Surgical Alternatives Used in the Treatment of Liver Hydatid Cyst. A Systematized Approach Based on Evidence (an Overview)

Alternativas Quirúrgicas Utilizadas en el Tratamiento del Quiste Hidatídico Hepático. Revisión Global de la Evidencia

Carlos Manterola*** & Tamara Otzen****


SUMMARY: Echinococcosis is an endemic zoonosis in the south of Chile; we therefore have occasion to treat a large number of patients, particularly in the liver. Hepatic echinococcosis (HE) has its own morbidity and mortality due to evolutionary complications, to which the risk of complications related to the surgical procedures is added, the morbidity which has been reported up to 80 %. This is associated with a history of previous surgeries for HE, evolutionary complications of the cyst, the need for additional procedures such as the treatment of the disease in other simultaneous locations, etc. Moreover, reported mortality is up to 10 %, a situation that remains unchanged despite technological and therapeutic advances. The surgical treatment of HE can be divided into four phases: isolation of the surgical area, evacuation of the cyst, treatment of the complications of the cyst and treatment of the residual cavity. HE surgical procedures can be classified as conservative (marsupialization, cystostomy, Posadas technique and cystojejunostomy) and radical (pericystectomy and hepatic resections). Finally, the role of laparoscopic surgery, which is still under evaluation, is also worthy of note. The aim of this article is to present a general evidence-based overview of some surgical aspects of interest in the treatment of HE. In this article issues of the different surgical options utilized for HE treatment and their results are discussed, based on published evidence.

KEY WORDS: Echinococcosis; Hydatidosis; Echinococcosis; Hepatic Echinococcosis; Hepatic/surgery]; Hepatic Hydatid Cyst; Pericystectomy; Hepatic resections; Capitonnage; Omentoplasty.

INTRODUCTION

Echinococcosis is an endemic zoonosis in Chile (Grosso et al., 2012), and particularly in the Region of the Araucanía, which reports high prevalence rates that have persisted unchanged in recent years (Cortés & Valle, 2010; Martínez 2014).

The liver is the most frequently affected (77 % to 84 %), as this organ is the first filter of the infecting protoscolices (Eckert & Deplazes, 2004). Hepatic echinococcosis (HE) has its own morbidity, which is associated with evolutionary complications of the disease, and secondary morbidity related to therapeutic procedures (Eckert & Deplazes; Manterola et al., 2010a). Postoperative morbidity (POM) persists in high numbers (11 % to 86 %) despite technical advances and perioperative support. This can be medical (alteration in respiratory, cardiovascular or renal functions, etc.) or surgical, such as complications in the access (development of seromas, infection of the surgical site, evisceration and eversion), in the surgical area (development of biliary fistula and residual cavity), septic phenomena (intra-abdominal abscesses, infection of residual cavity, respiratory or urinary infection), and in vicinity, such as ipsilateral pleural effusions and postoperative ileus (evidence type 3a, 4 and 5) (Martínez; Eckert & Deplazes; Manterola et al., 2010a; Manterola et al., 2014; Manterola et al., 2015a).

It has been verified that POM in HE surgery is associated with age, cyst location, type of surgery (open vs. laparoscopic surgery and conservative vs. radical or resective techniques), previous history of HE surgery, the presence of...
evolutionary complications of the cyst, additional surgery that may be required and concomitant treatment of other locations with this disease (evidence type 2a, 3 and 4) (Manterola et al., 2014; Mamarajabov et al., 2011; Tagliacozzo et al., 2011; Agayev & Agayev, 2008; Gourgiotis et al., 2007; Daradkeh et al., 2007; Manterola et al., 2005; Priego et al., 2008). This is not particularly low, because numbers up to 79.9 % have been reported (evidence type 2a and 4) (Tagliacozzo et al.; Agayev & Agayev; Gourgiotis et al.; Daradkeh et al.; He et al., 2015). Additionally, operative mortality figures reach up to 9.2 % in the literature, and recurrence rates up to 30.4 % (evidence type 4) (Tagliacozzo et al.).

