

# Hydatid cysts of the liver opening in the biliary tracts: about 120 cases comparative study between radical and conservative treatment

Ghofrane Talbi<sup>1</sup>, Wael Ferjaoui<sup>1</sup>, Sahir Omrani<sup>1</sup>, Mohamed Hajri<sup>1</sup>, Seifeddine Baccouche<sup>1</sup>, Nizar Cherni<sup>2</sup>, Lasaad Gharbi<sup>1</sup>, Nefaa Arfaa<sup>1</sup>, Hafedh Mestiri<sup>1</sup>, Rached Bayar<sup>1</sup>

<sup>1</sup> Department of General Surgery, Mongi Slim University Hospital, Faculty of Medicine of Tunis, Tunisia

<sup>2</sup> Department of Urology, Military Hospital of Tunis, Faculty of Medicine of Tunis, Tunisia

## ABSTRACT

**Background.** Hepatic hydatid disease is still common in Tunisia, is deemed benign disease but concerns its complications; the most frequent is the rupture in biliary ducts.

The aim of the work was to study the results of surgical treatment of hydatid cysts of the open liver in the bile ducts and to deduce the predictive factors of postoperative complications.

**Methods.** This was a retrospective study, done in the Department of General Surgery of Mongi Slim Hospital, over a period of 11 years (from January 2000 to December 2013) and collecting 115 patients. Judging criteria were specific morbidity, length of postoperative stay and recurrence.

**Results.** The median age was 43 years (62 women and 53 men). The clinic consisted of typical cholangitis episodes (6.95%), 6.95% of patients were asymptomatic.

50 radical interventions and 65 conservative interventions were performed, supplemented by treatment of the fistula as follows: Fistula blindness (43.47%), trans-fistulo-oddian drainage (3.4%), bipolar drainage (2.6%), PERDOMO (6.95%).

The overall mortality was zero. The overall morbidity was 27.69% with an overall specific mortality of 14.7% mainly represented by the suppuration of the CR and the external biliary fistulas. The median postoperative stay was 6 days; the recurrence rate was 1.7% with a delay of 30 months.

The predictive factors of postoperative complications in bi variance were: The size of the cyst ( $p = 0.02$ ), The location of a cyst in the hepatic dome ( $p = 0.002$ ), Surgical treatment: radical or conservative ( $p = 0.02$ ), The sex factor ( $p = 0.015$ ).

The multivariate study who found a significant association between sex only and postoperative complications.

**Conclusion.** Advances in hepatobiliary surgery and anesthesia are changing the surgical attitude in favor of radical procedures with favorable consequences in terms of morbidity, length of stay and recurrence. It is believed that the surgeon's experience is the most important factor in the success of surgical treatment. This helps to prevent complications.

**Keywords:** Hydatid cyst, Bile ducts, fistula, results

## INTRODUCTION

Hydatid cyst, or hydatid echinococcosis, is an endemic condition. It is due to the accidental development in humans of the larva of a parasitic dog worm: *ECHINOCOCCUS GRANULOSUS*.

This parasitosis can affect all organs, but the hepatic localization remains the most frequent. Its frequency can reach up to 5% of the population in highly endemic areas [1,2,3].

The opening in the biliary tract is the most frightening and the most frequent of the complications

*Corresponding author:*

Wael Ferjaoui

*E-mail:* farjaouiwael4@gmail.com

*Article History:*

Received: 25 February 2022

Accepted: 7 March 2022

[4]. This complication is found in 10 to 30% of cases [4, 5, 6,7].

Surgery is the standard treatment for open hydatid cysts in the bile ducts (KHFOVB). However, the management of FKB cystobiliary fistulas still remains a very controversial topic.

Postoperative complications of FKB are frequent, diverse and can be serious and fatal.

The aim of our work was to compare the results of the different surgical methods for the treatment of KHOVB in the short term (mortality, morbidity, length of postoperative stay) and in the long term (recurrence), to deduce the risk factors in order to better adapt our therapeutic management.

## METHODS

Our study was retrospective and descriptive. It was carried out over a period between January 1, 2000 and December 31, 2011 and relating to 115 patients operated on for KHFOVB.

The inclusion criteria chosen to conduct this study were:

Patients hospitalized in the general surgery department of Marsa for KHFOVB, regardless of age and sex, with single or multiple KHFOVB, isolated, regardless of the therapeutic method used: conservative radical treatment.

