# Double versus single T-tube drainage for frank cysto-biliary communication in patients with hepatic cystic echinococcosis: A retrospective cohort study with median 11 years follow-up

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## Abstract

Background: Partial peri-cystectomy is one of the major surgical approaches for hepatic cystic echinococcosis (CE) and has been practiced in most centers worldwide. Cysto-biliary communication (fistula, leakage, rupture) is a problematic issue in CE patients. T-tube is a useful technique in situations where exploration and decompression are needed for common bile duct (CBD), however, postoperative biliary complications for cystic cavity still remains to be studied in depth.

Materials and Methods: A retrospective cohort analysis of CE cases in our single center database from 2007 March to 2012 December was performed. Patients (n=51) were divided into two cohorts: double T-tube drainage (one at CBD for decompression and one at the fistula for sustaining in cystic cavity, n=23) group and single T-tube drainage cohort (only one at CBD for decompression, n=28). Short-/long-term postoperative complication focusing on biliary system was recorded in detail and they were followed-up for median 11 years.

Results: Overall biliary complication rates for double and single T-tube drainages were 17.4% vs. 35.7%. Short-term complications ranged from minor to major leakages, cavity infection and abscess formation, and prevalence was 17.4% vs. 21.4% respectively for double and single T-tube groups; most importantly, double T-tube drainage group had predominant advantage regarding long-term complications (0% vs. 14.3%), which was biliary stricture needing surgery and it was observed in single T-tube drainage group.

Conclusions: Double T-tube drainage had better outcomes without procedure specific postoperative biliary complications than single T-tube drainage. Meanwhile, we recommend long-term follow-up when comparing residual cavity related biliary complication in CE patients as it could happen lately.

## Background

Human cystic echinococcosis (CE) in one of the lethal infectious diseases and causes severe organ damage[1, 2]. Mortality of echinococcosis is > 90% within 10–15 years if left untreated or inadequately treated after initial diagnosis. Radical resection, which was mainly the total peri-cystectomy with non-opened cyst and liver resection are considered as the best curable options[2, 3]. However, not all cases indicate radical approach due to crucial cyst location or corresponding complications, then needs non-radical surgeries. The inflating growth pattern of CE within the liver often leads to various vasculature comorbidities[1, 4, 5]. Some of them presents certain therapeutic challenges for surgeons and consumes patience of the subjects by lowering life quality, and even takes lives. Cysto-biliary complications (fistula, leakage, rupture) is one of the most frequent complications[4, 6]. According to reports, communication between cystic cavity and the intra-hepatic biliary tracts could be classified as major (> 5 mm in diameter), minor (< 5 mm in diameter) and invisible (occult, hard to observe with naked eye but definitely exists). Invisible or occult communication occurs in 10–37% of CE patients, however, frank cysto-biliary communication (FCBC) is an open intercommunication between the cystic cavity and intra-hepatic bile ducts that allows the contents of the cyst to drain directly into the bile duct as well as biliary poring into the cystic cavity[7]. FCBC could cause obstructive jaundice, cholangitis, cystic infection, gastrointestinal discomfort and naplulaxis[8].

Therefore, cholecystectomy, CBD exploration as well as T-tube drainage has been practiced after partial peri-cystectomy (PPC) for eradicating intra-biliary debris and decompression,and gradually became routine procedure[9, 10]. In this situation, the ruptured site was managed by suturing, hepatectomy of relevant liver parenchyma, residual cavity drainage, omentoplasty etc., however, postoperative complications still

draw attention[9–12]. Long term follow-up after this surgical approach has revealed potentially serious postoperative complications, including biliary stricture, biliary fistula, wound infection as well as abscess formation[5, 6]. Additionally, most health care professionals have limited experience with such situations outside the endemic region. In our center, extra T-tube was introduced as a sustain of the FCBC site in order to achieve better outcome more than a decade ago, and we call it double T-tube drainage. Initially, it was an evolutionary process: non-T-tube catheter was placed in the cystic cavity for leakage drainage, and it evolved gradually into T-tube which was introduced right into the FCBC site. The first cohort that was designed specially to compare the biliary outcome in such patients from 2007 March, and those were enrolled in ten-year follow-up period. Currently, the follow-up was finished with comparable prognosis, which we thought would be helpful to academic society and infectious disease professionals.