In the 19th century, when a laparotomy represented high risks of morbidity and mortality, the treatment for HE was confined to the simple puncture and evacuation of the lesion(s), with or without injection of parasiticides, such as mercury salts, carbolic acid or sublimate. These methods were abandoned due to the high incidence of subsequent complications, like hemorrhaging and intra-abdominal infection, bile duct stricture and even sudden death. The surgical era of HE treatment began at the end of 19th century, when Kirschner (1879) described what is currently known as marsupialization, and Knowsley (1883) described the technique that would later be popularized by Posadas (Manterola et al., 2009a).

GENERALITIES OF SURGICAL TREATMENT

HE surgery can be divided schematically into four steps. The first corresponds to the isolation of the surgical field, the second to evacuation of the cyst (hydatid fluid, germinative membrane and daughter vesicles), the third to the treatment of biliary communications or other alterations caused by the parasite in the host, and the fourth to the treatment of the residual cavity (Manterola et al., 2009a).

1. First step

There is type 4 and 5 evidence regarding the difficult procedure of isolating the surgical field with compresses soaked in scolicides such as hydrogen peroxide, hypertonic saline, povidone-iodine or others. An additional maneuver, is the puncture and injection of the scolicides to the inside of the cyst, which has definitely been discarded due to the lack of supporting evidence (in in vitro observations, it has verified that the time of action required by the solutions of 50 % dextrose and 20 % NaCl are 30 and 45 min respectively to eliminate all protoscolices in a sample) (Caglar et al., 2008). With respect to the rigorous isolation of the surgical area, it is important to note that this is an increasingly controversial issue, because surgeons who work on this topic are no longer convinced of its use. It is likely, that this surgical procedure will soon be practically excluded due to the evidence with respect to cyst viability (evidence type 2b) (Manterola et al., 2006). As such, the concept of simple protection of the area without the use of scolicides to soak the compresses is maintained. At the moment, however, it continues to be a normal step in HE surgery, regardless of the surgical procedure used (evidence type 5) (Manterola et al., 2009a).

2. Second step

Evacuation of the cyst by aspiration of the fluid and exeresis of the germinal layer and daughter vesicles is the only point on which all surgeons agree about the need and method; therefore, it seems superfluous to discuss. Suffice to say that this phase is included in all existing surgical techniques (evidence type 4 y 5) (Mamarajabov et al.; Manterola et al., 2009a; Buttschoen & Carli Buttenschoen, 2003; Goksov et al., 2008).

3. Third step

Treatment of the alterations caused by the parasite in the host is a controversial and probably multifactorial issue. This stage does not apply in all cases, but rather only in those where one of the following situations exists:

Frank intrabiliary rupture: Cyst-biliary communication is a frequent evolutionary complication, the prevalence of which is around 26 % of patients with HE (Manterola et al., 2009b). In these cases, there is uncertainty based on a different type of evidence, which endorses simple suturing of these ruptures or the need to perform a decompression choledochostomy that is either isolated or associated with the suturing (evidence type 1b and 4) (Manterola et al., 2009b; Ramia et al., 2013; Shalayiadang et al., 2014). We are inclined to use absorbable sutures and not perform the choledochostomy, because the procedure is not without morbidity, and leaves the patient with an ostomy for a considerable period of time; therefore, this is indicated only where there is certainty of cholangiohydatidosis with or without secondary acute cholangitis, or when a coexisting choledocholithiasis is diagnosed (evidence type 1b) (Manterola et al., 2009b; Manterola et al., 2010b).

Cholangiohydatidosis: As already mentioned, cyst-biliary communication is frequent (Manterola et al, 2009b; Manterola et al., 2010b; Averininos et al., 2006); nevertheless, it is very different when, in addition to this rupture, a migration of parasitic elements occurs inside the bile duct, which causes biliary obstructive syndrome, which we call cholangiohydatidosis; this may or may not be accompanied...
by acute secondary cholangitis (Manterola & Otzen, 2016). Cholangiohydrodatisosis is a rare evolutionary complication of HE, and this may be the reason for few reports in the literature (evidence type 4), therefore, the paucity of reports collaborates with an adequate knowledge of natural history, clinical characteristics, evolution and therapeutic alternatives of this entity.

