

Title:

Radical surgery in hepatic hydatidosis: analysis of results in an endemic area

Authors:

José Manuel Ramia Ángel, Alba Manuel Vázquez, Cecilia Gijón Román, Raquel Latorre Fragua, Roberto de la Plaza Llamas

DOI: 10.17235/reed.2020.6722/2019 Link: <u>PubMed (Epub ahead of print)</u>

Please cite this article as:

Ramia Ángel José Manuel, Manuel Vázquez Alba, Gijón Román Cecilia, Latorre Fragua Raquel, de la Plaza Llamas Roberto. Radical surgery in hepatic hydatidosis: analysis of results in an endemic area. Rev Esp Enferm Dig 2020. doi: 10.17235/reed.2020.6722/2019.



This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.



OR 6722 inglés

Radical surgery in hepatic hydatidosis: analysis of results in an endemic area

José Manuel Ramia, Alba Manuel-Vázquez, Cecilia Gijón Román, Raquel Latorre-Fragua and Roberto de la Plaza-Llamas

Department of Surgery. Hospital Universitario de Guadalajara. Guadalajara, Spain

Received: 12/11/2019

Accepted: 19/11/2019

Correspondence: Alba Manuel-Vázquez. Department of Surgery. Hospital Universitario

de Guadalajara. C/ Donante de Sangre, s/n. 19002 Guadalajara, Spain

e-mail: alba_manuel_vazquez@hotmail.com

ABSTRACT

Introduction: radical surgery in hepatic hydatidosis is associated with less morbidity

and recurrence than conservative surgery.

Material and methods: a retrospective observational study of patients with liver

hydatid cyst surgery was performed. Seventy-one patients with 90 cysts were included

between 2007 and 2017, and radical surgery was performed in 69.01%.

Results: there was no difference in morbidity, mortality, biliary leakage or recurrence

according to surgery. Complicated cysts were associated with a longer hospital stay

and morbidity.

Conclusion: decision-making should consider age, comorbidity, cyst characteristics and

available resources. Radical surgery should be applied whenever feasible as it achieves

better results with an adequate selection of patients.

Keywords: Echinococcosis. Liver hydatidosis. Hydatid cyst. Liver hydatid cyst. Radical

surgery. Conservative surgery. Liver resection.

INTRODUCTION



Hepatic hydatid cyst (HLC) remains endemic in countries of the Mediterranean basin, including Spain and in Oceania, North Africa, South America, the Philippines, northern China and India (1). Emigration to non-endemic areas has made it a global disease (2-4). HLC has been a notifiable disease in Spain since 1982. In 2016, 111 new cases were reported, which is less than in 2015. The highest rates were in Navarra (1.88), Castilla y León (1.14), Aragón (0.5) and Castilla-La Mancha (0.49) (5).

The therapeutic strategies for the HLC vary according to symptoms and/or activity and also according to medical infrastructure and socio-sanitary development. There are several treatment options such as observation, antiparasitic, PAIR (puncture, aspiration, injection, re-aspiration) and surgery. However, surgical treatment is the only one that eliminates the parasite (6). There are two groups of surgical techniques: radical surgery (RS), which includes hepatic resection, peri-cystectomy and total cystectomy; and conservative surgery (CS). RS is associated with a lower morbidity and recurrence than CS (6-8).

The objective of this study was to determine the rate of RS in HLC at the Hospital Universitario de Guadalajara, Spain, and compare the results obtained according to RS/CS and clinical debut.

MATERIAL AND METHODS

A retrospective observational study was performed of patients that underwent HLC surgery at the Hospital Universitario de Guadalajara, from May 2007 to December 2017. All consecutive patients with planned HLC surgery were included in the study. The surgical indications were active HLC (CE1-CE3) and complicated HLC (CE1-CE5) (12). Emergent surgery, ineligible patients and those that refused to participate in the study were excluded.