## Patients Materials And Methods

From 2007 March to 2012 December, 51 patients without hepatobiliary surgical history were underwent PPC in our center due to CE with FCBC. Their most common complaints were abdominal pain, jaundice, nausea and fever. Patient demographics, cyst features and clinical symptoms were presented in Table 1. Preoperative computed tomography (CT) and magnetic resonance imaging and cholangiopancreatography (MR/MRCP) results indicated that, the certain bile duct where rupture occurred, mostly were lobular or segmental bile ducts (Fig. 1,2). PPC was performed in all patients; debris in biliary tracts was removed through CBD using choledochoscopy after cholecystectomy; biliary tract was explored by injecting methylene blue (1:250 dilution with normal saline) to discover the FCBC site (Fig. 3, also see supplementary file/video). Owing to the basis of FCBC, setting decompression T-tube and sustaining T-tube drainage has been introduced. Consequently, we retrospectively divided these special subjects into two groups based on their operative

procedures (double T-tube drainage group and single T-tube drainage group, specifically mentioned blow), forming this research cohort.

## Double T-tube drainage

Schematic diagram (Fig. 4A) presented this procedure which was performed in 23 patients. In this method, main operation steps includes: (i) PPC followed by exploration of inside surface of remnant peri-cyst to discover potential ruptured bile ducts; (ii) then cholecystectomy, choledotomy and choledochoscopic exploration, removal of feces or debris from cyst contents in biliary tracts were performed, then the biliary tree was tested by infusing methylene blue through CBD, minor leakages in the residual cavity were sutured by using $4-0$ or $5-0$ absorbable sutures; (iii) upon exposuring the FCBC bile duct, one sustaining T-tube was placed where it ruptured, followed by a regular decompression T-tube at the CBD was introduced, thereafter, the rupture as well as the biliary tracs was reexamined by flushing methylene blue through decompression T-tube until no leakage was noticed; (iv) after careful examination and assuring patency of biliary tree, other two routine catheters were placed respectively at the residual cavity and the hepatic hilar region for post-surgical observation of any bile leakage or infections.

## Single T-tube drainage with sutured fistula

Schematic diagram (Fig. 4B) showed this procedure which was performed in 28 patients. Main steps (i), (ii) and (iv) were same with above method; (iii) the specific ruptured bile duct was identified sutured using $5-0$ or $6-0$ absorbable sutures, then a regular decompression T-tube at CBD was set, thereafter, reexamination by flushing methylene blue through T-tube was performed until no leakage was noticed.

## Postoperative T-tube removal

All subjects hold decompression T-tubes for about one month after cholangiographic

examination was negative in both groups; and all sustaining T-tubes were removed if the results showed no leakage and indicates fully recover with the help of cholangiographic examination at three months after the surgery in double T-tube drainage group (Fig. 5).

## Patient follow-up and data analysis

Subject were followed-up after discharge from hospital at every 6 months for the first two years and at every 1–2 year afterwards. Follow-up results were obtained by visiting or interviewing through phone contacts. Attentive collection of all relevant data was finished. Finally, their clinical data were analyzed, including demographic characteristics, cystic lesion features, operative methods, hospital-stay, short-/long-term biliary complications.

## Results

In general, there were no mortality and no malignancy observed in any of these patients. In terms of prognosis, altogether 37 (72.5%) patients achieved clinical cure at early stage, and other 14 (27.5%) cases suffered from short-/long-term complications to certain degrees, but equally reached clinical cure after corresponding surgical interventions. Short term complication rate was 10 (19.6%) while long term complications occurred in 4 (7.8%) patients. Postoperative complications between the two groups were shown in Table 2.

## Short term outcomes

(a) Minor biliary fistula(≤ 250 ml/24 h) was observed in 3 (10.7%) cases in single T-tube group, while in 2 (8.7%) in double T-tube group, and the former three ceased within 1 week by extended peritoneal drainage, but the latter two stopped with longer duration of T-tube except from extended peritoneal drainage. (b) One case (3.6%) in single T-tube group struggled from major biliary fistula(□250 ml/24 h), unfortunately, the case developed into peritonitis and received reoperation with peritoneal lavage and

debridement. (c) Infection of cavity was occurred in 1 (3.6%) patient in single T-tube group, which was managed by wound care and debridement. Whereas, in double T-tube group, 2 (8.7%) subjects with infection of incisions were treated by antibiotic therapy and wound care by meticulous dressing and debridement. (d) Abscess or infection of residual cavity was only discovered in 1 (3.57%) case in single T-tube group, which was treated through the percutaneous drainage.

## Long term outcomes

In the long term, no complications were noticed in double T-tube group, however, biliary stricture was discovered in 4 (14.3%) in single T-tube group. Out of these patients, one of them, luckily, accomplished dilation of biliary stenosis and cured, thanks to decompression from percutaneous cholangiography and drainage of upper biliary tree. While, two of them recovered through reoperation with continuous T-tube sustaining which took half year to heal. Another one case had to receive left hepatectomy to stop continues biliary stenosis after two years.

