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Huge echinococcal cyst of the liver managed by hepatectomy: Report of two cases



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ABSTRACT

INTRODUCTION: Echinococcal cysts are predominantly located in the right liver. They are usually solitary and asymptomatic, but large cysts can cause compression symptoms.

CASE REPORTS: We report two cases of huge (25 cm and 20 cm in diameter, respectively) echinococcal cysts located in the left liver, which presented as a large palpable mass causing compression symptoms. Diagnosis was established with CT scan showing a cystic mass with the characteristic daughter cysts and reactive layer (pericystic wall) consisting of fibrous connective tissue and calcifications. Both patients were treated radically with left hepatectomy and had uneventful postoperative course and no recurrence upon follow-up.

DISCUSSION: The treatment of liver echinococcal cysts represent a unique surgical challenge. Even though conservative approaches are less technically demanding, the radical approach with resection has better outcome with less recurrences, when performed by experienced surgeons.

CONCLUSION: Resection rather than drainage is the management of choice for such huge liver echinococcal cysts.

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1. Introduction

Echinococcosis or hydatidosis is a widely endemic disease in many parts of the world and predominately affects the liver. In the majority of the cases it is caused by the canine parasite *Echinococcus granulosus* tapeworm which gives rise to the most common cystic form, while the fox parasite *Echinococcus alveolaris* (multilocularis) is responsible for the rare form of the disease. Definitive host in the parasite's cycle could be the dog, wolf, or fox and intermediate host the human, sheep, cow, or pig. The disease is usually asymptomatic but occasionally presents as an acute complication due to suppuration or rupture of the cyst into the biliary tree or the peritoneal cavity. Diagnosis is established by current imaging techniques (US, CT, MRI-MRCP), aided by serum serology tests for antibodies against hydatid antigens. It is important always to include echinococcosis in the differential diagnosis of cystic lesions of the liver [1–4].

Chemotherapy with albendazole is indicated as conservative treatment mainly for small not calcified multiple cysts or as postoperative adjuvant therapy. It is also the treatment of choice in most cases of *E. alveolaris* [5].

However, surgery is the main treatment modality in the management of liver echinococcosis, especially in large solitary cysts. There is an ongoing debate regarding which procedure is optimal for the treatment of liver echinococcal cysts i.e. drainage or resection. In this manuscript we report two cases of huge echinococcal cysts located on the left liver, which were successfully managed with left hepatectomy. This is a compelling report on unusual cases, which addresses current information about management and treatment of huge liver echinococcosis.

2. Report of cases

The patients gave informed consent for publication.

Case 1 was a 42-year-old man, ASA I score, with a 25 cm left liver echinococcal cyst, causing persistent epigastric pain and discomfort.

Case 2 was a 75-year-old man, ASA II score, with a 20 cm cyst also located in the left liver. He presented with epigastric discomfort and vomiting caused by compression and complete obstruction of the gastric outlet by the cyst.

Upon clinical examination, a large palpable mass was found in the upper abdomen of both patients. The diagnosis was established by CT scan, which showed the cystic mass with the characteristic daughter cysts and clearly delineated reactive layer (pericystic wall) consisting of fibrous connective tissue and calcifications (Figs. 1 and 2).

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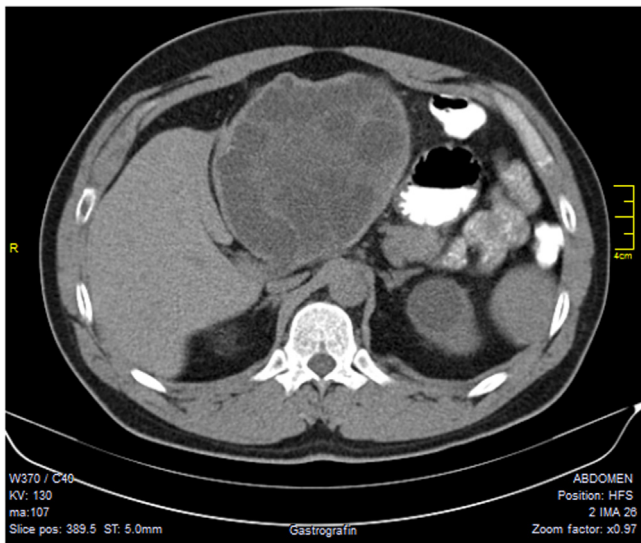


Fig. 1. CT showing the huge cyst (25 cm) with the characteristic daughter cysts and clearly delineated reactive layer (pericystic wall) in the first case.