The variables collected were age, sex, nationality, clinical debut, number/size/location of the HLC, World Health Organization (WHO) classification (9), surgery and surgical technique, hospital stay and morbidity at 90 days according to Clavien-Dindo classification (10), with \geq IIIa as major complications and recurrence. Clinical debut was classified as uncomplicated if patients were asymptomatic or had mild abdominal pain and complicated if there was obstructive jaundice (OJ), septic shock, hemoptysis,



cysto-biliary communication (CBC), cysto-cutaneous fistula, biliary-bronchial or cholecysto-cysts. Radical surgery was defined as the complete excision of the cyst by total cystectomy or hepatic resection and CS was defined as a partial resection and content removal, including any non-total cystectomy and Lagrot. The RS/CS decision was made based on preoperative/intraoperative findings and RS was the initial intention whenever feasible.

Follow-up was performed via serology/abdominal ultrasonography after six months and annually for up to five years. No further revisions were performed in patients with negative serology and imaging tests, unless requested by the patient. In the case of a suspicion of recurrence, puncture and microbiological confirmation were performed.

Statistical analysis

A descriptive study was performed and quantitative variables with a normal distribution are shown as the mean and standard deviation. Median and range were used to describe non-normal variables. The statistical study was performed with SPSS. Comparisons were made between the groups of RS/CS and clinical debut, and p \leq 0.05 was considered as statistically significant.

RESULTS

Seventy-one patients with 90 HLC were included in the study and the mean age was 53.9 ± 15 years. Sixty-four patients were Spanish (90.14%) and the rest were from Romania (five), Algeria (one) and Morocco (one). The median HLC per patient was 1 (range: 1-4) and 54 patients had a single HLC (76.06%), 16 patients had two lesions (22.53%) and one patient had four. Fourteen patients had been previously intervened (19.72%). The mean size of the lesion was 8.2 ± 3 cm and the data referring to clinical debut, location and WHO classification are shown in table 1. With regard to complicated HLC, nine debuted with OJ, five with septic shock, three with cystocutaneous fistula, two with biliary-bronchial fistula, one with CBC and one with hemoptysis.

Eighty-six surgical procedures were performed as a single procedure to resolve more than one HLC in some cases. RS was performed in 49 patients (69.01%) and the surgical



techniques are shown in table 1. Some type of bile duct (BD) procedure was associated to CBC in five patients; two choledochotomies with T-tube, two hilar plate reconstructions and one hepatic-jejunostomy.

The median hospital stay was eight days (range: 4-50) and complications and biliary leakage (BL) are shown in table 1. The patient with grade C fistula required reintervention via a right hepatectomy and hepatic-jejunostomy. There were two deaths; both patients had complicated CLH with septic shock and biliary-bronchial fistula and were treated by CS. The median follow-up was 50 months (range: 2-130), three patients died during follow-up from a HLC-unrelated cause and two patients returned to their native country. The two patients with recurrence had undergone a previous surgery and presented CBC. In one case, recurrence was contralateral and was treated by PAIR. The other patient declined treatment.

Table 2 compares RS/CS and shows statistically significant differences in HLC size and sex, with no differences in morbidity, mortality, BL or recurrence. According to clinical debut (Table 3), complicated HLCs were significantly associated with a longer hospital stay and complication rate, with no differences in RS rate.

DISCUSSION

The management of HLC is a challenge, especially in non-endemic areas, as the experience of the surgical team is considered to be a success factor (1,12). Comparing surgical techniques, CS offers acceptable mortality results, with a considerable morbidity rate (1,7,8,13) and residual cavity infection as an exclusive complication (11). RS presents fewer complications and recurrence, with no differences in mortality or hospital stay (1,4,7,8,13). A recent meta-analysis concluded that the benefit of RS on morbidity is greater in studies performed after 2006 in Europe and with a follow-up of less than 36 months (4,6).

The main problem is that both techniques are difficult to compare. CS is applicable in any center, for any cyst and by surgeons with limited experience, whereas RS is not feasible in all situations and requires greater technical and hospital infrastructure (11). RS is still not generally accepted as the treatment of choice for HLC, but it is being progressively implemented due to its advantages (4,8,11). The regionalization of



patients and the awareness of the surgical team of the advantages of RS are key factors to increase the percentage of RS (1,8,11,14).