## Discussion

FCBC with overt passage of intra-cystic material to the biliary tract is a serious complication, and the reported frequency is 3–37% in different case series[6, 12]. Intrabiliary rupture mainly results from compression of bile ducts from the gradually increasing cysts, causing bile stasis. The increased intraductal pressure induces fissure formation in the duct wall while local intracystic water pressure of up to 80 cm lead to the rupture of the cyst into biliary tract[13]. Intrabiliary rupture mainly established in centrally localized cysts near to the hilum in liver segments III, Ⅳ, V, Ⅵ, Ⅶ and there was also a correlation with the size especially those size over 10cm[9, 14, 15]. The main approach of surgery for hydatid cyst with intrabiliary rupture should be: cystic content

evacuation, cavity management, clearance of cystic material from biliary tract with assurance of patent biliary channel and restoration of normal biliary flow. Although opinion on cavity management vary, but there is general consensus that meticulous CBD exploration with clearance of cystic material and maintenance of normal patency of biliary channel is mandatory.

For attaining proper cavity management with proper healing of communication between cavity and biliary system, the role of decompression of biliary tract is fundamental. Various technique including choledochoduodenostomy, T-tube drainage, and transduodenal sphincteroplasty have been recommended to decompress intrabiliary pressure[5, 11]. Placement of T-tube enables easier monitoring and seems to be traditional method after intrabiliary rupture of CE. But use of T-tube is not free of complications. The most frequently encountered morbidities were external biliary fistulas and suppuration of the residual cavity and postoperative biliary strictures. To prevent these late complications such as stricture, after PPC and T-tube decompression, we further perform extra T-tube in the cystobiliary orifice of bile duct in residual cavity. However, since it still remains controversial whether or not double T-tube decreases the risk of postoperative complications, long term follow-up is necessary to evaluate the outcomes of double T-tube decompression.

In current study, we evaluated the outcomes in 51 patients with FCBC who were treated by double or single T-tube drainages. However, nonnegligible drawbacks have been reported to affect the outcomes of PPC. The main immediate postoperative complications were biliary leakage and fistulas, along with septic complications of the residual cavity leading to prolonged hospital stays[16]. Most of these complications may be ascribed to the pericyst lining the residual cavity. A pericyst left in situ, especially if thick and calcified, represents an obstacle to liver regeneration filling the residual cavity, thus leading to

serum and blood accumulation or liver abscess formation[5, 6, 16]. Furthermore, the persistence of the pericyst hides possible biliary communication in the residual cavity, considered the main reason for biliary leakage. However, it should be stressed that even when biliary communications are identified, their closure within a stiff, calcified pericyst wall would not be easy or effective. Furthermore, infection, biliary fistula, and slow reduction of the cyst cavity may lead to more serious complications, such as obstruction of main hepatic ducts or the portal vein[1, 9, 11]. And, for this reason, we recommend a long-term follow-up for such situations, which was our another aim to report this study results a after 11 years observation. In these cases, reoperation is quite complex, with high mortality due to the technical difficulties related to distorted liver anatomy, deteriorated liver function, and poor general conditions. In our study, the overall stricture rate was 14.3% vs 0%, with late complications being more frequent in single vs. double T-tube drainage groups.

Although the longer follow-up period in double T-tube drainage group was necessary, no complications of stricture have been observed to date. It suggested that in most cases of FCBC, the cyst involved major bile duct or located near to hilum, and the residual cavity get shrinked and fibrosed gradually due to persistent leakages and recurrent infections. Progressive shrinkage and fibrosis lead to development of traction in underlying bile duct which gets stenosed and resultant stricture formation. So, introducing the sustaining T-tube in orifice of residual cavity, bile duct maintained the patency and was protected from postoperative strictures in double T-tube drainage group. We believed that after PPC and T-tube decompression, a further T-tube insertion into orifice of residual cavity bile duct was essential for the prevention of postoperative residual cavity infections and strictures especially when the location of the cyst is near to the hilum.

Based on our single center experience and current study, we demonstrate that specific

indications may comply there instructions: (1) performing double T-tube drainage should be individualized, not just depend no FCBC size or location; (2) when the FCBC site is lobular or segmental bile ducts and near to hepatic hilum, or the duct was interrupted due to the rupture, a double T-tube drainage should be considered first; (3) when there is ulceration due to inflammation at the FCBC site, suturing may be difficult and double T-tube is recommended; (4) if corresponding upper level biliary tree is just a single or small-in-size liver segment, if there is liver atrophy at relevant liver parenchyma, if there is severe liver damage so it would not be saved properly, then a hepatectomy should be accompanied; (5) usually, > 5 mm diameter bile duct needs double T-tube drainage more than < 5 mm ones; (6) above items should be thought in an integrative way to achieve best practice and best patient outcome.

Shortcomings of this study is that it was a retrospective study based on relatively mall sample size, and higher quality randomized case control studies would be very helpful to achieve better evidence-based results. In addition, removal timepoint should be optimized due to late leakage could happen at original cysto-biliary communication site after withdrawal of decompression T-tube, in which occasion longer decompression was necessary.

