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ORIGINAL ARTICLE

Laparoscopic versus open surgery in children with choledochal cysts: a meta-analysis

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Abstract
Objective To compare the safety and efficacy between laparoscopic and open cyst excision with hepaticojejunostomy for children with choledochal cysts using meta-analysis.
Methods Studies comparing the laparoscopic and the open choledochal cyst excision that met the inclusion criteria for data extraction were identified from electronic databases (PubMed, Embase, Science Citation Index, and the Cochrane Library) up to November 2014. The proceedings of relevant congress were also searched. The outcomes were operative time, intraoperative blood loss, time to food intake, postoperative morbidity and mortality, length of hospital stay. Outcomes were calculated as odds ratios (ORs) with 95 % confidence intervals (CIs) using RevMan 5.2.
Results Seven retrospective studies were finally included, involving a total of 1016 patients, of whom, 408 cases underwent laparoscopic cyst excision and Roux-en-Y hepaticojejunostomy (LH) and 608 cases underwent open cyst excision and Roux-en-Y hepaticojejunostomy (OH). In LH group compared with OH group, the operative time was longer (MD = 59.11, 95 % CI 27.61–90.61, $P = 0.0002$), while the length of postoperative hospital stay was less (MD = −2.01, 95 % CI −2.49 to −1.54, $P < 0.00001$), the intraoperative blood loss was lower (MD = −37.14, 95 % CI −66.69 to −7.60, $P = 0.01$) and time to food intake was less (MD = −1.14, 95 % CI −1.61 to −0.67, $P = 0.01$). The rate of postoperative morbidity was more in the OH group, but there is no statistically significant difference between the two groups in postoperative morbidity (OR = 0.52, 95 % CI 0.13–2.06, $P = 0.35$).
Conclusion Laparoscopic surgery is a feasible, safe treatment of choledochal cyst with less postoperative morbidity, a shorter length of stay and a lower blood loss when compared with open approach. With the improvement of laparoscopic techniques and deftness of surgeons practice, laparoscopic surgery may become the first choice procedure for choledochal cyst.

Keywords Laparoscopy · Hepaticojejunostomy · Choledochal cysts · Children · Meta-analysis

Introduction

Choledochal cysts are rare congenital cystic dilations of the biliary tree that present primarily in female infants and young children and are more prevalent in East Asian populations [1, 2]. The exact etiology of choledochal cysts is unknown, but anomalous pancreaticobiliary duct union (APBDU) is seen in 30–70 % of all choledochal cysts where the common bile duct (CBD) and pancreatic duct junction occurs outside the duodenum, allowing reflux of pancreatic fluid into the biliary tree [3]. Besides the classic triad of abdominal pain, right upper quadrant mass and obstructive jaundice, choledochal cysts can be associated with serious complications such as cholangitis, pancreatitis, cholelithiasis and have a high risk in developing cholangiocarcinoma [4]. Thus, total cyst excision followed by Roux-en-Y hepaticojejunostomy reconstruction is

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recommended for most patients with choledochal cysts whenever possible [5]. Laparoscopic excision of choledochal cysts has increasingly gained acceptance and applicability since its first description by Farello [6]. The potential advantages of LH include shorter recovery time and improved cosmesis. However, its safety remains a major concern compared with the conventional open surgery. Therefore, we conducted this meta-analysis to compare the clinical safety and effectiveness of LH with OH.

Materials and methods

Searching strategy

We searched databases, including PubMed, Embase, the Science Citation Index, and Cochrane Library updated to November 2014, to identify all related published studies comparing the laparoscopic and the open choledochal cyst excision with hepaticojejunostomy. The keywords used in the search were as follows: laparoscopic, open, hepaticojejunostomy, choledochal cysts. The language was restricted to English only. The citations within the reference lists of the articles were searched manually to identify additional eligible studies.