Fig. 2. CT showing the huge cyst (20 cm) with the characteristic daughter cysts and clearly delineated reactive layer (pericystic wall) in the second case.

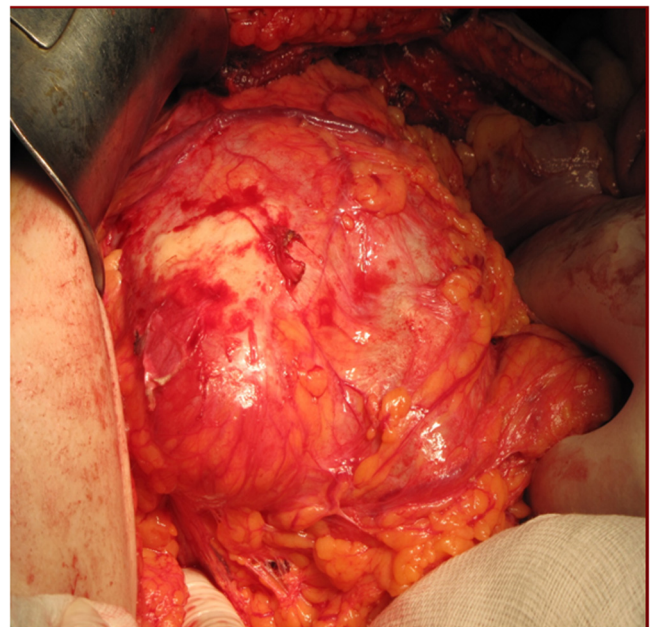


Fig. 3. Intact the resected specimen showing thickened the outer or adventitia fibrous layer of hitine in the first case.

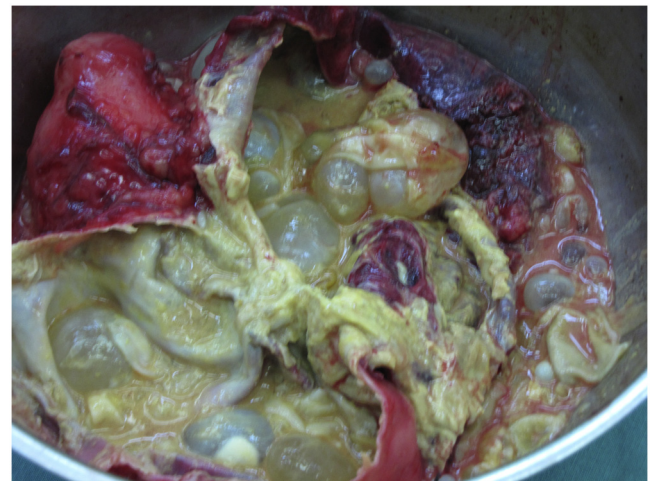


Fig. 4. Opened the huge cyst with contained daughter cysts and vesicles, hydatid debris material, the inner laminated or germinal membrane and clear water fluid in the first case.

The operative procedure was performed via a bilateral subcostal incision, followed by liver mobilization. The operative field was carefully isolated with packs enriched with hypertonic saline for prevention of dissemination of the parasite, followed by cautious dissection. Fortunately, no puncture or unwilling rupture occurred in any case.

Both patients underwent radical resection of the lesion by left hepatectomy with low blood loss (600 ml and 350 ml, respectively). Due to dense adhesions and scar formation, dissection in the first case proved more challenging. The intact resected specimen of this case is shown in Fig. 3 as well as the opened huge cyst (Fig. 4). After meticulous hemostasis a drain was left in the site of the resected cyst.