## Conclusion

This research evaluated feasibility, safety and efficacy of additional sustaining T-tube of intrahepatic bile duct where the rupture occurred in the residual cavity. Although a longer follow-up period was necessary, this study revealed satisfactory follow up results of double T-tube drainage (decompression plus sustaining) compared to single T-tube drainage (decompression) after PPC for CE.

## List Abbreviations

CBD=common bile duct, CE=hepatic cystic echinococcosis, CT=computed tomography, FCBC= frank cysto-biliary complication, MR/MRCP= magnetic resonance imaging and retrograde cholangiopancreatography, PPC=partial peri-cystectomy.

## Declarations

### Ethics approval and consent to participate

All procedures were performed in adherence to the terms of the latest version of the Declaration of Helsinki for Medical Research involving Human Subjects and approved by the Ethical Committee of the First Affiliated Hospital of Xinjiang Medical University. All included patients aged more than 18 years and written informed consent for anonymous collection and analysis of clinical data was provided by all patients before surgery in this retrospective cohort.

### Consent for publication

All patients signed written informed consent for publication of relevant data.

### Availability of data and materials

The datasets used/analyzed during the current study are within the manuscript and more data could be available from the corresponding author on reasonable request.

### Competing interests

The authors declare no conflict of interest regarding this manuscript.

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**Authors' contributions**

YY, TA, YS, HW designed the surgery, conceived the whole study, revised the manuscript and approved for publication; TA conceptualized and supervised during the research, acquired funds and critically revised the manuscript; PS, TJ and BR originally collected all clinical data; PS interpreted, analyzed and presented the data, drafted, and he was also responsible for data and quality control of the study; AAini collected radiological and photographic materials, edited the schematic diagram in this paper, and revised the article; RZ, QG, AAhan, AAbulizi participated the surgeries and assisted PS in data collection and patient follow-ups; TA, YS, PS, YY, TJ, BR, RZ, QG, AAhan participated patient management in hospital; YY assisted AAini in radiological data collection and clinical data providing. All authors together have read the final version and approved the manuscript.

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## Tables

Due to technical limitations, tables are only available as a download in the supplemental files section

## Figures

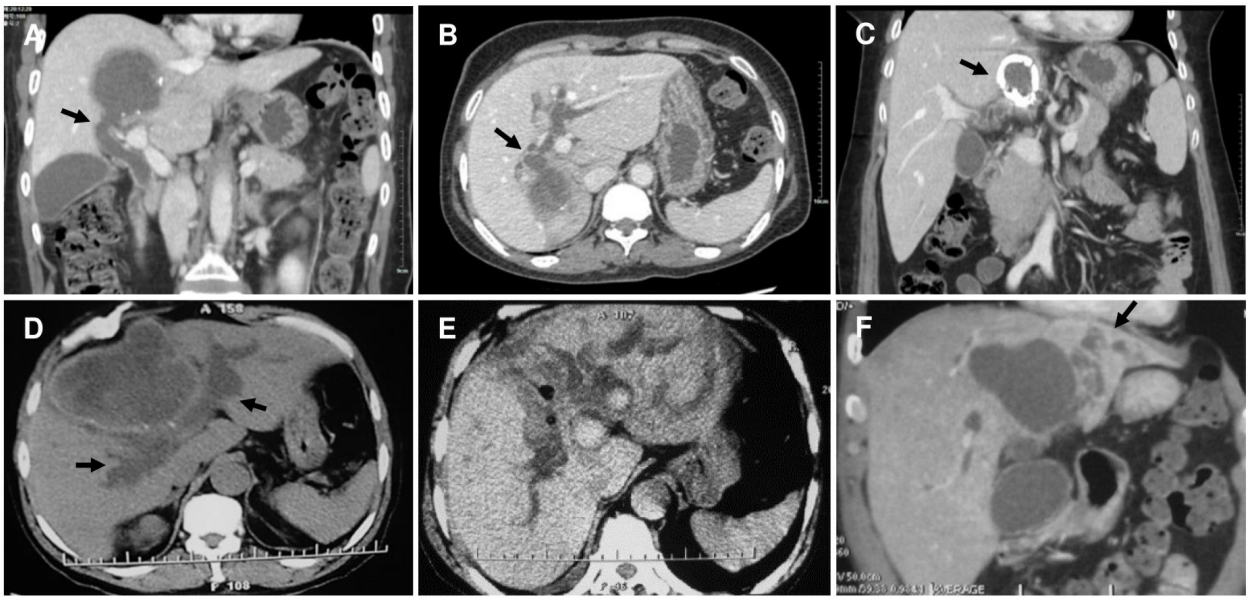



Figure 1

CT presentations of frank cysto-biliary communications. (A) Cysto-biliary communication (arrow) at right supra section and debris-filled common bile duct; (B) Cysto-biliary communication (arrow) at right posterior lobe; (C) Capsule calcified cyst (arrow) interlinking with left hepatic duct; (D) Medial lobular cyst presenting cysto-biliary communication to major hepatic ducts form both sides (arrows); (E) Severe left lateral lobe liver damage caused by cysto-biliary communication; (F) Hepato-atrophy (arrow) of left lateral lobe led by cysto-biliary communication.

