

Long-term results of hepatic hydatid disease managed using palanivelu hydatid system: Indian experience in tertiary center

P. Senthilnathan · Swati Inamdar · V. P. Nalankilli ·
Anand Vijay · S. Rajapandian · R. Parthsarathi ·
Praveen Raj · C. Palanivelu

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Background Incidence of hepatic hydatid disease is increasing due to globalization. Surgery is the gold standard treatment. Laparoscopy has gained enough evidence regarding its safety and efficacy. Complete evacuation of hydatid contents without spillage remains a challenge. We aimed to determine long-term results of hepatic hydatid disease managed laparoscopically using palanivelu hydatid system (PHS) at our institution.

Methods One hundred and five patients underwent laparoscopic surgical management using the PHS at our institute from May 1997 to May 2013. Clinical presentations, surgical strategy, postoperative morbidity, and long-term recurrence rate were evaluated.

Results Of the 105 patients, 76 were male and 29 female with a mean age of 32 years (range 14–71 years). The most

common presentation was abdominal pain in 61 patients (58 %). Sixteen patients had multiple cysts of which nine had involvement of both lobes. Seventy-seven (73.3 %) cysts were uncomplicated. Nineteen (18.09 %) had a cyst-biliary communication, two were ruptured cysts, and seven were recurrent cysts. All patients underwent successful laparoscopic management where conservative surgery was performed in 94 patients and radical surgery in 11 patients. Post-operative morbidity was seen in 18 (17.14 %) patients, which included deep cavity infection in two cases, post-operative bile leak in 13 cases, and duodenal injury in one case without any mortality. Mean long-term follow-up was 36 months (range 6 months–5 years) with recurrence in two cases.

Conclusion Our long-term results with PHS showed good outcomes in the laparoscopic management of hepatic hydatid disease with conservative surgery as the preferred approach reserving radical surgery only in selected cases.

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P. Senthilnathan (✉) · V. P. Nalankilli · A. Vijay
HPB Department, GEM Hospital and Research Center, Pankaja
Mill Road, Ramnathpuram, Coimbatore 641045, Tamil Nadu,
India
e-mail: senthilnathan94@yahoo.com

V. P. Nalankilli
e-mail: drnalanms@yahoo.co.in

A. Vijay
e-mail: anandvij@yahoo.com

S. Inamdar
General Surgery Fellow in Minimal Access Surgery (FNB),
GEM Hospital, Pankaja Mill Road, Ramnathpuram, Coimbatore,
Tamil Nadu 641045, India
e-mail: swatidinamdar@gmail.com

S. Rajapandian · R. Parthsarathi
Colorectal Department, GEM Hospital and Research Center,
Coimbatore, Tamil Nadu, India
e-mail: srajapandian@rediffmail.com

R. Parthsarathi
e-mail: parthu@me.com

P. Raj
Bariatric Surgery Department, GEM Hospital and Research
Center, Coimbatore, Tamil Nadu, India
e-mail: praveenraj@me.com

C. Palanivelu
GEM Hospital and Research Center, Coimbatore, India
e-mail: palanivelu@me.com

Incidence of hepatic hydatid disease is increasing due to globalization. Surgery still remains the gold standard treatment in spite of emerging evidence for safe and effective alternative treatment modalities for its management. Despite its benign nature, management of this disease is a dilemma in surgical clinics due to high complication and recurrence rate. Lots of studies have been published in the literature on laparoscopic management of hepatic hydatid disease [1–3]. However, fear of anaphylactic reaction resulting from spillage of cyst contents may be one of the discouraging factors preventing wider adoption of these techniques [4]. Various instruments have been described to evacuate contents of hydatid cyst which minimizes spillage, thereby reducing risk of anaphylaxis and recurrence [1], [5–7] and [8]. We have introduced Palanivelu Hydatid System (PHS) which addresses all these problems successfully [9]. We have previously published our early experience of safe and efficacious laparoscopic management of hepatic hydatid disease with PHS which has also been confirmed by other authors [9, 10]. We have continued the laparoscopic management of hepatic hydatid disease successfully using PHS at our institution. In this study, we have evaluated our 17 years data for short and long-term results of laparoscopic management of hepatic hydatid disease using PHS.