The presence of a Large fistula whose size is greater than or equal to 5 mm

We calculated absolute frequencies and relative frequencies (percent) for the qualitative variables.

We calculated means, medians and standard deviations and determined the extreme values for the quantitative variables.

For the quantitative variables: the comparison of two means on independent series of the two variables was carried out by means of the Student's t-test and in case of invalidity of this test by the nonparametric test of Mann-Whitney.

For the qualitative variables: The comparison of percentages on independent series was carried out by Pearson's Chi-square test and Fisher's two-tailed test.

In order to identify the risk factors independently linked to the event, we carried out a multivariate analysis in Cox regression, descending step by step method (in the first step, we introduce all the factors including the "p" are <0.05 in bivariate, and step by step we remove the factor which has the least significant "p"). The multivariate analysis made it possible to calculate adjusted relative risks, measuring the specific role of each factor.

For all statistical tests, the significance level was set at 0.05.

## RESULTS

In this work, we reported 115 cases of Hydatid cysts opening in the biliary ducts out of 430 cases of hydatid cysts operated on in the general surgery department of MongiSlim hospital in La Marsa, i.e. 26.7%, over a period of 11 years between 2000 and 2011. Our patients were divided into 62 women and 53 men with a sex ratio F/M of 1.16. The median age of our patients was 43 years

Ultrasound allowed the location of 130 cysts. The computed Tomography (CT) scan allows a better analysis of the vascular and biliary relationships of the cyst.

The most widely used technique has been resection of the protruding dome. It was performed in 65 patients (56.5%). Total perikystectomy was performed in 42 patients, i.e. 36.5%. Liver resection was performed in 8 cases. This resection consisted of a right hepatectomy in 2 cases and a left lobectomy in 6 cases. Cholecystectomy was performed systematically 110 times in our series, i.e. 95.6%. The others, 5 were already cholecystectomized.

Suturing of the fistula into healthy tissue was performed in 50 patients after radical treatment.

In the case of conservative treatment, our attitude with respect to the fistula has been summarized in the table above:

Technique	Effective	%
Simple suture after intra-lamellar perikystectomy	50	43.47
Trans-hepatic-cystic choledocostomy according to PERDOMO	8	6.95
trans-fistulo-oddian drainage	4	3.4
Bipolar drainage	3	2.6

The length of stay for our patients varied from 3 to 15 days with an average of 6 days. The postoperative effects were simple in 80.8% of patients.

The rate of specific complications was 14.7%.

Suppuration of CR was found in 8 patients. Seven patients were treated with antibiotics with prolonged drainage. Only one patient underwent CT-guided percutaneous drainage.

In our series, we observed 4 FBE exteriorized by drainage. All the fistulas ended up spontaneously drying up after prolonged intracavitary drainage.

Cholera peritoneum: was observed in 2 patients requiring revision surgery, peritoneal cleansing and establishment of effective drainage.

Two patients presented a residual hydatid obstruction which required endoscopic treatment.

One patient presented with right subphrenic suppuration requiring surgical revision for flattening and drainage.

A large right pleural effusion was found in a case requiring thoracic drainage.

A wall abscess was noted in two cases, requiring flattening and antibiotic therapy. Fixed evisceration was noted in 1 patient.

No deaths were reported in our study. The recurrence rate was 1.7% with a mean delay of 30 months

The comparison of patients who had a radical procedure and those who had a conservative procedure showed that the length of postoperative stay was long in the conservative treatment group.

Morbidity and specific morbidity were significantly higher in the conservative treatment group.

In order to identify the prognostic factors predicting morbidity in our patients, we divided our population into two groups: group with simple postoperative consequences and group with complicated post-operative outcomes. The predictive factors tested are divided into preoperative and intraoperative factors.

We carried out a bivariate analysis then a multivariate analysis.

We compared the preoperative and intraoperative variables for the two groups "simple postoperative = 93" and "complicated postoperative = 22". The gender factor was significantly associated with postoperative morbidity. The intraoperative variables significantly associated with postoperative morbidity were: The size of the cyst ( $p = 0.02$ ), The location of the cyst in the dome ( $p = 0.002$ ), Surgical treatment: radical or conservative ( $p = 0.02$ )

In multivariate analysis, only sex was retained as an independent predictor of postoperative morbidity ( $p = 0.018$ ).

