BMJ Open

Factors affecting the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric cases of congenital choledochal malformation: a retrospective case study in Southeast China

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To cite: Guo W, Zhan Y, Fang F, *et al*. Factors affecting the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric cases of congenital choledochal malformation: a retrospective case study in Southeast China. *BMJ Open* 2018;**8**:e022162. doi:10.1136/bmjopen-2018-022162

► Prepublication history and additional material for this paper are available online. To view these files, please visit the journal online (http://dx.doi.org/10.1136/bmjopen-2018-022162).

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Received 6 February 2018
Revised 2 May 2018
Accepted 11 May 2018

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ABSTRACT

Objective The aim of this study was to evaluate factors affecting the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric cases of congenital choledochal malformation (CCM).

Design A 3-year retrospective study was undertaken between January 2013 and December 2015 in four centres in China.

Setting This involved a retrospective chart review of paediatric patients with CCM in four large hospitals in Southeast China.

Participants Sixty-five paediatric patients with CCM were included in this study. We derived all available information on patient demographics, clinical characteristics, preoperative complications and surgical methods from the charts of all these patients.

Interventions Univariate and multivariate logistic regression analyses were used to evaluate factors significantly affecting the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric cases of CCM.

Results Twenty-three of the 65 case surgeries were performed using laparoscopic technique, and 42 surgeries were performed by conventional open surgery. The median operating time was 215 min (range 120–430 min). The morphological subtype of CCM and the presence of cholecystitis or cholangitis were the only factors found to affect the operating time ($p<0.05$). Logistic regression analysis confirmed cholangitis as an independent risk factor.

Conclusions The morphological subtype of CMM and the presence of cholecystitis or cholangitis are factors affecting the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric cases of CCM, whereas cholangitis is an independent risk factor.

Strengths and limitations of this study

- This was a multicentre retrospective study undertaken over a 3-year period in Southeast China.
- A stepwise logistic regression analysis of 65 paediatric congenital choledochal malformation (CCM) cases was performed in an attempt to identify factors affecting the operating time, and the accuracy of the logistic regression model for the obtained results was further assessed by the receiver operating characteristic curve.
- The sample size was relatively small; however, CCM is a rare paediatric disease. Selection bias may have been a factor.

INTRODUCTION

Congenital choledochal malformation (CCM) is a rare congenital anomaly that presents as extrahepatic and/or intrahepatic bile duct dilatation, and its incidence is much higher in Asia than in Western countries.[1–4] The majority of cases seen in the clinic are in infants and children. Most CCM cases present clinically with abdominal pain, jaundice and a mass, sometimes accompanied by vomiting and fever. CCM leads to higher rates of cholestasis, stone formation, pancreatitis, biliary hyperplasia, atypical epithelial growth of the bile duct or gallbladder, and tumour formation; the latter is thought to be caused by pancreaticobiliary maljunction (PBM) with a two-way reflux of bile and pancreatic juice.[5–8]

Early prevention of long-term complications such as recurrent cholangitis, gallstones, pancreatitis and especially malignant tumours in the bile duct and gallbladder is of essence. Complete excision of the cyst and Roux-en-Y hepaticojejunostomy is the main stay of treatment for children with CCM.[9–11] Cyst excision and hepaticojejunostomy by either open surgery or laparoscopic surgery

in paediatric CCM are feasible and popularly undertaken.[12 13] Jackson *et al* and Jenkins *et al* reported that the complexity and difficulty of a surgical procedure were reflected by the operating time.[14 15] Qiao *et al* reported on an extensive series of CCM cases in seven centres and found that postoperative complications were associated with longer operating times.[11]

Up to now, only few studies have focused on factors affecting the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric cases of CCM. We retrospectively studied the clinical presentation and surgical management of 65 paediatric cases of CCM that had undergone complete cyst excision and Roux-en-Y hepaticojejunostomy between January 2013 and December 2015 in four centres in Southeast China, in an attempt to identify factors affecting the operating time.