Inclusion and exclusion criteria

The studies that published up to and including November 2014 were considered eligible if they met the following inclusion criteria: (1) study reported on at least one of the outcome measures mentioned below: operative time, intraoperative blood loss, time to food intake, postoperative morbidity (including bile leakage, pancreatic leakage, cholangitis, pancreatitis, adhesive intestinal obstruction, abdominal bleeding, etc.,) and mortality, length of hospital stay; (2) population: children younger than 18 years with choledochal cysts; (3) intervention: laparoscopic cyst excision and Roux-en-Y hepaticojejunostomy (LH) versus open cyst excision and Roux-en-Y hepaticojejunostomy (OH). Abstracts from conferences and full texts without raw data available for retrieval, duplicate publications, letters, non-randomized trials, retrospective analyses and reviews were excluded. If publications were reporting on the same study population, the most informative article was included.

Study quality assessment

The quality of the literature was assessed independently by two authors (Huo-Jian Shen and Ming Xu) using the 9-star Newcastle–Ottawa Scale [7]. A full score is 9 stars, and a score $\geq$6 stars is considered to be high quality.

Data extraction and statistical analysis

Two reviewers (Huo-Jian Shen and Ming Xu) abstracted relevant information from each eligible article using a standardized form independently. Information about the characteristics of the study population, authors, publication year, study period, country, sample size, interventions, outcomes details of the surgical techniques used, and relevant outcomes were recorded. Disagreements between reviewers regarding data abstraction were resolved through discussion. Statistical analysis of dichotomous variables was performed using the odds ratio (OR) as the summary statistic, while continuous variables were analyzed using the weighted mean difference (MD). For both variables, 95 % confidence intervals (CI) were reported. Heterogeneity was assessed using the I^2 statistic. The heterogeneity among the studies was evaluated using the Mantel–Haenszel Chi-squared test, with its significance

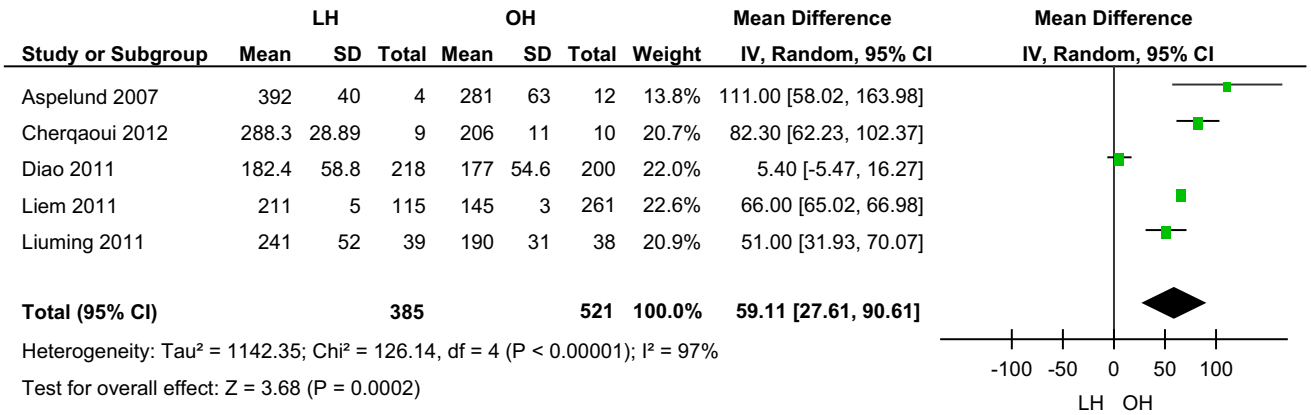

Fig. 1 Comparison of the operative time between laparoscopic group and open surgery group (random-effects model)

set at $P < 0.1$, and the extent of inconsistency was assessed by the I^2 statistic [8]. I^2 values of <25 % were defined as low heterogeneity. Those between 25 and 50 % were defined as moderate heterogeneity and those >50 % as high heterogeneity. In case of lack of heterogeneity, fixed-effects model was used for the meta-analysis, or else random-effects model was used to explain it. The estimates of the mean and SD were required to calculate the CIs for continuous data. For these tests, a P value of <0.05 was considered statistically significant. The analysis was conducted using Review Manager Version RevMan 5.2.