Materials and methods

We have reviewed departmental database of all cases of hepatic hydatid disease managed surgically using the PHS at our institute over a period of 17 years from May 1997 to May 2013.

Diagnosis

In all patients, diagnosis of hepatic hydatid disease was made based on clinical findings and morphological features detected by imaging techniques (ultrasonography and computed tomography scan). Serology (indirect hemagglutination assay-IHA) was selectively used only in doubtful and atypical cases where imaging was not diagnostic. Pre-operative endoscopic retrograde cholangiopancreatography (ERCP) was considered only when a cyst-biliary communication was suspected.

All patients were treated preoperatively with albendazole (10 mg/kg/day) for 2 weeks which was continued postoperatively for 4 weeks.

The exclusion criteria

Severe coagulopathy, cysts more than 3 cm deep to the surface of liver, cysts smaller than 3 cm in diameter,

pregnancy or women who refused contraception during the period of albendazole treatment and cysts with heterogeneous complex mass (Gharbi type 4) or a calcified wall (Gharbi type 5).

Technique

The unique design of PHS has already been described in detail in our last publication [9]. (PHS is designed by us and manufactured by “Om surgical” Mumbai, India). After introducing camera port through umbilicus, the location of hydatid cyst is identified. The PHS trocar with cannula (Fig. 1) is introduced into peritoneal cavity directly over the hydatid cyst. After piercing the peritoneal cavity, trocar is removed and the cannula alone is advanced until its tip is in total contact with hydatid cyst surface. Suction is applied through the side channel to maintain contact between cyst and cannula. Continuous suction creates a vacuum seal between cyst wall and rim of cannula, which prevents spillage. Povidone iodine-soaked gauze is placed around the trocar so that if at all accidental spillage occurs, it should get absorbed into the gauze having scolicedal effect. Thereafter the trocar which is connected to separate suction apparatus is introduced into the cannula and by steady pressure pushed into the cyst along with cannula. Any spillage on puncturing the cyst wall is immediately suctioned either into body of the hollow trocar through its fenestrated tip or into outer cannula. Once the PHS enters hydatid cyst, the trocar is removed. Aspiration of hydatid fluid without spillage is the first step in all types of surgeries. This is followed by complete removal of all laminated membranes, daughter cysts, and debris. The cavity is irrigated with normal saline through main channel while simultaneously maintaining continuous suction through side channel. Once returning fluid is clear, CO₂ is insufflated at low pressure (3–4 mmHg) and separate telescope (to minimize risk of contamination) is introduced into the cavity to visualize interior of the cyst to confirm complete evacuation of daughter cysts and to look for any overt cyst-biliary communication. In early period of this study, 1.5 % cetrimide was used as scolicedal agent in the absence of overt cyst-biliary communication (verified by absence of bile staining in suctioned fluid) and normal saline (0.9 %) when it was present. Normal saline (0.9 %) alone was used irrespective of cyst-biliary communication during the last 8 years. The idea is to use large volume of normal saline to clear cyst contents (based on our concept that volume is more important than concentration of saline). Conservative surgeries that followed evacuation included deroofing, partial pericystectomy with or without omentoplasty. Radical surgeries include complete pericystectomy and liver resection. Partial pericystectomy involved resection of exposed cyst wall along with thinned out liver parenchyma

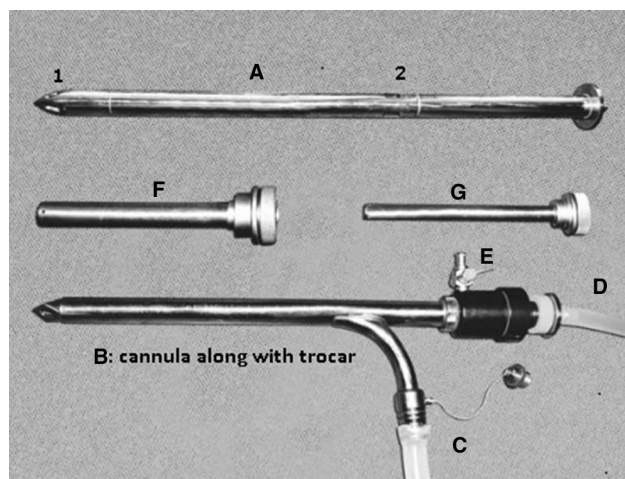


Fig. 1 The palanivelu hydatid system (PHS). *A* Hollow trocar with fenestrations at tip (1) and in midshaft (2). *B* Cannula with trocar. *C* Side channel for connection to one suction machine. *D* End channel for connection to another suction machine. *E* Air insufflation channel. *F* 12–5 mm reducer. *G* 12–10 mm reducer