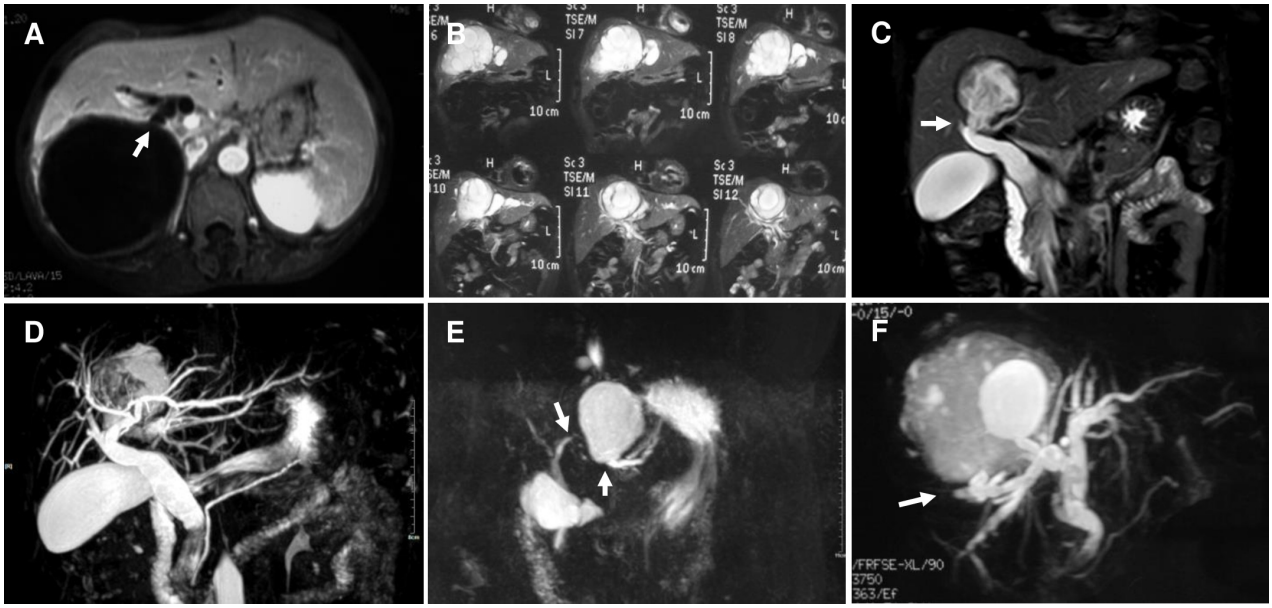
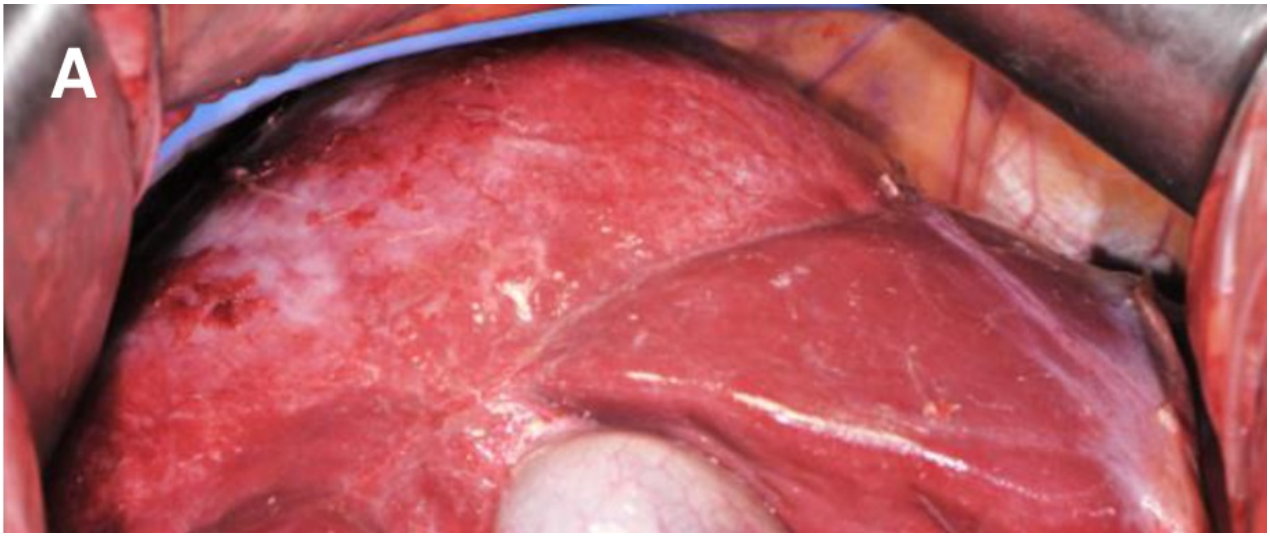