Results

Search results and reporting quality

According to the search strategy described previously, a total of 162 citations were obtained for review of title and abstract (Fig. 1). Of the 162 citations, 14 duplicates were removed by the Endnote X5 software, and 131 irrelevant studies were excluded through scanning titles and abstracts. Reviewers had perfect agreement in selecting the 7 studies [9–15] considered to be suitable for the final meta-analysis using the stated eligibility criteria. All 7 included studies are retrospective series. They included total of 1016 patients. Of these, 408 (40.16 %) underwent LH, and 608 (59.84 %) had OH. The characteristics, quality assessment, and outcomes for the included studies are summarized in Table 1.

Meta-analysis results

Operative time

There were five trials [9, 11–13, 15] that included information of mean and the SD about the operative time; the pooled estimates of those studies showed that the operative time was longer in the LH group (MD = 59.11, 95 % CI 27.61–90.61, $P = 0.0002$), and significant heterogeneity was present in the trials ($\chi^2 = 126.14$, $P < 0.00001$, $I^2 = 97$ %), a random effect model was considered (Fig. 1).

The intraoperative blood loss

There were only two studies [11, 15] that reported the intraoperative blood loss. The intraoperative blood loss in the LH group was significantly less than that in the OH group (MD = −37.14, 95 % CI −66.69 to −7.60, $P = 0.01$), there is a significant heterogeneity present in the trials ($\chi^2 = 3.08$, $P < 0.08$, $I^2 = 67$ %), a random effect model was considered (Fig. 2).

Table 1 Characteristics and quality assessment of the included studies

References	Study period	Group		Age (years)		Sex (M/F)		Diameter of cyst (cm)		Follow-up (months)		NOS
		LH	OH	LH	OH	LH	OH	LH	OH	LH	OH	
Ng et al. [14]	2006–2012	13	22	3.04	3.04	5/8	3/19	4.4 (1.7–15.1)	2.45 (0.7–10.7)	35	41	8
Cherqaoui et al. [13]	2000–2009	9	10	4.48	5.21	Not mentioned		2.6 (1.2–8)	2.5 (1.2–8)	8.11	67.8	7
Liuming et al. [11]	2001–2003 (LH) 1999–2001 (OH)	39	38	4	5	9/30	7/28	5 (2.4–18)	4 (2.2–16)	60		8
Liem et al. [12]	2007–2010 (LH) 2001–2006 (OH)	115	261	4 ± 0.2	5.19 ± 3.36	Not mentioned		4.76 ± 1.5	4.78 ± 1.5	Not mentioned		7
Diao et al. [15]	2001–2009 (LH) 1993–2001 (OH)	218	200	4.16	4.59	56/162	51/149	2.5–21.0	3.5–17.0	38	146	8
She et al. [10]	1978–2008	10	65	3.75	3.75	Not mentioned		Not mentioned		60		6
Aspelund et al. [9]	2000–2006	4	12	4.48	4.55	Not mentioned		Not mentioned		1	37	5

Time to food intake

Two studies [11, 15] reported that the time to food intake in the LH group was significantly shorter than that in the OH group (MD = −1.14, 95 % CI −1.61 to −0.67, $P = 0.01$), there is a significant heterogeneity in the trials ($\chi^2 = 4.40$, $P = 0.04$, $I^2 = 77$ %), a random effect model was considered (Fig. 3).

Length of hospital stay

Six studies [9–12, 14, 15] reported the length of hospital stay. It was significantly shorter in the LH than in the OH group (MD = −2.01, 95 % CI −2.49 to −1.54, $P < 0.00001$), there is a significant heterogeneity in the trials ($\chi^2 = 9.62$, $P = 0.02$, $I^2 = 69$ %), a random effect model was considered (Fig. 4).