**Liver abscess of hydatid origin:** This also constitutes a frequent evolutionary complication, the reported prevalence of which is around 25% of patients with HE. Its pathogeny is not clear, because in addition to contamination through biliary communications, contamination by hematogenous dissemination has been suggested. The clinical manifestations are non-specific, and are associated with a larval evolution over prolonged periods, which appears abruptly as a sepsis of biliary origin, at times indistinguishable from an acute cholangitis (evidence type 3a and 4) (Buttenschoen & Carli Buttenschoen, 2003; Manterola et al., 2003a; Manterola et al., 2015b).

**Thoracic involvement of HE:** This is a less frequent evolutionary complication, with an estimated prevalence between 2% and 11% (Gastaca et al., 2015; Kilani et al., 2001). We intervene abdominally and once the liver has been mobilized, we treat the cyst. Depending on the degree of advancement, we perform a simple phrenoplasty with or without a right pleurectomy, and occasionally a phrenoplasty with prosthetic material (Manterola et al., 2009c). Nevertheless, controversy remains in terms of whether access should be abdominal or thoracic (evidence type 4).

**Cystic rupture into the peritoneal cavity:** When a hydatid cyst ruptures into the abdominal cavity, with subsequent hydatid peritonitis. After initial resuscitation in cases of anaphylaxis, an arduous cleaning of the peritoneal cavity with a scolicide (for example diluted povidone) and treatment of the complicated cyst and possible coexisting lesions must be performed as soon as possible in a single surgery. This reduces the likelihood of hydatid seeding with subsequent presentation of peritoneal or pelvic hydatidosis or in some other abdominal extra-hepatic location (evidence type 4 and 5) (Manterola et al., 2003b; Yilmaz et al., 2012).

**4. Fourth step**

Finally, treatment of the residual cavity may be the most controversial phase. The evidence that endorses the different options is inconsistent; therefore, it cannot be guided by strict norms, but rather must be adapted to each individual case. Generally, the options to consider could be schematized as:

**Opening to the exterior or to a bowel loop:** This alternative is used when marsupialization (Fig. 1a) and cystostomy (Fig. 1b) are performed, and when a Roux-en-Y cystojejunostomy is performed (Fig. 1c) (evidence type 4 and 5) (Gourgiotis et al., 2007; Manterola et al., 2009a; Acar et al., 2014).

**Capitonnage:** This procedure, which term is derived from the French meaning “to quilt”, consists of folding the residual cavity by means of a series of loose absorbable sutures between one end of the pericyst and the other, thus enabling the progressive reduction of the cavity, which is sometimes obliterated (Fig. 2a). There is evidence that indicates it is associated with greater POM than the use of omentoplasty (Tagliacozzo et al.); however, a more recent study has concluded that it generates lower POM than omentoplasty (evidence type 4 and 2b) (Manterola et al., 2013).

**Omentoplasty:** In this case, a greater omentum flap is prepared that is sufficiently voluminous and vascularized to fill the cavity; and then it is fixed to the residual pericyst with loose absorbable sutures (Fig. 2b). As mentioned previously, its association with the development of POM is controversial compared with capitonnage (evidence type 2b) (Manterola et al., 2013; Muftuoglu et al., 2005).
Abstention from action: On some occasions, according to the technique used (pericystectomy and hepatic resections), it is possible to choose to leave the residual pericyst untouched, especially when it is very small and does not constitute a cavity, or is located on the underside of the liver where the force of gravity will act in favor of spontaneous drainage into the peritoneal cavity (Manterola et al., 2009a).