Figure 2

MR/MRCP manifestations of frank cysto-biliary communications. (A) Cysto-biliary communication (arrow) at right posterior lobe; (B) Cysto-biliary communication of “nested” cyst at main ductal branches of the liver; (C) Cysto-biliary communication (arrow) at right supra section; (D) Cysto-biliary communication (arrow) at right posterior lobe; (E) Left lateral lobular cyst that played as “drainage pool” of the bile via cysto-biliary communication (lower arrow), note that proximal end of left hepatic duct was cramped (upper arrow) due to functional disuse; (F) Cysto-biliary communication (arrow) at right lobe and biliary disuse of superior tributary.



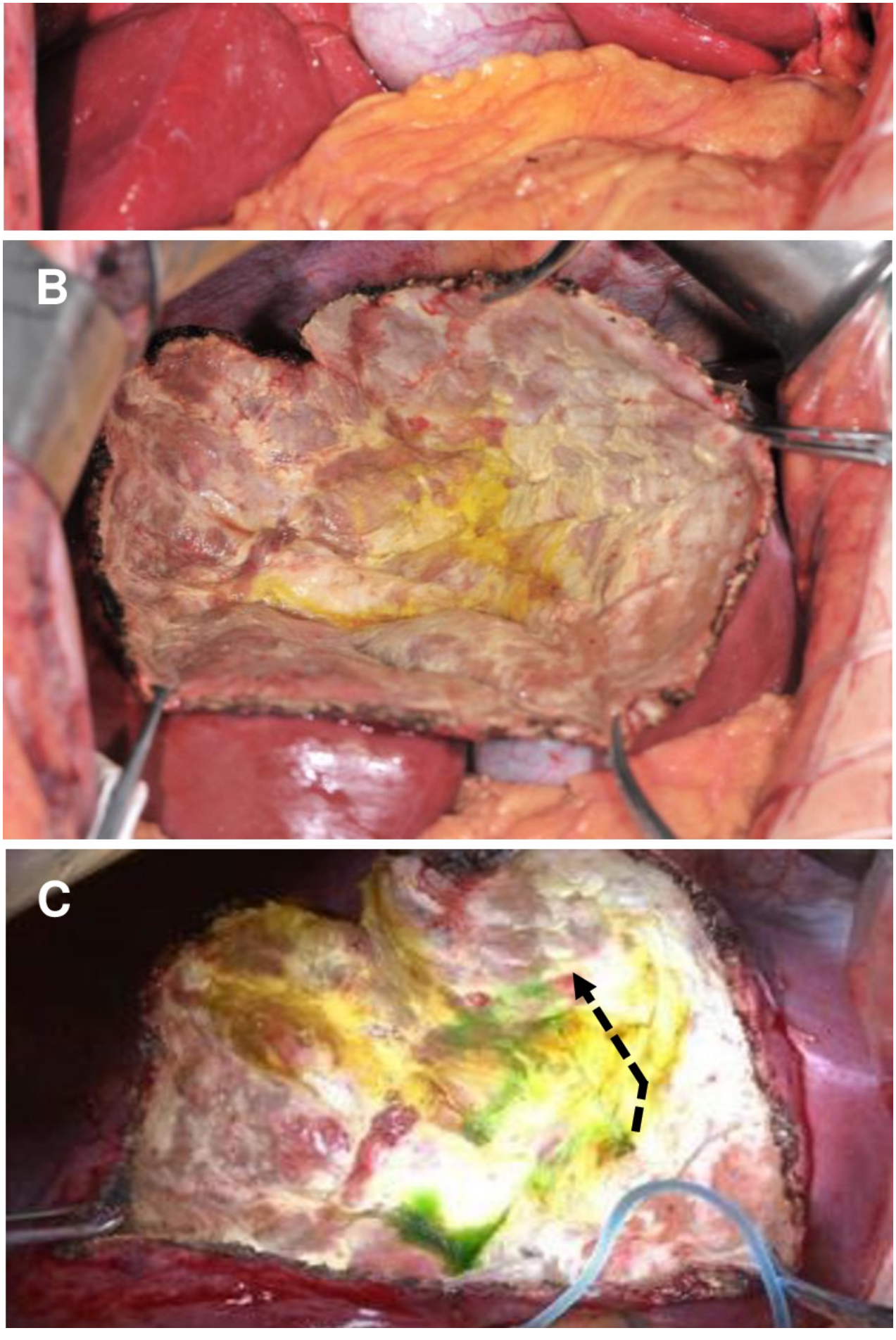

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Figure 3