Postoperative morbidity and mortality

Total patient morbidity was 24/408 (5.88 %) in the LH group and 129/608 (21.22 %) in the OH group. There was no statistically significant difference between LH strategy and the OH approach (OR = 0.52, 95 % CI 0.13–2.06, $P = 0.35$), but the rate of postoperative morbidity was much more in the OH group (5.88 versus 21.22 %), there is a significant heterogeneity in the trials ($\chi^2 = 37.42$, $P < 0.00001$, $I^2 = 84$ %), a random effect model was considered (Fig. 5). There was no early or late mortality postoperatively in all studies. Thus, we cannot compare the mortality data between the two study groups.

Discussion

The incidence of choledochal cysts is approximately 1 in 100,000–150,000 live births in Western countries [16] and 1 in 13,000 individuals in Japan [17]. About 80 % of choledochal cysts are diagnosed in childhood within the first decade of life [18, 19]. Since choledochal cysts can confer significant morbidity in childhood and have a high risk of biliary tract malignancy in adulthood [20], early diagnosis and treatment is very important. The optimal definitive treatment is complete excision of cysts with Roux-en-Y hepaticojejunostomy. Laparoscopic approach to choledochal cysts was first described by Farelo et al. [6] in a 6-year-old child. Since then, it has become popular and evolved at an unprecedented pace in the treatment of children with choledochal cysts [21, 22]. Because of small operative space, risk of injury to vital structures and a prolonged pneumoperitoneum, the laparoscopic surgery may potentially be a technical challenge in treatment of children with choledochal cysts for many surgeons. The safety and efficacy of laparoscopic procedure remain uncertain.

In our study, compared with the open group, less blood loss was seen in the laparoscopic surgery. It may be attributed to a better vision of the deep anatomic structures and improved accuracy provided with the magnified view in laparoscopy [23]. The time for patients to resume their diet is shorter and postoperative hospital stay is less in the LH group, it may imply a quicker recovery in the LH group. We found less postoperative complications in the group operated by laparoscopy than by laparotomy (5.88 versus 21.22 %), but there was no significant difference (OR = 0.52, 95 % CI 0.13–2.06, $P = 0.35$). Laparoscopy presented longer operative time than open laparotomy; the LH is challenged by significantly prolonged operative time, which may be associated with increased operative and anesthetic risks. There is a steep learning curve in the initial stage, the operative time gradually shortened after the surgical teams further mastered the surgical techniques, and in the more recent cases in the seven centers in different areas, the complete operation could be accomplished within 3 h, which is not inferior to that of open surgery [15, 22].

In the treatment of children with choledochal cysts by laparoscopic surgery, we think it has a variety of advantages. First, laparoscopic techniques provide a better vision of the deep anatomic structures and avoid injuring the

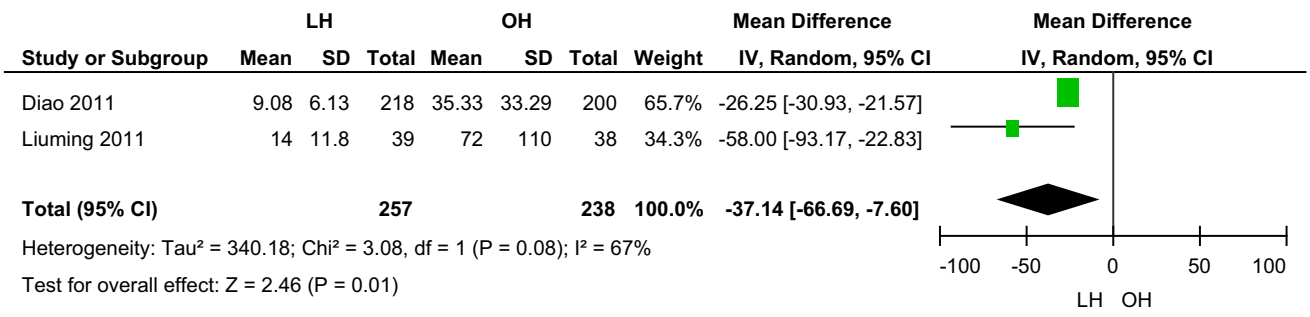

Fig. 2 Comparison of the intraoperative blood loss between laparoscopic group and open surgery group (random-effects model)

