after surgery; the findings and results of the ERCP; the type of surgery performed, LC vs open cholecystectomy (OC); whether IOC, open CBD stone extraction, and/or Roux-en-Y hepaticojejunostomy (RYH) was performed; the duration of surgery; the need for conversion to laparotomy; major complications; and the length of the postoperative stay. The follow-up of the patients ranged from 3 to 122 months (median, 17 months).

### 2. Results

During the studied period, 223 patients were diagnosed with cholelithiasis at our institution (approximately 12 patients per year). Of these patients, 119 were female, and the median age was 11 years (range, 2-17 years).

Comorbidities were present in 177 patients (79.3%); the most common are listed in Table 1.

<table>
<thead>
<tr>
<th>Comorbidity</th>
<th>No. of patients (%)</th>
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<tbody>
<tr>
<td>Hemolytic disorders</td>
<td>139 (62.3)</td>
</tr>
<tr>
<td>History of bariatric surgery</td>
<td>16 (7.1)</td>
</tr>
<tr>
<td>Others (portal hypertension and cardiac disorders)</td>
<td>22 (9.9)</td>
</tr>
</tbody>
</table>

A cholecystectomy was done for all patients with cholelithiasis, even if they were asymptomatic. Symptoms were present in 134 patients (60.0% of all patients), as detailed in Table 2. Symptoms were present in 61 patients with hemolytic disorders (63.3% of them) and in 5 patients with previous bariatric surgery (31.2% of them). A total of 56 patients (25.1% of all patients) presented with complications as the first sign of cholelithiasis, including pancreatitis in 28, choledocolithiasis in 12, and acute calculous cholecystitis (ACC) in 16. Multiple gallstones were observed in 146 patients (64.4% of all patients), and 83 (37.2% of all patients) had 1 or 2 calculi in the gallbladder.

An LC was performed in 204 patients, OC was performed in 17 patients, and 2 patients underwent RYH. The reasons for initial OC indication are shown in Table 3. One 16-year-old adolescent girl had Mirizzi syndrome, which is characterized by extrinsic compression of the common hepatic duct by stones impacted in the cystic duct or the gallbladder neck.

Regarding LC, the average duration of the procedure was 80.8 minutes (range, 40-240 minutes), and the average hospital stay was 1.1 days (range, 1-3 days). A conversion to an OC was necessary in 4 cases (1.9% of LC patients), 2 of which were cases of ACC and 2 of which were patients with portal hypertension. All 4 conversions occurred near the beginning of our experience in the 1990s. An IOC was performed in 15 cases where there was suspicion for choledocolithiasis and yielded positive results in only 1 patient (0.4% of all patients). The child underwent ERCP to extract the stone after LC.

Preoperative ERCP was performed on 11 patients, and stone extraction was possible in 9 cases. The remaining 2 cases had a dilated CBD with stones. At the time of the laparotomy, a hepaticojejunostomy was performed when CBD stones extraction was not successful. Because of the

<table>
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<tr>
<th>Indications for OC</th>
<th>No. of patients</th>
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<tr>
<td>Portal hypertension</td>
<td>6</td>
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<tr>
<td>Cardiac malformations</td>
<td>6</td>
</tr>
<tr>
<td>Mirizzi syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Others (asthma, previous gastroschisis, and neurofibromatosis)</td>
<td>4</td>
</tr>
</tbody>
</table>
complexity of these cases, other procedures, such as laparoscopic CBD exploration or even open transduodenal sphincteroplasty for duct clearance, were not considered.

Major postoperative complications were observed in 2 patients. Despite preoperative and intraoperative care, 1 girl with sickle cell disease developed acute thoracic syndrome on the first postoperative day. Clinical management with hydration, blood transfusion, and antibiotics resolved this condition. The other major complication involved another girl with sickle cell disease who developed hemoperitoneum secondary to bleeding at a trocar site, and a laparotomy was required to resolve this condition. There were no injuries to the hepatic artery or to the CBD in our series.

Regarding the patients who underwent an OC or RYH, the mean lengths of hospital stay were 3.2 days (range, 1-4 days) and 18 days (range, 6-30 days), respectively, and there were no complications. During the follow-up, we did not detect symptoms that could be attributable to postcholecystectomy pain or any other morbidity related to the operation.

3. Discussion

Gallstones are increasingly found in childhood, not only because of the use of ultrasonography in the workup of abdominal pain but also because of the increase in incidence in obesity worldwide and the high frequency of hemolytic anemias in all countries [1-5].

In addition to hemolytic anemias, a history of bariatric surgery is frequently associated with cholelithiasis. Therefore, some authors recommend concurrent prophylactic cholecystectomy during laparoscopic Roux-en-Y gastric bypass in adults, considering the low morbidity associated with cholecystectomy and the high index of ulcer biliary disease [11]. We believe that concurrent prophylactic cholecystectomy may also be considered in the adolescent population.