Resection: When there is a complete resection of the cyst, either by total pericystectomy or, through a ruled or atypical liver resection, it is evident that no residual cavity will remain. This is possibly the best option, unless the patient presents some clinical or evolutionary criterion of the disease indicating that a less aggressive approach is advisable. Our objective is to avoid leaving a residual cavity, implementing either a total or partial pericystectomy, with a remaining small piece of pericyst that does not require treatment. When, in spite of everything, a cavity remains, the decision can be made to perform a capittonnage or omentoplasty depending on location and shape of the cavity (evidence type 2b, 4 and 5) (Manterola et al., 2009a; Manterola et al., 2013; Muftuoglu et al.; Kapan et al., 2006; Sekulic et al., 2011).

CONSERVATIVE SURGICAL PROCEDURES

Procedures are called conservative only when the cyst is treated without resecting either the pericyst or surrounding hepatic parenchyma. These procedures have gradually been abandoned; some due to the cumbersome nature of the technique and others due to high POM index involved (evidence type 4) (Tagliaezzo et al.; Daradkeh et al.; Hofstetter et al., 2004; Arikan et al., 2007; Reza Mousavi et al., 2005; Utkan et al., 2001; Engin et al., 2010). These include:

1. Marsupialization

This consists of evacuation, cystostomy, extirpation of the germinal layer and hydatid material, and then communication of the residual cavity to the exterior by fixing the pericyst to the abdominal wall. The cavity is drained with gauzes that are exteriorized (Fig. 1a). There is little evidence to support its current use (evidence type 4) (Hofstetter et al.; Safioleas et al., 2006).

2. Cystostomy

After aspiration, cystostomy and extirpation of the germinal layer, a drain is installed in the residual cavity that is exteriorized through a counter opening (Fig. 1b) (Agayev & Agayev; Gourgiotis et al.; Arikan et al.; Reza Mousavi et al.; Utkan et al.). In cases where the cavity of the cyst cannot be put in contact with the abdominal wall, it is possible to resort to covering the tube with the greater omentum to avoid intraperitoneal effusion of the fluid. This technique is just a variety of marsupialization because a permanent course is created between the residual cavity and the exterior, with the only difference being that this has a smaller diameter. It is a simple treatment alternative; however, it presents the disadvantage of a torpid and prolonged evolution, with chronic suppuration through the drainage tube, and then (once this is removed) even through the orifice that this leaves (Gourgiotis et al.). A variant of this technique is called “bipolar drainage” (Manterola et al., 2009a), a cystostomy, to which a cholecustomy is added in order to decompress the main biliary tract. This variant can be of use on certain occasions; nevertheless, the disadvantages previously described are similar and are associated with the morbidity and risks typical of a cholecustomy. The use of these alternatives is endorsed only by study of evidence level type 4 (Agayev & Agayev; Gourgiotis et al.; Hofstetter et al.; Arikan et al.; Reza Mousavi et al.; Utkan et al.; Engin et al.).

Posadas technique: This is a cystostomy, evacuation of the cyst, extirpation of the germinal layer, suture of biliary communications, and then closing of the cyst cavity (Fig. 3a). It is simple, fast and effective. Its main indication is small and peripheral cyst, not complicated or calcified. Its main disadvantage is the risk of infection of the persistent cavity; this is why at the beginning of the 20th century, and simultaneously, Llobet & Varsi described a modification to this technique consisting of fixation of the pericyst to the abdominal wall, leaving some stitches outside the skin (Fig. 3b) to be able to access the cavity through direct puncture in order to drain any possible intracavitary collections, or if the situation warrants, to perform a cystostomy later on (evidence type 5) (Manterola et al., 2009a).
Cystojejunostomy: Proposed by Pegulio & Pellisier in 1959, this consists of communication of the cyst cavity with a defunctionalized jejunal loop (Fig. 1c). It is a difficult technique which is not useful and has been progressively carried out less in practice due to the number of complications. The biggest drawbacks are the inadequate drainage of the residual cavity, anastomotic stenosis and alteration in intestinal function from the manipulation of the normal intestinal transit (evidence type 4) (Acar et al.; Sahin et al., 2010).