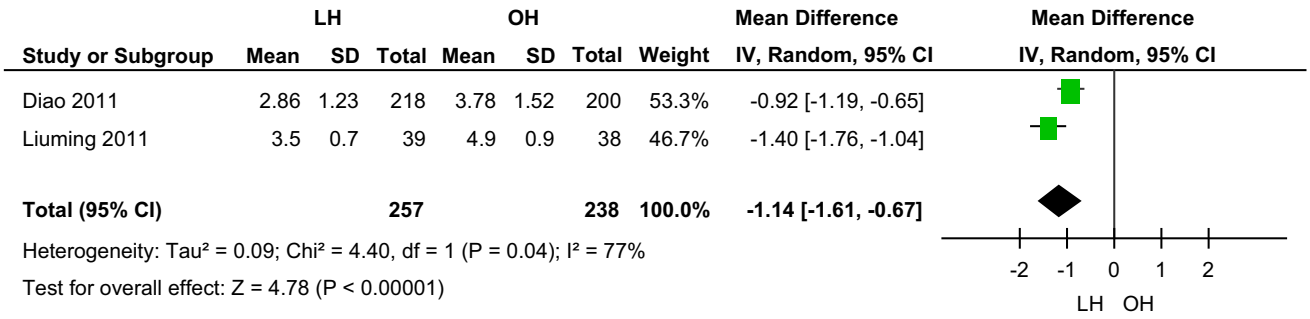

Fig. 3 Comparison of the time to first postoperative food intake between laparoscopic group and open surgery group (random-effects model)

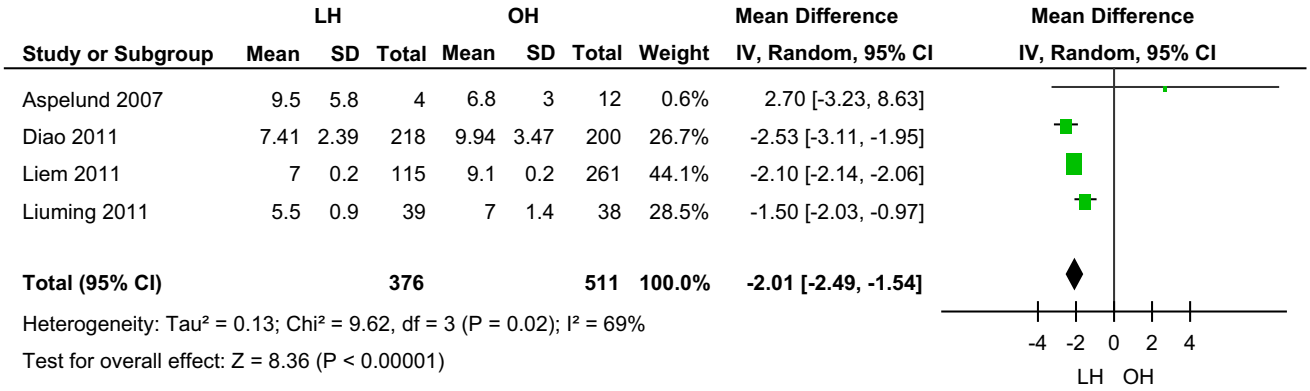

Fig. 4 Comparison of the length of postoperative hospital stay between laparoscopic group and open surgery group (random-effects model)