No well-established consensus exists regarding the indications for cholecystectomy in asymptomatic pediatric patients [3]. Some authors believe that it is indicated only in patients with comorbidities, particularly hemolytic anemias [12]. In the present series, we observed that significant proportion of all patients (25.1%) developed complications related to the presence of gallstones (eg, ACC, pancreatitis, cholecdocholithiasis), and the complication was the first sign of cholelithiasis. Herzog and Bouchard [3] described a very high incidence of complications (58.0%), such as pancreatitis, cholestasis, and choledocholithiasis in patients with idiopathic gallstone disease. In another publication, 10 of 14 initially asymptomatic patients with sickle cell disease became symptomatic and required surgery. In addition, the operative time, morbidity rate, and postoperative stay were greater among symptomatic patients who underwent cholecystectomy than in those who underwent the surgery when asymptomatic [12]. Another argument is that the presence of gallstones is a major risk factor for carcinoma of the gallbladder [13]. Finally, we may consider the psychologic consequences for the child, who will live for more than 90 additional years, and for the family when a surgical disease is diagnosed.

However, we must consider the potential morbidity of “unnecessary” cholecystectomies performed on asymptomatic patients (some cholecystectomies can also be performed if the patient will need a laparotomy for other causes—eg, splenectomy in hemolytic disorders and surgery for morbid obesity). In fact, this argument should be considered before an LC is indicated for an asymptomatic patient. Besides, it is not known the long-term course of all children with asymptomatic cholelithiasis, although in adult patients with silent gallstones, it was shown no long-term risk of symptoms or adverse events leading to cholecystectomy [14]. Although we had no complications in our series of pediatric patients, a prospective follow-up study of a large population of asymptomatic patients would be necessary to answer this question.

The other interesting finding in our series was the high incidence of comorbidities (79.3%). We believe that acute complications such as pancreatitis and cholecystitis could be life threatening in these patients. In addition, the first clinical presentation of gallstone disease in 25.1% of patients included some of these complications. Finally, we may argue that, in both cases in which an RYH was necessary, the decision to perform an LC or OC earlier in the disease course would avoid this more extensive and morbid surgery.

Careful clinical and anesthetic preoperative evaluation is crucial for detecting conditions that could be dangerous to a laparoscopic approach. In such cases, we believe that open surgery is the best approach; there were no complications after laparotomy in our series (16 cases; 7.3% of patients) and no drawbacks other than a larger scar. Furthermore, the judicious use of the laparoscopic approach combined with adequate anesthetic techniques and laparoscopic skills was responsible for the low conversion (4 cases; 1.9% of LC patients) and complication rates in our series.

Another important point of discussion refers to the lessons learned by the surgeons, which were derived from previous surgical interventions in the liver or in the biliary tract, primarily liver transplantation, from deceased or living donors, or hepatic tumor resections [15,16]. Our group’s significant experience with these procedures gave us appreciable skill in completing an LC quickly (in a little more than 1 hour) and with a low conversion rate to an OC (1.9%). Finally, thanks to our technique of performing the dissections quite close to the gallbladder, we caused no injuries to the CBD or to the hepatic artery.

Routine IOC, even in suspected cases of choledocolithiasis, is often associated with negative results [9], and we identified only 1 positive case among the 15 patients studied. Although we believe that IOC may also be indicated for a better definition of the biliary anatomy and that it is a useful tool in liver and biliary tract surgeries, it is not necessary during an LC because all of the dissection procedures are performed very close to the gallbladder. In addition,
Preoperative ultrasound examinations permit the precise evaluation of the biliary tract anatomy.

Endoscopic retrograde cholangiopancreatography can be safely and effectively performed in suspected children by experienced endoscopists, during both the postoperative and preoperative periods [17]. Laparoscopic exploration of the CBD is feasible, but it is a procedure that requires training and equipment not usually available in all institutions. Moreover, unlike what is observed in adults, CBD stones in children usually pass spontaneously without any significant complication [8,9]. When ERCP failed to remove the stones, even surgery with opening of the CBD was not successful and RYH was required.

Based on our experience, we can propose a new algorithm to guide the approach to cholelithiasis in children (Fig. 2). The final conclusion is that cholecystectomy should be performed laparoscopically in pediatric patients with gallstones. This procedure has low rates of postoperative complications. In children, when a previous diagnosis of choledocolithiasis or choledochus dilation has been made, ERCP is necessary if obstructive symptoms persist, either before or after an LC. Intraoperative cholangiography and laparoscopic CBD exploration, although feasible, are not mandatory. An RYH or an open sphincteroplasty may be required when the stones present in dilated CBD cannot be extracted by ERCP. Finally, other studies using large multi-institutional databases will be able to definitely answer and confirm our conclusions.

**References**