Example of intraoperative technique to discover cysto-biliary communication. (A) Surgical exposure of large echinococcal cyst; (B) Posing cystic cavity for potential biliary leakage after partial peri-cystectomy plus total endo-cystectomy; (C) Liquid spray (dotted arrow) form leakage site when injecting methylene blue (1:250 diluted with normal saline).

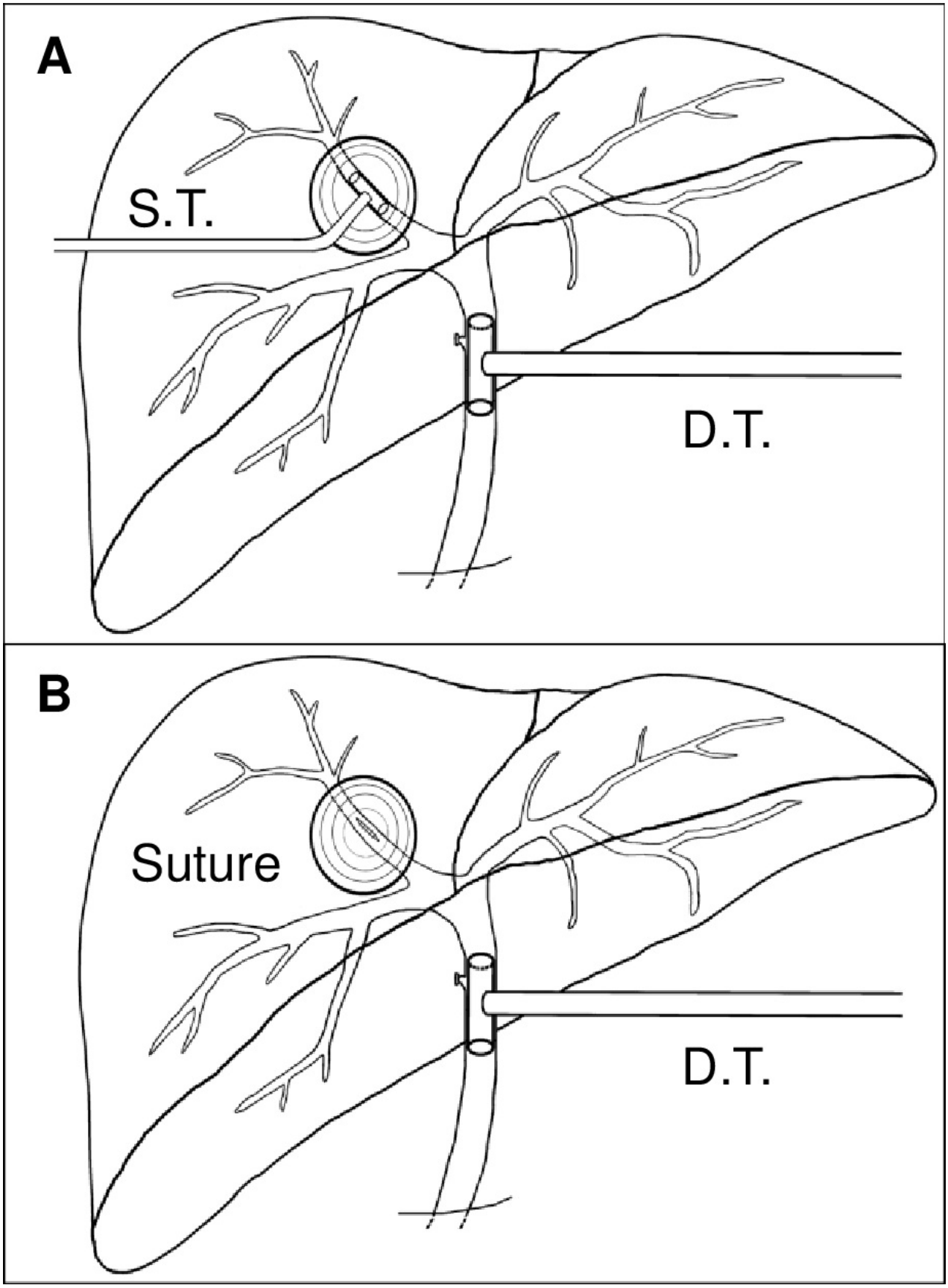


Figure 4

Schematic diagram of different biliary surgical techniques for cysto-biliary

communications. (A) Double T-tube drainage due to fistula; (B) Single T-tube drainage with sutured fistula; (C) Single T-tube drainage with untreated cystic cavity due to inapparent cysto-biliary communication. (D.T.= decompression T-tube at common bile duct; S.T.= sustaining T-tube at cysto-biliary communication site; Suture= suturing of the cysto-biliary communication site; Note that there were routine cystic cavity draining catheters within all these three techniques but they were omitted in order to magnify T-tubes in the figure).