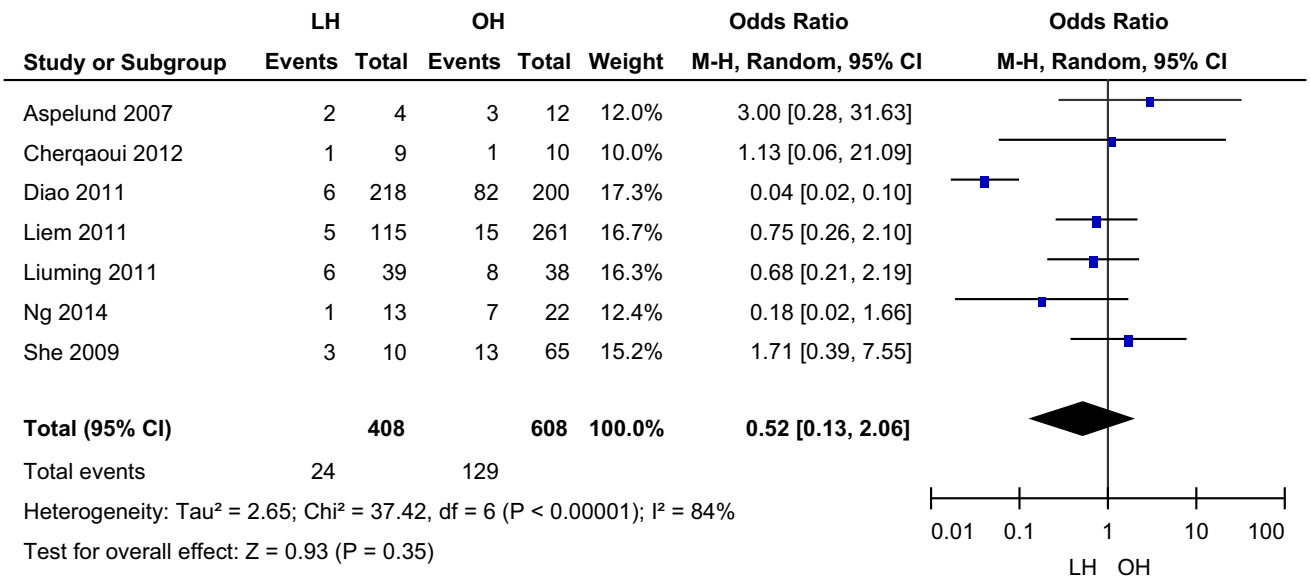

Fig. 5 Comparison of the incidence of postoperative complications between laparoscopic group and open surgery group (random-effects model)

surrounding organs, especially in childhood because of the small abdominal cavity space; second, decreased postoperative pain and small incision are beneficial to the growth of children in psychological; third, less blood loss and a quicker recovery of gastrointestinal function are also beneficial to the growth of children in physiological.

This meta-analysis has some limitations that should be taken into account when considering the results. First, there

are no randomized trials comparing the two procedures. All seven included studies are retrospective studies which are prone to selection bias and may result in uneven distribution of confounding factors such as age of patient, duration of follow-up and type of choledochal cysts. Second, except one study [14], the others did not compare outcomes between the laparoscopic and open group within the same study period, the main drawback in drawing conclusions from different study period is that factors other than the surgical approach, such as changes in perioperative practices or improved equipment, may introduce bias. Third, in some studies, the number of patients was too small, leading to low-power analyses. Caution should be given in the interpretation and generalization of this meta-analysis as the included study numbers are not high and also the overall quantity of these studies is insufficient. At last, heterogeneity among the studies is high. Therefore, a randomized, controlled trial is warranted to compare the safety and efficacy of laparoscopic versus open cyst excision and hepaticojejunostomy.

In conclusion, laparoscopic cyst excision with Roux-en-Y hepaticojejunostomy is a safe and effective treatment option for choledochal cysts in children with comparable outcomes to open resection. LH was associated with a shorter length of postoperative hospital stay and a lower blood loss when compared with open resection. With more refinement in equipment and technique, it will be possible to consider LH as a gold standard for treatment of pediatric choledochal cysts.

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