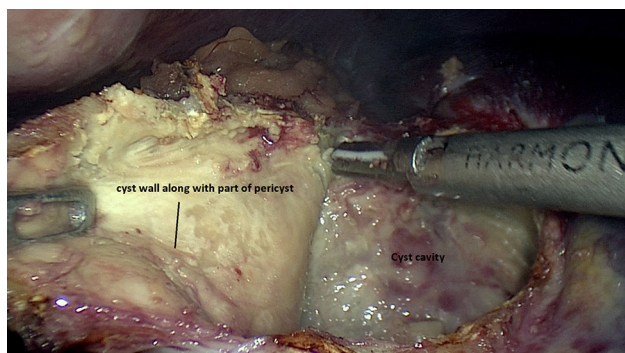


Fig. 2 Partial pericystectomy: resection of the exposed cyst wall along with thinned out liver parenchyma and pericyst at its junction with the liver using ultrasonic scalpel (Harmonic scalpel). Residual cavity managed with drains without omentoplasty since residual cavity is small and superficial

and pericyst at its junction with liver (Fig. 2). Complete pericystectomy involved non-anatomical resection of liver parenchyma 1–2 cm beyond cyst margin. Most of our surgeries were conservative reserving the radical surgery only for selected cases. Small biliary communications seen in the cyst wall were managed by laparoscopic intracorporeal suturing with 3–0 vicryl. Two drains were used, one in sub hepatic space and other in residual cavity in all cases. These drains were kept for a period of 3–10 days depending upon volume and nature of the fluid. Patients requiring a longer duration of drainage than expected were discharged with drain in situ after applying stoma bag over the tube. Drainage tube was removed when output was minimal. A persistent bile leak was defined as any measurable bile output beyond the third postoperative day. In

Table 1 Details of clinical symptoms seen in patients with hepatic hydatid disease

Symptoms	<i>n</i> (no. of patients)	Frequency (%)
Pain	61	58.10
Dyspepsia	25	23.81
Lump	12	11.43
Fever	3	2.86
Jaundice	2	1.9
Cough	2	1.9

case of persistent bile leak or a rising trend in biliary output, patients were intervened by ERCP.

Follow-up

The follow-up period ranged from 6 months to 5 years with a mean follow-up of 36 months. Imaging by ultrasound with or without aspiration cytology was used for follow-up to confirm recurrence. A CT scan was done whenever any complication or recurrence was suspected with an inconclusive ultrasound.

Results

From May 1997 to May 2013, we surgically managed 116 patients with hepatic hydatid disease. Eleven of those patients were not treated using PHS and hence were excluded from this study. Of the 105 patients, 76 were male and 29 female with a mean age of 32 years (range 14–71 years). The most common presentation was pain in 61 patients (58.10 %). Table 1 lists the most common presenting symptoms and their frequency. Eighty-nine patients had a single cyst and 16 patients had multiple cysts. Average size of the cyst was 10.4 cm (range 5–20 cm). Nine patients had involvement of both lobes. The right lobe was involved more frequently ($n = 63$, 60 %). All segments were involved with variable frequency including segment VIII (three cases) and segment V (four cases). The majority of patients ($n = 77$, 73.3 %) had uncomplicated cysts.

Complicated cysts were seen in 28 patients which included recurrent cyst ($n = 7$, 6.6 %), cyst-biliary communication ($n = 19$, 18.09 %), and ruptured cyst ($n = 2$, 1.9 %). The management of cyst-biliary communication is highlighted in Table 2.

Left lateral segmentectomy was performed during the early period of this study. Other procedures performed are mentioned in detail in Table 3.