MATERIALS AND METHODS

Study subjects

Data were collected after study protocol approval from the institutional review boards of each participating institution, and informed consent was signed by the guardians of the subjects. All methods were carried out in strict accordance with the relevant institution guidelines regarding the acquisition and use of human tissues. Sixty-five paediatric cases of CCM were recruited into this clinical study. Twenty-nine subjects were from the Children's Hospital of Soochow University, 11 subjects were from Xuzhou Children's Hospitals, 12 subjects were from the Affiliated Hospital of Guizhou Medical College and 13 subjects were from the Affiliated Hospital of Nantong Medical College. All patients were diagnosed by ultrasound, CT, MR cholangiopancreatography (MRCP) and intraoperative cholangiography (IOC). All patients underwent surgical excision of their choledochal cyst and hepaticojejunostomy by the way of similar laparoscopic or conventional open surgical approaches, performed by paediatric surgeons of the four participating hospitals. All procedures were undertaken by expert paediatric surgeons who had advanced skills and were capable of independently completing surgical procedures in cases with huge choledochal cysts. A total of 23 patients underwent their surgical procedure by way of the laparoscopic technique. In all of these patients, a similar laparoscopic surgical approach was used by the operating surgeons in the participating hospitals. The operating times (from incision to skin closure) of each surgical team were analysed among the four subject cohorts.

Factor analysis

The patients were assessed for gender, age, common bile duct shape, Todani type and presence of a stone in the choledocal cyst. Ages were grouped as infant (<1 year of age) or paediatric (>1 year of age). Common bile duct shape, Todani type and presence of cyst stones were assessed by ultrasound and MRCP combined with IOC. Common bile duct shape was classified as either cystiform or fusiform. Clinical symptoms at presentation, preoperative complications, operative methods (conventional open or laparoscopic cyst excision and hepaticojejunostomy) and operating time were analysed. Cholangitis was defined as any symptom of the Charcot triad (abdominal pain, jaundice or fever) with an increase in total bilirubin, Aspartate transaminase (AST) and Alanine aminotransferase (ALT) levels above the normal range. Cholecystitis was confirmed by ultrasound combined with pathological findings. In 80% (52/65) of cases, the operating time was less than 300 min. The operating time was then dichotomised as <300 min or ≥300 min (figure 1).

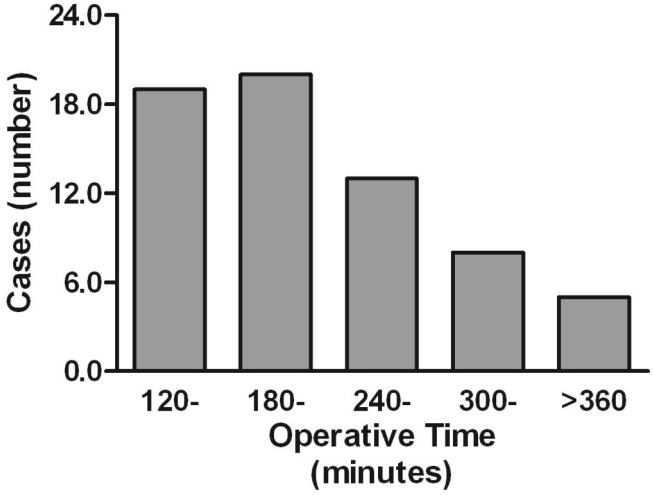

Figure 1 Distribution of the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric congenital choledochal malformation cases. The operating time in 80% (52/65) of cases was <300 min.

Patient and public involvement

There was no patient or public involvement in this study.

Statistical analysis

Analyses were performed with SAS software, JMP V.9.0 (SAS Institute). Data are presented as number (n), percentage and median. Multigroup ordered variable data comparisons were performed using Kruskal-Wallis test. Univariate comparisons were performed using non-parametric one-way Wilcoxon rank-sum, χ^2 or t-test, depending on the statistical distribution. To evaluate risk factors affecting the operating time, logistic regression analysis was performed. The receiver operating characteristic (ROC) curve was used to assess the accuracy of the logistic regression model. P<0.05 was considered statistically significant.

RESULTS

Clinical characteristics and general findings of paediatric CCM cases

The present clinical study comprised 65 paediatric CCM cases diagnosed by ultrasound, CT, MRCP and IOC. Among these 65 patients, there were 25 (38.5%) males and 40 (61.5%) females with CCM. The median age of

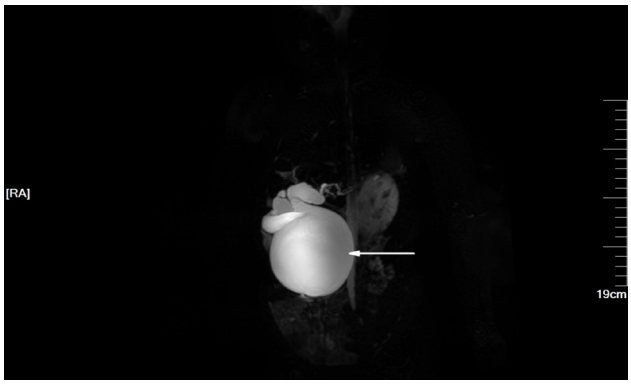

Figure 2 A paediatric patient with congenital choledochal malformation (Todani type, type IV). MR cholangiopancreatography shows the cystic dilatation of the common bile duct (arrow).