Both patients had an uneventful postoperative course and were discharged on the 6th and 8th postoperative day, respectively. 3-year follow-up for the first patient 2-year follow-up for the second patient showed no CT evidence of recurrence and normal liver func-

tion tests. One-month and seven-month postoperative CT scans from the first patient are shown in Figs. 5 and 6.

3. Discussion

Echinococcal or hydatid disease of the liver has been known since the time of Hippocrates, who described it as “liver full of water”. The disease is common in some geographical areas (Africa, Middle East, Mediterranean Sea, South America, Australia, New Zealand), but its frequency tends to be decreased with the improvement of sanitary conditions and the development of effective management procedures. The disease affects predominantly the liver (70%), mainly the right lobe and the cyst is solitary in 80%. The lung is the second most frequent location (20–25%), mainly the lower lobe. Lung cysts are usually solitary without calcification, but in 20% they can be multiple or bilateral. Rarely locations include muscle, brain, spleen, bones, pancreas, kidney, heart, mediastinum



Fig. 5. CT showing the liver remnant one month after hepatectomy and no other abnormal findings in the first case.

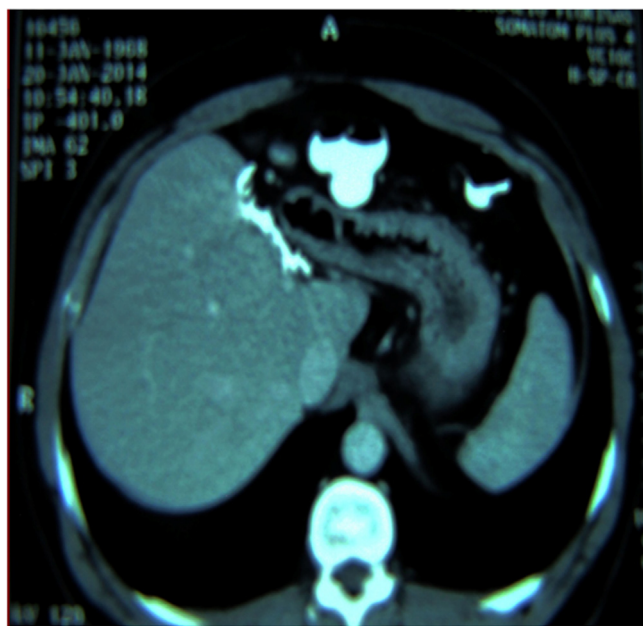


Fig. 6. CT showing the liver remnant with compensatory hyperplasia seven months after hepatectomy and no recurrence or other abnormal finding in the first case.

or diffuse intraabdominal dissemination. Hepatic echinococcal disease is usually asymptomatic and in the majority of the cases (60%) it is detected incidentally in routine imaging. Presentation depends heavily on cyst size. When cyst diameter exceeds 5 cm, abdominal pain or heaviness will possibly ensue. On physical examination, a palpable mass can be found in the right upper quadrant or epigastrium. However, a complication may occur manifested by corresponding acute appearance [3,4,6].

Surgery remains the cornerstone of the management of hepatic hydatid disease [3,6,7–9]. Care must be taken to overcome operative difficulties and achieve optimal outcome. Necessary actions include meticulous dissection and careful isolation of operative field by packing enriched with hypertonic saline solution or other

scolicidal agent. This action is important for prevention of parasite dissemination in case of accidental or intentional breaching of the cyst wall, thus avoiding the recurrence. Treatment is indicated only for viable echinococcal cysts, while degenerated or calcified cysts need not be operated.