In special situations like cysts situated in segment VIII, the cyst wall was accessed through the intercostal space by

Table 2 Details of management of patients with cyst-biliary communication with hepatic hydatid disease

Cyst-biliary communication (Total, <i>n</i> = 19)	Intraoperative suturing	Lap CBD exploration	ERCP	Conservative management with drain tube
Pre-operative diagnosis (1)	–	–	1 (Pre op)	–
Intra -operative diagnosis (6)	5	1	1 (where intraoperative suturing done but failed to control bile leak postoperatively)	–
Post-operatively became evident (12)	–	–	2 (Post op)	10

Table 3 Details of the types of surgery performed in patients with hepatic hydatid disease

Type of surgery	<i>n</i> (%)
Evacuation and cystectomy/partial pericystectomy	91 (86.7)
Evacuation and left lateral segmentectomy	10 (9.5)
Total pericystectomy	1 (0.95)
SILS (single incision laparoscopic surgery) partial pericystectomy with partial splenectomy	1 (0.95)
Laparoscopic lavage and removal of all daughter cysts along with good wash with normal saline, in 2 cases of ruptured hydatid cyst.	2 (1.9)

Table 4 Complications

Post-operative complications	<i>n</i> (%)
Persistent bile leak	13 (12.38)
Secondary deep cavity infection	2 (1.9)
Duodenal injury	1 (0.9)
Post op recurrence	2 (1.9)
Anaphylactic shock	Nil
Mortality	Nil

PHS trocar under laparoscopic guidance. Lung injury was avoided by reducing the tidal volume. We have also done single-incision laparoscopic surgery for liver hydatid cyst along with splenic hydatid cyst where we did partial pericystectomy for liver hydatid with partial splenectomy.

The average duration of surgery was 52 min. No obvious intraoperative spillage or anaphylaxis was seen in any of the patients. Average hospital stay was 3.2 days (range 2–8 days). Overall, morbidity was seen in 18 patients (17.14 %). The complications occurred during this study are mentioned in Table 4.

One asymptomatic patient with large cyst-biliary communication which was not suspected preoperatively required CBD exploration to clear daughter cysts during laparoscopy. Choledochotomy was done with lavage and T tube placement. This patient was not having cholangitis, and no preoperative investigations were suggestive of intrabiliary communication. He improved well without any morbidity.

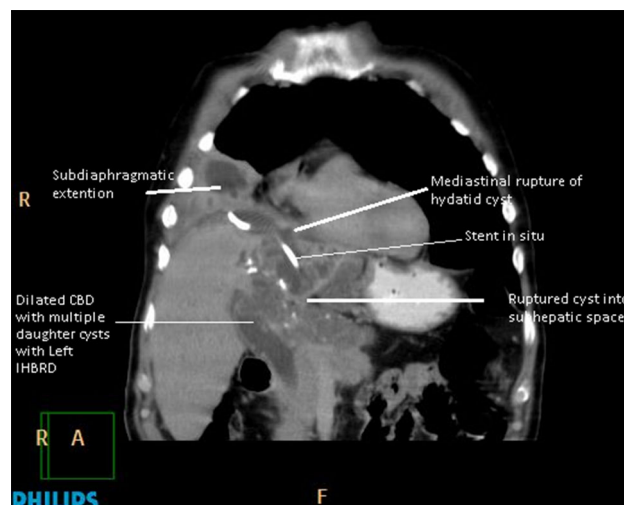


Fig. 3 CT scan showing ruptured hydatid cyst into *right* subdiaphragmatic and the subhepatic space extending into the mediastinum. Hydatid cyst has also ruptured into biliary system. Dilated CBD with multiple daughter cysts in it and *left* intrahepatic biliary radical dilatation (IHBRD) seen. Pigtail stent which was placed during ERCP into stomach opening was seen in *right* subdiaphragmatic space

Another patient presented with cholangitis. CT scan of abdomen was suggestive of ruptured liver hydatid into mediastinum along with frank intrabiliary rupture (Fig. 3). ERCP along with clearance of all daughter cysts and stenting was done (Fig. 4). On withdrawing scope, we noticed another small opening into pyloric region. Stent was placed in that opening. When cholangitis was under control, all daughter cysts in the mediastinum were evacuated laparoscopically (transhiatal) and adequate saline (0.9 %) wash was given. The stent which was placed into the stomach opening was seen in mediastinum (Fig. 5). He recovered well without any morbidity and no recurrence in 1-year follow-up. This type of rare presentation and its laparoscopic management is not reported in the literature until now.