## DISCUSSION

The opening in the VB is the main complication of hydatid cyst [8,9]. Its frequency in the literature varied from 10 to 30% [10, 11, 12,13]. In our series, the opening frequency was 26.7%.

Thus, the surgical treatment is based on several techniques, conservative or radical, the indications of which are debated and depend on the location and the number of cysts, but also on the technical platforms available and on the competence of the surgeon who takes care of them.

Thus, two types of methods are proposed: Conservative Methods and Radical Methods. These methods are attractive because they both solve the problem of residual cavity and fistula. They require a perfect knowledge of the surgical anatomy of the liver. Although the risk of haemorrhage is significant, they have become increasingly recommended thanks to developments in liver surgery and resuscitation methods [14,15,16,17]. In our series 43.47% of our patients who had received radical treatment. Most authors such as Tagliacozz et al [18] and Motie et al [19] reported a shorter length of stay after radi-

cal treatment than after conservative treatment. This difference was related to a longer drainage and a more frequent occurrence of postoperative complications in patients treated by conservative methods [20]. In our series, the length of stay was shorter after radical treatment than after conservative treatment. The difference was significant ( $p = 0.03$ ).

The postoperative mortality rate varied in the literature from 2 to 5% [21,22,23,24,25,26]. It varies from one author to another and depending on the treatment chosen.

In our series, there were no deaths.

Several series have compared the overall morbidity rate according to the procedure performed (conservative or radical)

In the literature, the morbidity rate varied from series to series. Thus, most authors concluded that conservative treatment was associated with a high morbidity rate.

In a prospective study published in 2002 comparing 102 patients operated on by radical techniques to 250 patients operated on by conservative techniques, Tasev et al [27] conclude that radical surgical procedures are associated with lower postoperative morbidity rates.

In our study, we had a morbidity rate of 27.69% after conservative surgery. While this rate was 8% after radical treatment. The difference was statistically significant ( $p = 0.03$ )

The incidence of non-specific medical complications ranged from 7% to 20.8% in different series in the literature [28,29]. These complications were of variable severity, and were mainly represented by respiratory complications (infectious lung disease, lobar or segmental atelectasis, infectious or reactive inflammatory pleurisy), urinary tract infections and thromboembolic complications.

In our series, the incidence of medical complications was 1.7% and represented by one case of myocardial infarction and one case of pleural effusion.

Suppuration on the residual cavity, a serious complication mainly linked to the persistence of a residual cavity, is the prerogative of conservative surgery [30,31]. In the different series of conservative treatment of KHFOVB, the incidence of this complication was 12-26% [29].

This complication was non-existent after radical surgical treatment, since radical methods in principle remove the pericyst and therefore remove any residual cavity [30]

In our study, we had a morbidity rate of 27.69% after conservative surgery. While this rate was 8% after radical treatment. The difference was statistically significant ( $p =$

Biliary fistula poses a problem of fluid, electrolyte and protein loss as well as a deficiency of fat-soluble vitamins. These losses must be compensated by a suitable hydro-electrolyte and protein intake.

In our series, we had 4 cases of biliary fistula. All of these fistulas have been reported after conservative surgery. The evolution was favorable in all cases.

Suppuration of residual cavity: In the literature, the incidence of this complication ranged from 6% to 26% [22].

In our series, there were 8 cases of CR suppuration. The evolution was favorable in all cases.

Postoperative choléperitoneum is a rare complication whose incidence has ranged in the literature from 0.4% to 2.6% [29,30,31].

In our series, we reported 2 cases of cholera peritoneum treated surgically with good outcome.

Right subphrenic abscess: Several authors have described this complication in their series.

In our series, we reported only one case of subphrenic abscess (1.9%) treated with surgery.

Several published series have mentioned variable recurrence rates. These results varied depending on the treatment chosen (radical or conservative). However, in most of the series consulted, the recurrence rate was higher in conservative treatment.

In a series by CIRENEI et al [25], the recurrence rate was 12 times higher in patients who had had conservative treatment (11.2%) than in those who had radical treatment (0.9%).

In our series, the recurrence rate was 1.7%, all of which were after conservative treatment.

Few of the studies have investigated the predictors of postoperative complications. For some authors, a morbidity rate greater than 10% should require the surgical team to re-evaluate the operative technique.