Unroofing: Surgery that began to report in 1980. It consists of a cystotomy, evacuation of the cyst and extirpation of the germinal layer, leaving the residual cavity intact. It is a simple technique, and thus within the reach of any surgeon, and indicated essentially for small and peripheral cysts. The evidence to support it is limited (evidence type 4) (Tekin et al., 2008).

RADICAL SURGICAL PROCEDURES

These are so-called because they affect exeresis of the cyst, pericyst and a larger or smaller fragment of surrounding hepatic parenchyma. They require certain experience of the surgical team, but their results in terms of POM are superior to those reported with the use of conservative surgical techniques (evidence type 2b, 3a) (Priego et al.; He et al.; Yüksel et al., 2008). They include:

Total pericystectomy: Described by Bourgeois in 1961 (Manterola et al., 2009a), it has also been called a cystopericystectomy. It consists of the total extirpation of the parasite and pericyst through a cleavage plane with the healthy hepatic parenchyma. This resection can be complete, leaving only the hepatic parenchyma exposed (total pericystectomy Figure 4a, or incomplete, leaving a piece of residual pericyst, which is generally related to the hepatic pedicle or perihilar vessels (partial pericystectomy). The exposed hepatic parenchyma, prior to hemostasis and closing of any possible biliary communications, can be left intact or covered by means of omentoplasty. It is one of the surgical procedures of choice (evidence type 3a and 4) (Priego et al.; He et al.; Sekulic et al.; Reza Mousavi et al.; Elhamel, 1990; Botrugno et al., 2010).

Partial pericystectomy: Variation on the total pericystectomy. This consists of the evacuation of the cyst, cystotomy, extirpation of the germinal layer, suture of biliary communications, and almost total resection of the pericyst, leaving only a part, which is usually found in relation to the hepatic pedicle, suprahepatic vessels, or the vena cava (Fig. 4b). A section of the pericyst can be carried out with surrounding hepatic tissue, the amount of which will vary according to the location of the cyst and its depth in the liver (evidence type 1b and 4) (Manterola et al., 2010a, 2010b, 2013, 2015a; Priego et al.). If the residual cavity remains, according to its location, it can be treated with capitonnage or omentoplasty (Manterola et al., 2013; Muftuoglu et al.) (Figs. 2a and 2b).

Hepatectomies: These can be divided into typical major and minor resections and atypical resections; understanding atypical resections to be when the resection of a hepatic tissue fragment is performed that does not correlate with its vasculature or precise anatomical limits (generally resections that include all or part of the lesion). The indication of resection for the treatment of persistent residual cavity and multiple HE is accepted, particularly when this is located in...
the left lobe. There is no evidence to support the practice of systematic lobectomies or hepatectomies due to the great number of parenchyma that must be resected, unless it has disappeared because of the progressive development of the parasite, a situation in which the denomination “hepatic resection” does not seem justified unless we give the parasite credit for the resection. In addition, and although there is evidence of the regenerative capacity of the liver, it must be remembered that subjects who have undergone surgery mainly return home where, despite education, the risk of a new parasitization exists, which is not rare and also raises doubts about the real utility of major resections for a benign disease (evidence type 4) (Priego et al.; Nari et al., 2014; Birnbbaum et al., 2012; Martel et al., 2014). Various access routes have been reported in the literature and range from simple laparotomies such as supraumbilical midline and subcostal to more complex procedures like bilateral subcostal, midline with bilateral or single transverse extension (“in square” or “in inverted T”) and J-laparotomy, which is the one we routinely use, which involves the general mobilization of the liver, release of the adhesions of the cyst to the parietal peritoneum, diaphragm and abdominal viscera (evidence type 4) (Manterola, 2014).