Liver resections were previously considered as an excessively aggressive surgery. However, they allow RS without cyst opening, reducing BL and recurrence, and are usually reserved for severe BD/hilar plate involvement, vascular involvement with lobar atrophy or recurrence (15). In our series, the rate of liver resection was 30.98%, although a high percentage of these resections were a left lateral sectionectomy.

The RS rate in our series was 69.01%, which was not related to the presence of complications. According to clinical debut, there were differences in morbidity and hospital stay, which supports the idea that the presence of complications should be considered when selecting treatment (4,11,14). We must highlight that the two recurrences in our study occurred in the RS group; both patients had previously undergone surgery and required reconstruction of the hilar plate, which is a risk factor for recurrence (14).

The limitations of the study include its retrospective nature and the limited number of patients, which is probably why no significant differences were found. The non-random nature of the study may have repercussions on its strength. However, it would not be ethical to perform CS if RS is feasible given the current evidence. In addition, this study was performed at a center located in an endemic area with extensive experience.

Currently, surgery is still the gold standard of HLC (8,9,14). Decision-making should include patient age/comorbidities, location and contact with biliary/vascular structures (11) and surgeon experience. The application of RS to all HLC patients whenever feasible is considered acceptable (12).

REFERENCES

- 1. Aydin U, Yazici P, Onen Z, et al. The optimal treatment of hydatid cyst of the liver: radical surgery with a significant reduced risk of recurrence. Turk J Gastroenterol 2008;19:33-9.
- 2. World Health Organization. Available from: https://www.who.int/news-room/fact-sheets/detail/echinococcosis.



- 3. Dziri C, Haouet K, Fingerhut A. Treatment of hydatid cyst of the liver: where is the evidence? World J Surg 2004;28:731-6. DOI: 10.1007/s00268-004-7516-z
- 4. Pang Q, Jin H, Man Z, et al. Radical versus conservative surgical treatment of liver hydatid cysts: a meta-analysis. Front Med 2018;12(3):350-9. DOI: 10.1007/s11684-017-0559-y
- 5. Red Nacional de Vigilancia Epidemiológica, Instituto de Salud Carlos III. Resultados de la vigilancia epidemiológica de las enfermedades transmisibles. Informe anual. Año 2016. Available from: http://gesdoc.isciii.es/gesdoccontroller?action=download&id=25/01/2019-d8ee271b6f
- 6. Gómez I, Gavara C, López-Andújar R, et al. Review of the treatment of liver hydatid cysts. World J Gastroenterol 2015;21(1):124-31. DOI: 10.3748/wjg.v21.i1.124
- 7. He YB, Yao G, Tuxun T, et al. Efficacy of radical and conservative surgery for hepatic cystic echinococcosis: a meta-analysis. Int J Clin Exp Med 2015;8:7039-48.
- 8. Priego P, Nuño J, López-Hervás P, et al. Hidatidosis hepática. Cirugía radical vs no radical: 22 años de experiencia. Rev Esp Enferm Dig 2008;100(2):82-5. DOI: 10.4321/S1130-01082008000200004
- 9. Brunetti E, Kern P, Vuitton DA. Writing panel for the WHO-IWGE. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. Acta Trop 2010;114:1-16. DOI: 10.1016/j.actatropica.2009.11.001
- 10. Dindo D, Demartines N, Clavien PA. Classification of surgical complications, a new proposal with evaluation in a cohort of 6336 patients and results of a survey. Ann Surg 2004;240(2):205-13. DOI: 10.1097/01.sla.0000133083.54934.ae
- 11. Ramia JM, Figueras J, De la Plaza R, et al. Cysto-biliary communication in liver hydatidosis. Langenbecks Arch Surg 2012;397(6):881-7. DOI: 10.1007/s00423-012-0926-8
- 12. Secchi MA, Pettinari R, Mercapide C, et al. Surgical management of liver hydatidosis: a multicentre series of 1412 patients. Liver Int 2010;30(1):85-93. DOI: 10.1111/j.1478-3231.2009.02116.x
- 13. Tagliacozzo S, Miccini M, Amore Bonapasta S, et al. Surgical treatment of hydatid disease of the liver: 25 years of experience. Am J Surg 2011;201:797-804. DOI: 10.1016/j.amjsurg.2010.02.011