In the literature, there is an estimated female predominance of 70% in most world series [18,16]. This may be linked to the activities of women who are more involved than men with livestock, dogs and agricultural work [18, 16]. In our series, we noted preferential involvement of the female sex. This is why, in the various studies carried out, none have demonstrated an association between the male sex and the risk of complications.

In our series, female sex was a predictor of morbidity in the univariate analysis ( $p = 0.015$ ) and it is the only factor that remains significant in the multivariate analysis ( $p = 0.018$ )

Age > 40 years was found in a study conducted by Bediou et al [31] as a predictor of postoperative mortality and morbidity. Indeed, patients whose age

was 41 to 60 years and  $\geq 61$  years were 2.8 and 9.4 times more likely, respectively, to develop complications than those patients aged  $\leq 40$  years. This is mainly explained by the fact that older patients had associated co-morbidities which makes their management more difficult.

In our study, we were unable to establish a causal link between age and postoperative complications.

Most authors do not take this factor into account or exclude patients with a history of hydatid cyst surgery from their studies [30].

In our study, we were unable to establish a causal link between the history of KHF surgery and postoperative complications.

In the literature, the most implicated predictor of opening hydatid cyst morbidity is the type of surgery: conservative surgery or radical surgery.

A few authors have reported a high rate of postoperative complications in patients who underwent conservative treatment compared to those who underwent complete surgical resection

In our study, we were able to demonstrate the influence of the surgical procedure on the occurrence of complications, 4 patients presented postoperative complications after radical surgery against 18 patients who underwent conservative treatment. The difference was statically significant ( $p = 0.02$ )

In the literature, the large size of the hydatid cyst was a predictor of cysto-biliary communication in several series [12,13]. The hypothesis correlates the importance of the size of the KHF with the increase in intracystic pressure, causing necrosis of the adjacent bile ducts, and therefore the opening of the cyst in these bile ducts [14]. The hydatid cyst diameter threshold predicting the presence of BKF was greater than 10 cm [14].

Our study concluded that the size of the hydatid cyst is a predictor of postoperative morbidity ( $p = 0.02$ ).

## CONCLUSION

Radical treatment of opening hydatid cysts is the method that offers the best results in terms of specific morbidity, length of postoperative stay and recurrence rate. However, this procedure must be performed in a young patient without signs of seriousness, with sufficient blood reserve and by a surgeon who is an expert in hepatic surgery.