Besides disease parameters and the patient's physical status, surgeon's personal experience constitutes a principal criterion for selection of the best operative procedure. Total pericystectomy and hepatectomy are considered as the most demanding radical procedures. Conservative methods include partial cystectomy with external drainage, omentoplasty and capitonnage (approximating sutures), are easier to perform and were popular surgical options in the past decades [10]. Whenever possible, complete resection should be the prime choice even though technical difficulties, required expertise and need for advanced instrumentation make the procedure less attractive. Radical surgical procedures are better tolerated by the patients and provide better results in terms of long-term recurrence rates [10]. It has been proposed that subadventitial cystectomy should be considered as the standard surgical treatment, since it allows complete restoration of the operated liver [11] and should be performed in centres with accumulated experience in hepato-pancreato-biliary surgery. Laparoscopic surgery was also proven to be safe and effective both in radical liver resection and subadventitial pericystectomy [12].

The PAIR procedure is a recently reported conservative treatment of liver echinococcal cysts. It refers to the therapeutic puncture of the cyst, aspiration, injection of a scolicidal agent into the cyst and re-aspiration and has been proposed as a minimally invasive alternative approach to surgery [2,4]. The medical treatment with albendazole, 400 mg per os twice daily for 28 days, followed by a cessation for 14 days, until completion of 3–6 cycles is the usual scheme. Throughout medical treatment, serum transaminase levels must be followed [4,5]. It has also been already preliminary discussed the increased susceptibility and severity of echinococcal infection in patients with HIV infection making possible an association [13].

To the best of our knowledge, only a few cases of huge liver hydatid cysts have been reported so far [14–17]. Two cases were managed by subtotal pericystectomy dealing with the residual cavity, but none by hepatectomy as in our cases. One of the reported cysts had a diameter of 18 cm occupying both parts of the liver [14] and the other of 20 cm occupying almost the entire left lobe [15]. The performed hepatectomy in our cases ensured that no cyst opening would occur. To this effect, major complications such as allergic reaction or most importantly dissemination of the disease and recurrence have been prevented. Besides, two other cases with exophytic growth originating from the right lobe of the liver have been described as well. The first case was a cyst of a diameter of 32 cm that was managed by total pericystectomy without cyst rupture [16]; the second case, a cyst of 30 cm in diameter was managed by right hepatectomy [17]. We can surmise that huge echinococcal cyst of the liver is an infrequent entity which is associated with some operative challenges. Nowadays, advances that have emerged in surgical technique, utilization of proper instrumentation and state-of-the-art technology, contribute that hepatectomy can be feasible in such difficult cases, in order to provide radicality. Partial pericystectomy and dealing with the residual cavity should be avoided as first choice procedure.

In our cases, cyst size was 25 cm and 20 cm, respectively, affecting the left liver and presenting with persistent symptoms of space occupying lesion. Characteristic CT images with daughter cysts, well-delineated thickened wall established the diagnosis. Left hepatectomy was considered a reasonable option, since it ensured radicality and adequate functional residual liver parenchyma and was absolutely justified by the excellent long-term outcome. This manuscript has been reported in line with the SCARE criteria [18].

In conclusion, resection (hepatectomy or total cystopericystectomy), rather than partial cystectomy and drainage, is currently the procedure of choice in the management of liver hydatid disease, even when the cysts are huge and especially for those located in the left liver.

Conflict of interest

Nothing to declare.

Funding

Nothing to declare.

Ethical approval

Ethical approval was not needed. This is a well-described procedure on two patients with extremely large cysts. No new intervention was performed and informed consent from the patients was obtained. Personal details of the patients were not disclosed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Authors contribution

Study concept and design: ET Pavlidis, TE Pavlidis.

Data collection: ET Pavlidis.

Data analysis: N Symeonidis, K Psarras.

Writing the paper: ET Pavlidis, TE Pavlidis.

Final revision: N Symeonidis, TE Pavlidis.

Registration of research studies

Registration number pending.

Guarantor

TE Pavlidis.