the patients was 3 years (range 3 months to 13 years). Fifty-two cases were defined as PBM by MRCP with IOC. The morphological subtype of bile duct dilatation was cystic (figure 2) in 40 cases (eight were large cysts descending into the introitus of the pelvis) and fusiform (figure 3) in 25 cases. All 65 cases underwent complete cyst excision and Roux-en-Y hepaticojejunostomy. Twenty-three of these cases were performed using laparoscopic technique, whereas the other 42 cases were performed using conventional open surgery. The median operating time was 215 min (range 120–430 min). Figure 4 and online supplementary table 1 illustrate the distribution of operating time between the four centres, and there was no significant difference in the operating time across centres.

Related risk factors affecting the operating time in paediatric CCM cases

Table 1 depicts a comparison of the clinical characteristics, preoperative complications and surgical methods for the <300 min and ≥300 min operating time groups. Online supplementary table 2 depicts a comparison of the operating time of surgical methods between laparoscopic surgery and open surgery. Univariate analysis revealed

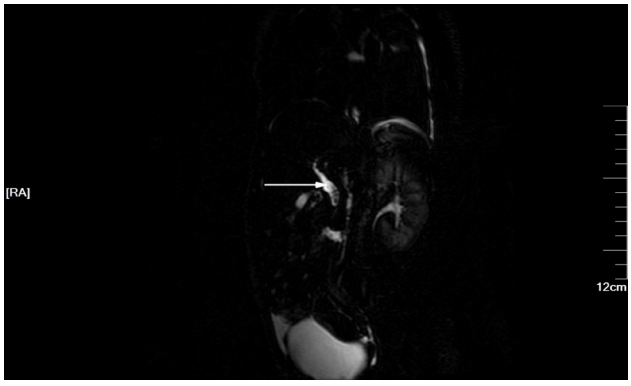

Figure 3 A paediatric patient with congenital choledochal malformation (Todani type, type I). MR cholangiopancreatography shows the fusiformis dilatation of the common bile duct with stones (arrow).

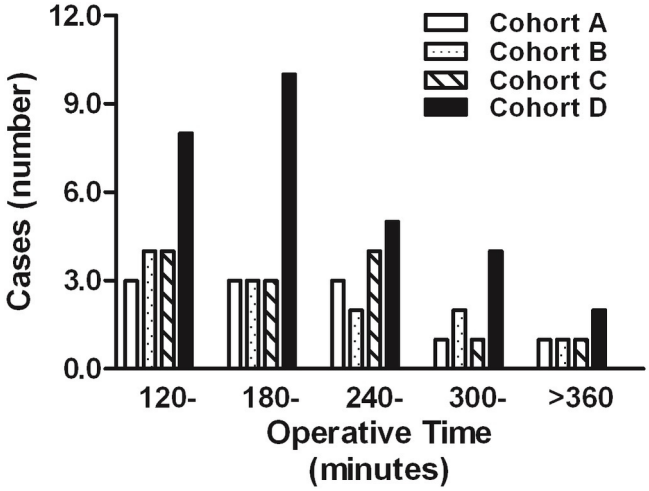

Figure 4 Distribution of the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric congenital choledochal malformation cases from four cohorts. There was no significant difference in the distribution of operating time among the four surgical teams (χ^2=0.7189, p=0.9489).

that the morphological subtype and the presence of cholecystitis or cholangitis affected the operating time (p<0.05). No other factors were found to affect the operating time. Logistic regression analysis was performed to assess independent predictors that affect the operating time in 65 paediatric CCM cases. Again, the presence of cholangitis was the most important risk factor (table 2). These data agreed with Hosmer and Lemeshow's goodness-of-fit test (p=0.4191). To further assess the accuracy of logistic regression model for the obtained results, analysis of the ROC curve was performed with the area under the curve at 0.8913 (figure 5).