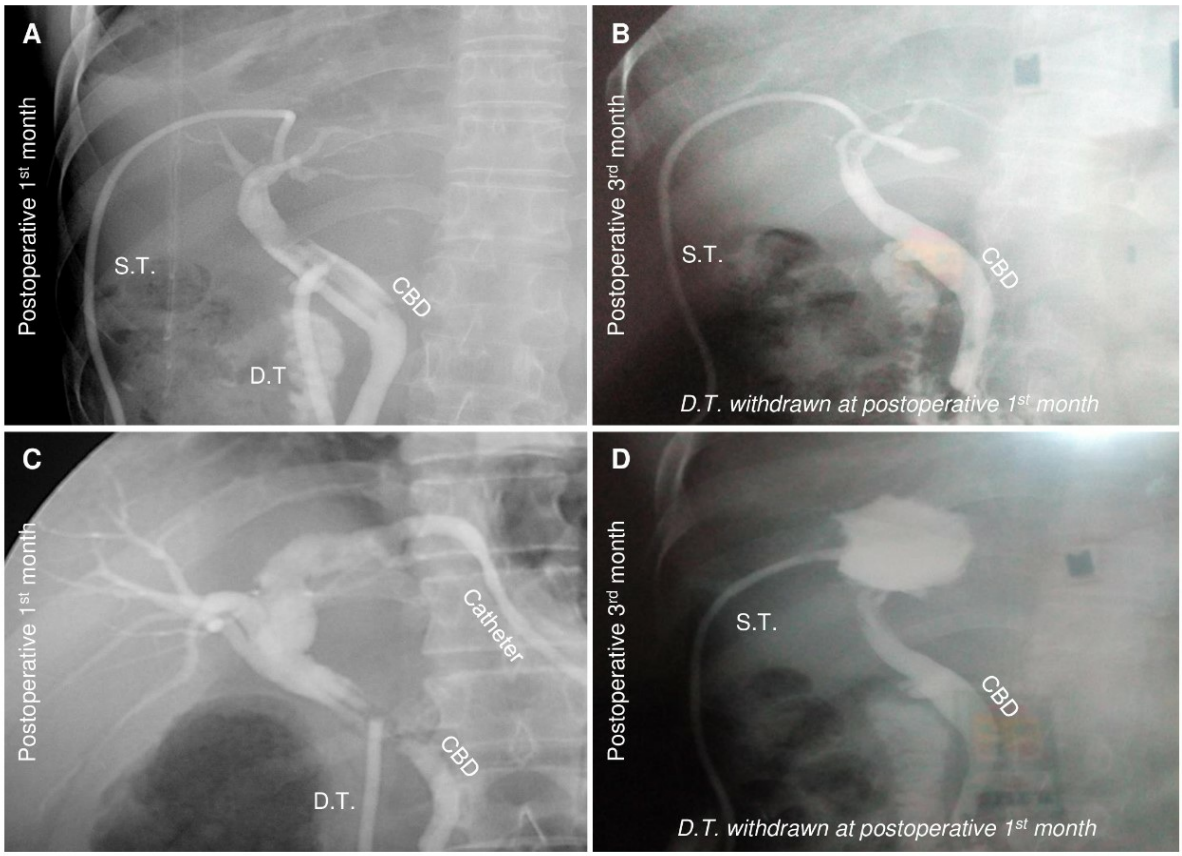


Figure 5

Postoperative cholangiograms for the tubes. (A) Satisfactory drainage without obvious leakage at postoperative first month (corresponding to double T-tube drainage); (B) Satisfactory healing of the cavity without apparent leakage at postoperative third month after withdrawal of decompression T-tube at postoperative one month (corresponding to double T-tube drainage for the same patient in A in this figure); (C) Qualified drainage without obvious leakage at postoperative first month (corresponding to single T-tube drainage with sutured fistula); (D) Unsatisfactory outcome of the original leakage in cystic cavity at postoperative third month, note that this leakage had not been observed at postoperative first month, which indicated late leakage at original cysto-biliary communication site after withdrawal of decompression T-tube at first month (corresponding to single T-tube drainage with sutured fistula).

## Supplementary Files

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Additional file 1 (Video, 15sec, 20.2MB, 1920X1080, mp4).mp4
Tables.pdf