Fifteen patients (14.28 %) were lost to follow-up. Till date, recurrence was seen in only two cases; both were detected within 6 months post-operatively: one was intrahepatic and other in peritoneal cavity; both were managed laparoscopically.

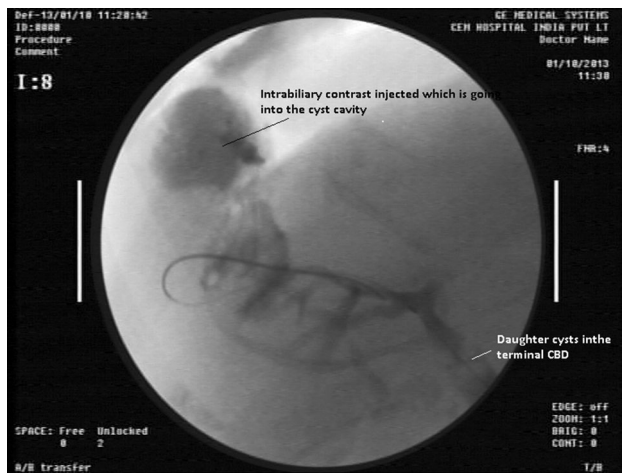


Fig. 4 ERCP is suggestive of frank intrabiliary rupture. The contrast injected in CBD is going into the cyst cavity showing frank cystobiliary communication. Daughter cysts can be seen in CBD causing CBD dilatation

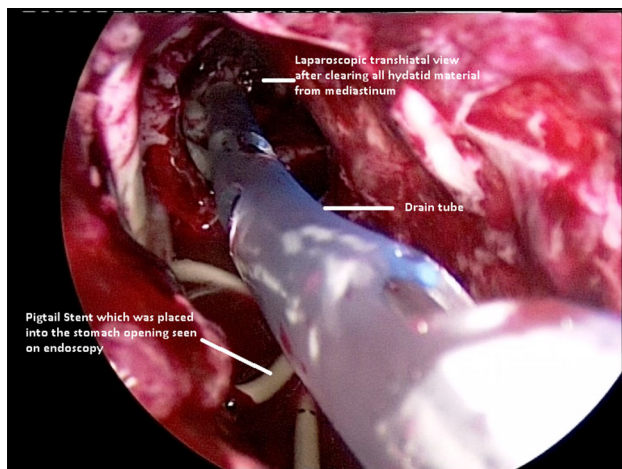


Fig. 5 Laparoscopic view of transhiatal approach for mediastinal-ruptured hydatid cyst. After thorough wash and removal of all hydatid materials along with daughter cysts from subdiaphragmatic space, mediastinal extension was noticed. Through transhiatal approach mediastinal-ruptured hydatid contents removed completely and the final view can be seen in figure after drain tube placement. Pigtail stent which was placed into stomach opening was seen in the mediastinum. This was suggestive of intra-gastric extension of ruptured hydatid cyst (this is unique case not yet reported)

Discussion

Hydatid cyst is a parasitic disease caused by the tapeworm *Echinococcus granulosus*. It can be present with any organ involvement, liver being the most common. Due to globalization many surgeons across the world can encounter cases of hepatic hydatid disease. Surgery is the mainstay of treatment although medical management and percutaneous aspiration, injection and re-aspiration (PAIR) have been reported in the recent literature to have equally good results

in selected cases [11]. The first report of laparoscopic treatment of hepatic hydatid disease was published in 1994 [1]. Soon thereafter the first report of anaphylactic shock complicating its treatment was published [12]. This was followed by an exaggerated fear of anaphylaxis which seemed to discourage surgeons from widely adopting minimal access techniques for its treatment [4]. However, gradually reports started appearing in the world literature detailing laparoscopic management of hepatic hydatid disease [1–3, 11, 13, 14]. Laparoscopic treatment produces encouraging results with the added advantages of laparoscopy, though its feasibility and safety have been questioned. No prospective randomized clinical trials comparing laparoscopic with open surgical treatment of hepatic hydatid disease have been reported. Our series of 105 patients is one of the largest series in the literature till date, and our selection criteria were truly permissive.