DISCUSSION

General findings

Due to the obstruction of flow of bile or pancreatic juice, paediatric cases of CCM often present with abdominal pain, jaundice and an abdominal mass. They also harbour a risk of malignancy as well as other serious complications. Pre-emptive surgery is recommended once diagnosis of CMM is confirmed. The operating time has been used previously as a measure of operative difficulty and complexity.[14 15] Sorokin *et al* reported that increased postoperative complications were associated with the prolonged operating time.[16] Qiao *et al* reported similar findings on assessing 956 paediatric cases with CCM, in which all the patients had undergone complete cyst excision and Roux-en-Y hepaticojejunostomy.[11] The mean operating time in the said study was 3.52 hours (range 2.21–5.75 hours). This operating time is similar to that in our study. However, there has been only little research focusing on the risk factors affecting operating time in CCM surgery. Our study on a multicentre cohort of paediatric patients who underwent complete cyst excision and hepaticojejunostomy yielded operating times

Table 1 Risk factors associate with operating time in paediatric CCM cases

Variables	Duration of surgery <300 min (52 cases)	Duration of surgery ≥300 min (13 cases)	P values
Gender (F)	35	5	0.0559
Infant	7	2	1.0000
Type IV	22	5	
Type I	30	8	0.7472
Fusiformis*	24	1	0.0114
Cyst stones	7	4	0.2092
Biliary tract infection***	3	10	<0.0001
Abdominal pain	41	9	0.4617
Jaundice	16	3	0.7385
Mass	15	3	1.0000
Vomit	33	10	0.2551
Fever	25	10	0.0620
Cholecystitis*	28	12	0.0114
Pancreatitis	2	0	1.0000
Laparoscopy	19	4	0.6972
Preoperative complications	10	–	0.0856
Gastrointestinal polyps	1	–	
Cyst rupture	1	–	
Appendicitis	2	–	
Ascites	1	–	
Pleural effusion	1	–	
Upper respiratory tract infection	4	–	

*P<0.05, ***P<0.001.
CCM, congenital choledochal malformation; F, female.

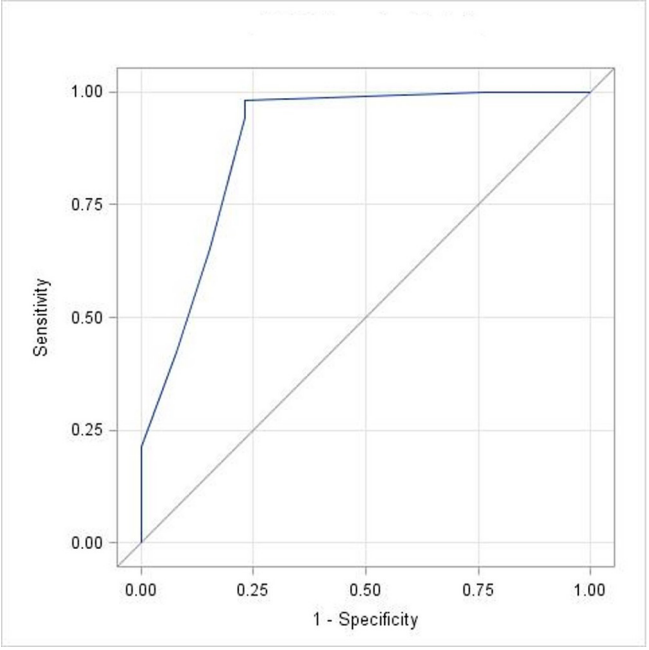

Figure 5 Use of the receiver operating characteristic curve to assess the accuracy of the logistic regression model. The area under the curve is at 0.8913 for the obtained results.

ranging from 120 to 430 min, similar to those previously reported.[17] Thus, we are persuaded that the variable of operating time can be used as a measure of operative difficulty and complexity. Moreover, we also focused on factors that potentially affected the operating time.

Factors affecting the operating time in paediatric CCM cases

To investigate factors that potentially influenced the operating time, we described the clinical characteristics, preoperative complications and the surgical methods employed in our series of paediatric CCM cases. Univariate analysis indicated that the morphological subtype and the presence of cholecystitis or cholangitis affected the operating time for complete cyst excision and Roux-en-Y hepaticojejunostomy. Patients with cystic biliary dilatations had longer operating times compared with those with fusiform biliary dilatations, and this was especially true for those with larger cysts. Patients with cystic biliary dilatations often presented with displacement of vessels, severe biliary obstruction and coagulopathy, consequently making surgery difficult and with a greater risk of injury to the surrounding tissues. This in turn increased the operating time. These results are comparable with those previously reported.[18] This indicates that the operative difficulty and complexity in patients with cystic biliary dilatations is greater than those with fusiform biliary dilatations. In addition, we compared operating times for laparoscopic and conventional open surgery. We found that the operating time for laparoscopic surgery was longer compared with conventional open surgery, although the difference in operating times between the two groups was not statistically