Additional surgeries: It is frequently necessary to employ other surgical procedures. Cholecystectomy is used when the gallbladder is found to be forming part of the wall of the cyst, when its proximity to the cyst prevents an adequate resection of the pericyst, or for coexistent cholelithiasis (evidence type 1b and 4) (Manterola et al., 2009b, 2010a, 2010b, 2014, 2015a; Manterola & Otzen), or exeresis of other abdominal hydatid lesions (peritoneal, mesenteric, etc.) in patients with extraparenchymal parasitization, which is 14% of cases (evidence type 4) (Manterola et al., 2003b; Silva et al., 2012).

LAPAROSCOPIC TREATMENT

The first reports of laparoscopic techniques for HE treatment were based on the evacuation of the content of the cyst by simple aspiration. A series of special cannulae were designed that enabled suction of hydatid fluid and germinative membrane. These procedures demonstrate the risk of hydatid dissemination and problems stemming from the formation of residual cavities, a conversion percentage of up to 30%, higher POM between 0% and 54%, which may be associated with inadequate patient selection, average hospitalization between 1 and 8 days, and recurrence between 0% and 11%. These data are presented in a systematic review of non-randomized studies, which summarizes the outcomes of 914 patients treated by this type of access, taken from 57 primary articles (evidence type 3a) (Tuxun et al., 2014).

Despite the potential problems that this alternative provides us, there are a series of advantages that must be taken into consideration, for instance the possibility of inspecting the residual cavity in search of biliary communications, reduction during the hospital stay and reduction in the infection of the surgical site (evidence type 4) (Manterola et al., 2002).

Finally, one systematic review of non-randomized studies regarding the treatment of complicated HE is worthy of note: it was possible to conclude that there is little evidence to support decision-making and that more studies are needed, particularly with supporting evidence, to answer the many questions remaining (evidence type 3a) (Dziri et al., 2004). Similar thinking is reflected in an editorial (evidence type 5) (Ramia & Figueras, 2010).

PUERCURE, ASPIRATION, INJECTION, REASPIRATION (PAIR)

Just a few words concerning this technique, developed since 2002 (Vuitton et al., 2002). There is a systematic review based on non-randomized studies, which concludes that PAIR seems promising, but the evidence is insufficient to support its use for treating patients with uncomplicated hepatic echinococcosis and that well-designed randomized clinical trials are needed to clarify its real effectiveness (evidence type 1a) (Nasseri-Moghaddam et al., 2011).

Previously stated facts along with brief follow-up time of most studies, indicate that recommendations implemented by various groups have been downgraded in recent years. Indeed, it is suggested that although beneficial in some cases, it is indicated for only the more favorable cases of type I and II cysts, according to Gharbi’s classification (Gupta et al., 2011).
cambios a pesar de los avances tecnológicos y terapéuticos. El tratamiento quirúrgico de la EH se puede dividir en cuatro fases: aislamiento de la zona quirúrgica, evacuación del quiste, tratamiento de las complicaciones del quiste y tratamiento de la cavidad residual. Los procedimientos quirúrgicos de la EH se pueden clasificar en conservador (marsupialización, cistostomía, técnica y cistoyeyunostomía de Posadas) y radical (periquistectomía y resecciones hepáticas). Por último, el papel de la cirugía laparoscópica, que todavía está en proceso de evaluación, también es digno de mención. El objetivo de este artículo es presentar una visión general basada en la evidencia de algunos aspectos quirúrgicos de interés en el tratamiento de la EH. Se discuten los temas desde las diferentes opciones quirúrgicas utilizadas para el tratamiento de la EH y sus resultados, sobre la base de la evidencia publicada.

PALABRAS CLAVE: Equinococosis; Hidatidosis; Equinococosis hepática; Cirugía hepática; Quiste hidatídico hepático; Periquistectomía; Resecciones hepáticas; Capitonage; Omentoplastia.

REFERENCES


Correspondence to:
Dr. Carlos Manterola, MD, PhD.
Department of Surgery and CEMyQ
Universidad de La Frontera
Temuco
CHILE

Phone: 56-45-2325760
Fax: 56-45-2325761

Email: carlos.manterola@ufrontera.cl

Received: 20-02-2016
Accepted: 18-03-2016