*Conflict of interest:* none declared

*Financial support:* none declared

## REFERENCES

1. Bellaiche G, Sultan S, Hassaine Y et al. Rupture d'un kyste hydatique du foie dans les voies biliaires: intérêts diagnostique et thérapeutique de la cholangiographie rétrograde endoscopique. *Acta Endoscopica*. 1993;23(5):75-81.
2. Zaouche A, Haouet K. Traitement chirurgical des kystes hydatiques du foie. *Encycl Med Chir. (Elsevier Masson, Paris), Techniques chirurgicales, Appareil digestif*. 40-775, 2006.
3. Botezatu C, Mastalier B, Patrascu T. Hepatic hydatid cyst - diagnose and treatment algorithm. *J Med Life*. 2018;11(3):203-9.
4. Sakhri J, Ben Ali A. Le kyste hydatique du foie. *J Chir*. 2004;141:381-9.
5. Zaouche A, Haouet K, Jouini M. Management of liver hydatidcysts with large bilio cystic fistula: multicenter retrospectivestudy. *World J Surg*. 2001;25:2839.
6. Baraket O, Feki MN, Chaari M et al. Hydatid cyst open in biliary tract: therapeutic approaches. Report of 22 cases. *J Visc Surg*. 2011;148:e211–e6
7. Beyrouth MI, Kharrat M, Elleuch S. Drainage interne trans fistulo-oddien des kystes hydatiques ouverts dans les voies biliaires. *Presse Med*. 2001;30:1863–7.
8. Bouzidi A, Chehab F. Traitement chirurgical des fistules bilio kystiques d'origine hydatique: 83 cas. *J Chir*. 1997;134:114–8
9. Dziri C, Haouet K, Fingerhut A, Zaouche A. Management of cystic echinococcosis complications and dissemination: where is the evidence? *World J Surg*. 2009;33:1266–73
10. Bulbul N, Ilhan Y, Kirkil C, Yenicierioglu A, Ayten R, Cetinkaya Z. The results of surgical treatment for hepatic hydatid cysts in an endemic area. *Turk J Gastroenterol*. 2006;17(4):273-8.
11. Tounsi A, Baroudi S, Ahallat M, Housni K, Oudanane M. Place de la résection du dôme saillant dans le traitement du kyste hydatique du foie. *Médecine du Maghreb*. 1997;66:7-9.
12. Prousalidis J, Kosmidis C, Anthimidis G, Fachantidis E, Harlaftis N, Aletras H. Forty-four years experience (1963–2006) in the management of primarily infected hydatid cyst of the liver. *HPB*. 2008;10(1):18-24.
13. Castaing D, Salloum C. Technique des hépatectomies par laparotomies. *Encycl Med Chir. (Elsevier Masson, Paris), Techniques chirurgicales-Appareil digestif*. 40-763, 2011.
14. Boutallaka H, Seddik H, Loubaris K, Bouhamou F, Aomari A, Morabit S, et al. Traitement endoscopique des complications des kystes hydatiques du foie rompus dans les voies biliaires: Expérience d'un service marocain. Vol. 50. 2018.
15. Chourak M, Mjabar A, Najih M. Kystes hydatiques du foie rompus dans les voies biliaires. *Gastroenterol Clin Biol*. 2009;33:589-95.
16. Moumen M, El Alaoui M, El Mokhtari M. Les kystes hydatiques du foie. A propos de 670 cas dont 552 compliqués. *Sem Hop Paris*. 1993;69:722-5
17. Chahbani S. Les kystes hydatiques du foie ouverts dans les voies biliaires [Thèse]. Médecine: Monastir; 1997.
18. Tagliacozzo S, Miccini M, Bonapasta SA, Gregori M, Tocchi A. Surgical treatment of hydatid disease of the liver: 25 years of experience. *Am J Surg*. 2011;10:797-804
19. Motie MR, Ghaemi M, Aliakbarian M, Saremi E. Study of the Radical vs. Conservative Surgical Treatment of the Hepatic Hydatid Cyst: A 10-Year Experience. *Indian J Surg*. 2010;72(6):448–52.
20. Ghariani B. Les kystes hydatiques ouverts dans les voies biliaires [Thèse]. Médecine: Tunis; 1994.
21. Hachaichi A. Contribution au traitement chirurgical des kystes hydatiques du foie ouverts dans les voies biliaires. A propos de 84 cas [Thèse]. Médecine : Tunis ; 1985.
22. Daali M, Fakir Y, Hssaida R, Hajji A, Hda A. Les kystes hydatiques du foie rompus dans les voies biliaires. À propos de 64 cas. *Ann Chir*. 2001;126:242-5.
23. Bouzidi A. Kyste hydatique du foie. *Encycl Med Chir. (Elsevier Masson, Paris), Hépatologie*, 7-023-A-10, 1993, 16p.
24. Atli M, Kama NA, Yuksek YN. Intra biliary rupture of a hepatic hydatid cyst: Associated clinical factors and proper management. *Arch Surg*. 2001;136:1249-55.
25. Sayek I, Onat D. Diagnosis and treatment of uncomplicated hydatid cyst of the liver. *World J Surg*. 2001;25:21–7.
26. Ammann RW, Eckert J. Cestodes. Echinococcus. *Gastroenterol Clin North Am*. 1996;25:655–89
27. Tasev V, Dimitrova V, Draganov K, Bulanov D, Popadiin N, Gaïdarski R. Hepatic echinococcosis: radical or conservative surgical treatment. *Chirurgia*. 2002;58:10-3.
28. Safioleas M, Misiakos E, Manti C et al. Diagnostic evaluation and surgical management of hydatid disease of the liver. *World J Surg*. 1994;18:859–65.
29. Lewall DB, McCorkell SJ. Rupture of echinococcal cysts: Diagnosis, classification, and clinical implications. *Am J Roentgenol*. 1986;146:391-4.
30. Bonifacino A, Carino R, Caratozollo M. L'échographie dans l'hydatidose. *Med Chir Diag*. 1989;18:304.
31. Bedioui H, Ayari H, Bouslama K, Maghrebi H, Hsairi H, Jouini M, et al. Les facteurs prédictifs de récurrence du kyste hydatique du foie : l'expérience tunisienne. *Bulletin de la Société de pathologie exotique*. 1 oct 2012;105(4):265-9.