Table 2 Logistic regression model for factors affecting the operating time in paediatric congenital choledochal malformation cases

Variable	χ^2	OR	95% Wald confidence limits	P values
Biliary tract infection***	11.2195	30.609	4.134 to 226.607	0.0008
Cystiform	1.1194	3.377	0.354 to 32.191	0.2900
Cholecystitis	1.8755	5.892	0.465 to 74.573	0.1708

Hosmer and Lemeshow goodness-of-fit test (p=0.4191)
***P<0.001.

significant. This finding is consistent with the report by Diao *et al*.[19]

The presence of cholecystitis was another factor affecting the operating time. Forty of our cases presented with cholecystitis, of which 75.0% were defined as having CCM with PBM. Sugai *et al* previously compared the preoperative ultrasound measurements of gallbladder wall thickness and the actual wall thickness measurements in resected gallbladder specimens; the results showed a concordance rate of 89%, with 25.9% cases showing chronic inflammation.[20] Park *et al* reported that CCM cases with PBM were often found to have clinical cholecystitis, and that this was strongly related to inflammatory changes in the duct and periductal structures.[21] It is likely that chronic cholecystitis resulting in the formation of adhesions is a factor, contributing to the increase in the difficulty of procedures and dissection time. As such, in CCM cases with PBM having cholecystitis, meticulous planning is needed to decrease the risk of operative complications.

Cholangitis is a common preoperative comorbidity that may increase the operating time in CCM. Morine *et al*, on assessing 952 paediatric biliary dilatation cases, reported that 16.2% of these patients also had cholangitis.[22] Ouaissi *et al* and Visser *et al*, in their studies assessing several CCM cases, respectively, reported 26.3% and 21% of patients having cholangitis.[23 24] Lal *et al* compared cases with and without infective complications and concluded that CCM with infective complications required meticulous planning and a multimodal approach, with more time required for the operative procedure.[25] In our study, 20.0% of cases had infective complications. Multiple logistic regression analysis confirmed that the presence of cholangitis was an independent factor affecting the operating time, suggesting that such cases needed more operating time and meticulous planning to decrease the risk of operative complications. Qiao *et al* reported that paediatric CCM with cholangitis led to increased operating time and postoperative complications. This finding is further supported by Diao *et al*.[11 18] It is conceivable that the increased operating time during complete cyst excision and Roux-en-Y hepaticojejunostomy contributes to the operative complications and postoperative morbidity in these cases. However, given that the operating time reflected the difficulty of surgical procedures, and also the relative experience of the surgeon, efforts were made to ensure that the paediatric surgeons involved in this study across all four centres were similarly and suitably trained. While the operating time is seemingly not an adequate surrogate for predicting long-term postoperative complications, our findings highlight the importance not only of meticulously planning surgical intervention for paediatric patients with CCM but also of the necessity to schedule these cases so as to allow for the effective use of operating room time in order to improve the quality of patient care.

Limitations

Our study has a number of limitations. First, the sample size was relatively small; however, CCM is a rare paediatric disease. Selection bias such as surgical methods may have been a factor. Second, this was a retrospective multicentre study of only 3 years' duration. A prospective multicentre study over a longer period is a necessary future endeavour.

CONCLUSIONS

In this series of complete cyst excision and Roux-en-Y hepaticojejunostomy in paediatric CCM cases, the median operating time was 215 min (range 120–430 min). The morphological CCM subtype and the presence of cholecystitis or cholangitis affected the operating time, with cholangitis as an independent risk factor. These findings are potentially important and will be useful in planning surgical interventions for paediatric patients with CCM in China. This has the potential to improve the quality of patient care in CCM in China .

Contributors S-gH and JW designed the study. W-lG, S-gH, FF, ZY, Y-bD and J-gZ collected data. Y-bD, J-gZ, FF and ZY analysed data. W-lG, S-gH and JW wrote the manuscript, and all authors read and approved the final version of the manuscript.

Funding This work was supported by Jiangsu Province Health and Family Planning Projects (H201519 and H201520) and Jiangsu Province Social Development Program - Standardized Diagnosis and Treatment of Key Disease in Clinic (BE2015643). Fundamental and Application Research in Health Care of Suzhou (SYS201563). Science and Echnology Development Project of Suzhou (SS201428).

Competing interests None declared.

Patient consent Guardian consent obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement No additional data are available.

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