The surgery was based on principles of complete evacuation of the cyst without spillage, followed by a thorough saline wash to achieve complete evacuation of contents (in the earlier period of study the focus was on sterilization of cyst contents by scolicidal agent) and obliteration of the cavity. One of the problems faced in laparoscopic treatment is the difficulty in evacuating particulate contents, the daughter cysts and the laminated membrane without spillage. Various instruments have been described in the literature [1, 5–8]. As there is high pressure in the cyst, any attempt to puncture it even with a fine needle may result in a leak around the puncture site. The puncture needle gets obstructed very easily by the germinative membrane itself or by the daughter cysts, necessitating withdrawal that causes further leakage. Ramachandran et al. [15] inserted a 12-gauge aspiration needle into the cyst and positioned a 5-mm suction cannula next to the needle to aspirate any spillage. This maneuver can be helpful in open surgery but might be dangerous during laparoscopy since vision and control over the operative field may be lost with sudden decrease in intra-abdominal pressure. The vacuum seal created using PHS helps maintain the negative pressure inside, segregating it from high pressure pneumoperitoneum giving undisturbed laparoscopic vision. Due to two separate suctions, double negative pressure is created which prevents spillage and assists complete evacuation of the cyst. The theoretical risk of anaphylactic shock after spillage is minimal once the cyst fluid is adequately sterilized by scolicidal agents along with perioperative albendazole cover.

Although many procedures and technical variations have been described in the literature, none of them can be considered as gold standard. Instead a tailored approach is required. Many techniques like simple drainage, deroofting, capitonnage [16], introflexion [17], omentoplasty [18], pericystectomy [3, 19], and hepatic resection [20] have

been performed alone or in combination. Cysts in deeper location and with a harder pericyst may cause cavity-related problems after simple drainage. This problem can be solved with the use of more advanced laparoscopic techniques such as pericystectomy or hepatic resection which in return may increase morbidity. As per Ediz Altinli [18] omentoplasty decreased the deep cavity-related complications effectively with less morbidity compared to radical surgery. We have done omentoplasty in all our cases where deeper cavity was formed after cyst evacuation. Few authors advocate radical procedures, mainly pericystectomy due to lower rate of recurrence and deep cavity-related complications [2, 11, 13, 19]. Others find it time consuming with increased blood loss and not justifiable for a benign disease [15]. Dziri et al. [21] found that intraoperative vascular injuries occurred more frequently in association with pericystectomy. Main advantage of total pericystectomy in open surgery is that it literally eliminates the problems of spillage and cavity management but in laparoscopy, cyst management without prior evacuation carries the risk of perforation and dissemination of disease. This procedure of evacuation is done by PHS effectively.

We used PHS in all cases as the initial step to evacuate all hydatid materials without spillage irrespective of conservative or radical procedure. Radical surgery was done only in 11 cases (10.47 %), while others were managed by conservative surgery. Although some proponents of radical surgery [22, 23] claim low recurrence rate, in our study, we had only 1.9 % ($n = 2$) recurrences even after 89.53 % ($n = 94$) of patients being managed by conservative surgery. We managed two ruptured cysts laparoscopically by thorough lavage and suctioning of all daughter cysts along with albendazole cover.

A recent meta-analysis of 14 studies analyzing the outcomes of surgery in endemic areas reported minor and major complication rates of 33 and 25.1 % respectively, with a mortality rate of 0.7 % [24, 25]. We had overall morbidity of 17.14 % ($n = 18$) with zero mortality. Cyst-biliary communication is one of the most common but still underestimated and troublesome complication. It is reported to occur at a relatively high rate of 5–30 % with wide range of complications, such as biliary obstruction, fistula, infection and secondary biliary cirrhosis [26]. We came across 19 cases (18.09 %) of cyst-biliary communication. The pressure dynamics arising from higher intracystic pressures than the resting bile duct pressure may explain its pathophysiology [26]. Most of them are occult in nature and cannot be diagnosed preoperatively. Several studies reported a history of cholangitis, high bilirubin, high alkaline phosphatase level, a cyst larger than 10 cm and the presence of ultrasound findings which suggest intrabiliary rupture as clinical predictors of intrabiliary rupture. ERCP was suggested to confirm the biliary communication [27–

29]. We could not find any significance of such predictors in our study. One out of 19 cases of cyst-biliary communication was diagnosed preoperatively because he presented with cholangitis with diagnostic ultrasound findings of daughter cysts in CBD. Preoperative ERCP followed by laparoscopic transhepatic drainage of hydatid cyst was performed in this case.