- 14. El Malki HO, El Mejdoubi Y, Souadka A, et al. Predictive factors of deep abdominal complications after operation for hydatid cyst of the liver: 15 years of experience with 672 patients. J Am Coll Surg 2008;206:629-37. DOI: 10.1016/j.jamcollsurg.2007.11.012
- 15. Ramia JM, Serrablo A, Serradilla M, et al. Major hepatectomies in liver cystic echinococcosis: a bi-centric experience. Retrospective cohort study. Int J Surg 2018;54:182-6. DOI: 10.1016/j.ijsu.2018.04.049



Table 1. Clinical presentation, location, size and WHO classification of the cysts (12), surgical technique, global complications after 90 days and biliary fistula of the liver hydatid cysts cohort intervened in the Hospital Universitario de Guadalajara, Spain, from 2007 to 2017

Series				
Patients (n)	71			
Cysts (n)	90			
Presentation				
Complication	21 (29.58%)			
Location				
Right liver	33 (46.48%)			
Left liver	30 (42.25%)			
Bilobar	8 (11.27%)			
WHO classification (6)				
CE 1	10 (14.08%)			
CE2	24 (33.80%)			
CE3	33 (46.48%)			
CE4	4 (5.63%)			
CE5	0			
Type of surgery				
Radical surgery	49 (69.01%)			
Conservative surgery	18 (25.35%)			
RS-CS	4 (5.63%)			
Surgical technique				
Total cystectomy	41			
Subtotal cystectomy	20			
Left lateral sectionectomy	13			
Left hepatectomy	7			
Lagrot and epiploplasty	3			
Right hepatectomy	2			
	J			

Complications (90 days)	27 (38.03%)
Clavien < IIIa	12
Clavien ≥ IIIa	15
Illa	12
IIIb	1
V	2
Biliary leakage	13 (18.30%)
А	3
В	9
С	1



Table 2. Distribution by type of surgery (radical surgery *vs* conservative surgery) of the liver hydatid cysts cohort intervened in the Hospital Universitario de Guadalajara, Spain, from 2007 to 2017

	Radical surgery	Conservative surgery	p			
Patients (n)	49	22				
Cysts (n)	61	30				
Women	25 (51.02%)	4 (18.18%)	p < 0.05			
Presentation	Presentation					
Complicated	12 (24.49%)	9 (40.91%)	p			
			0.161			
Size (cm)	7 ± 3.5	10 ± 3.8	p <			
			0.05			
Complications						
Global	18 (36.73%)	10 (45.45%)	p			
			0.487			
Mortality	0 (0%)	2 (9.09%)	p			
			0.093			
Biliary leakage	8 (16.33%)	4 (18.18%)	p			
			0.985			
Recurrence	2 (4.08%)	0 (0%)	p 1.00			



Table 3. Distribution according to complications in the clinical presentation of the liver hydatid cysts cohort intervened in the Hospital Universitario de Guadalajara, Spain, from 2007 to 2017

	Uncomplicated cysts	Complicated cysts	р
Patients (n)	50	21	
Location			
Right liver	24 (48.00%)	9 (42.86%)	p 0.849
Type of surgery		A. 1	
Radical surgery	36 (72.00%)	14 (66.67%)	p 0.161
Complications			
Global	13 (26.00%)	14 (66.67%)	p < 0.05
Mortality	0 (0%)	2 (9.52%)	p 0.085
Biliary leakage	6 (12.00%)	6 (28.57%)	p 0.147
Hospital stay (days)	8.9 (4-36)	16.4 (4-50)	p < 0.05
Recurrence	1 (2%)	1 (4.76%)	p 0.521