The therapeutic options for intra-operatively detected cyst-biliary communication are multiple and related to the size of the communication, the location of the cyst, and the experience of the hepatobiliary surgeon. Laparoscopy has the advantage of magnified view in detecting even minute biliary communication. Although we have done suture ligation in five cases of which only one required ERCP eventually, we have not encountered any reports on the success rate of suture ligation attempts in the literature. In fact, more failure rates have been reported by Zeybek et al. [26]. Another technique described is intraoperative CBD exploration with T tube placement which is not commonly required due to advent of ERCP. It was required only in one of our cases where biliary opening was too big to suture and frank rupture of cyst contents into CBD was obvious. Post-operatively patient recovered well without any complications.

In spite of several surgical techniques used to control intra-operatively detected cyst-biliary communication, 10–32 % of cases eventually develop a postoperative biliary fistula [28]. Certain fistulae may not be detected intraoperatively even after meticulous search with magnified laparoscopic view. Other than technical failure, this may be partially explained by the fluctuating pressure dynamics in the biliary system arising from intermittent contractions of sphincter of Oddi [28]. This explains 11.43 % ($n = 12$) rate of postoperative biliary fistula where intraoperatively there was no evidence of biliary communication, in our study. There is no significant difference in the development of postoperative biliary fistula based on the type of procedure (conservative or radical) [26]. ERCP has been advocated in high-output fistulae with duration of more than 1 week and in low-output fistulae with duration of more than 3 weeks with no signs of reduction in biliary output [26]. Due to routine placement of drain we were able to manage ten out of 12 cases of persistent bile leak without re-exploration while remaining two of them required ERCP.

A recurrence rate of 0.9–22 % is reported in literature for open surgery [30, 31]. With conservative and radical laparoscopic treatment, a number of authors have shown recurrence rates comparable to those of open operations thereby establishing the efficacy of laparoscopy [15]. Spillage should cause intraperitoneal recurrences but most of recurrences are intrahepatic. Baskaran et al. [14] believe that recurrence is possibly due to missed ectocysts that

escaped the scolicide and our scrutiny due to their ectocystic location. Alonso Casado O et al. [32] suggested that near zero recurrence can be obtained with radical operations since we will be removing all possible ectocysts en mass. There are no prospective randomized clinical trials available in the literature comparing conservative versus radical approach in terms of safety, complications and recurrences. All the available data is in the form of case series and single institute experience with very small number of cases. In our study, we had only 1.9 % (two cases) recurrences even after 89.53 % (94 cases) being managed by conservative surgery with the help of PHS. Both were detected within 6 months post-operatively. Recurrence was intrahepatic in one case and into peritoneal cavity in other one. Both were managed laparoscopically with no further recurrence until now. In spite of less evidence in literature favoring conservative approach, we have found it to be the most appropriate and adequate surgery with comparable recurrence rate.

Conclusions

Laparoscopic management of hepatic hydatid disease is feasible and safe in expert hands. Our long-term follow-up shows low recurrence rate with PHS which proves the efficacy of the apparatus. All complicated hydatid cysts mainly related to biliary communication, recurrent cysts, and even ruptured cysts can also be effectively managed by laparoscopy with all its added advantages. ERCP may eliminate the need of CBD exploration during same surgery or re-exploration in selected cases of cyst-biliary communication. We recommend conservative surgery with PHS as preferred approach since it has comparable recurrence rate to that of radical approach published in the literature with minimum complications reserving radical surgery only in selected cases.

Disclosures Dr. P. Senthilnathan (corresponding author), Dr. Swati Inamdar, Dr. V.P. Nalankilli, Dr Anand Vijay Dr. S Rajapandian, Dr. R.Parthasarathi, Dr. Praveen Raj, Dr. C. Palanivelu have no conflict of interest or financial ties to disclose.

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