References

- [1] T. Pakala, M. Molina, G.Y. Wu, Hepatic echinococcal cysts: a review, *J. Clin. Transl. Hepatol.* 4 (2016) 39–46.
- [2] J. Richter, E. Profis, M.C. Holtfreter, A. Orhun, I. Müller-Stöver, H. Dedelen, R. Kubitz, Anaphylactic shock ensuing therapeutic puncture of an echinococcal cyst, *Parasitol. Res.* 114 (2015) 763–766.
- [3] N. Symeonidis, T. Pavlidis, M. Baltatzis, K. Ballas, K. Psarras, G. Marakis, A. Sakantamis, Complicated liver echinococcosis: 30 years of experience from an endemic area, *Scand. J. Surg.* 102 (2013) 171–177.
- [4] G. Garcea, A. Rajesh, A.R. Dennison, Surgical management of cystic lesions in the liver, *ANZ J. Surg.* 83 (2013) 516–522.
- [5] T.E. Pavlidis, P.T. Katsinelos, P.Z. Tsiaousis, K.S. Atmatzidis, Intrabiliary rupture of a large liver echinococcal cyst in an adolescent managed with endoscopic sphincterotomy and albendazole, *J. Laparoendosc. Adv. Surg. Tech. A* 16 (2006) 493–496.
- [6] D. Voros, D. Katsarelias, G. Polymeneas, A. Polydorou, L. Pistiolis, A. Kalovidouris, A. Gouliamos, Treatment of hydatid liver disease, *Surg. Infect. (Larchmt)* 8 (2007) 621–627.
- [7] S. Halezeroglu, E. Okur, M.O. Tanyü, Surgical management for hydatid disease, *Thorac. Surg. Clin.* 22 (2012) 375–385.
- [8] A. Benkabbou, A. Souadka, B. Serji, H. Hachim, R. Mohsine, L. Ifrine, A. Belkouchi, H.O. El Malki, Changing paradigms in the surgical management of cystic liver hydatidosis improve the postoperative outcomes, *Surgery* 159 (2016) 1170–1180.
- [9] I. Gomez, C. Gavara, R. López-Andújar, T. Belda Ibáñez, J.M. Ramia Ángel, Á. Moya Herraiz, F. Orbis Castellanos, E. Pareja Ibars, F. San Juan Rodríguez, Review of the treatment of liver hydatid cysts, *World J. Gastroenterol.* 21 (2015) 124–131.
- [10] G.K. Georgiou, G.D. Lianos, A. Lazaros, H.V. Harissis, A. Mangano, G. Dionigi, C. Katsios, Surgical management of hydatid liver disease, *Int. J. Surg.* 20 (2015) 118–122.
- [11] K. Mohkam, L. Belkhir, M. Wallon, B. Darnis, F. Peyron, C. Ducerf, J.F. Gigot, J.Y. Mabrut, Surgical management of liver hydatid disease: subadventitial cystectomy versus resection of the protruding dome, *World J. Surg.* 38 (2014) 2113–2121.
- [12] H. Lv, Y. Jiang, X. Peng, S. Zhang, X. Wu, H. Yang, H. Sun, Echinococcus of the liver treated with laparoscopic subadventitial pericystectomy, *Surg. Laparosc. Endosc. Percutan. Tech.* 23 (2013) e49–53.
- [13] A. Javed, R. Kalayarsan, A.K. Agarwal, Liver hydatid with HIV infection: an association, *J. Gastrointest. Surg.* 16 (2012) 1275–1277.
- [14] L. Bonfrate, F. Giuliani, G. Palasciano, J.T. Lamont, P. Portincasa, Unexpected discovery of massive liver echinococcosis. A clinical, morphological, and functional diagnosis, *Ann. Hepatol.* 12 (2013) 634–641.
- [15] N.S. Salemis, Giant hydatid liver cyst: management of residual cavity, *Ann. Hepatol.* 7 (2008) 174–176.
- [16] D.A. Sahin, R. Kusaslan, O. Sahin, O.N. Dilek, Huge hydatid cysts that arise from the liver, growing exophytically, *Can. J. Surg.* 50 (2007) 301–303.
- [17] G.M. Ettorre, G. Vennarecci, R. Santoro, A. Laurenzi, C. Ceribelli, A. Di Cintio, et al., Giant hydatid cyst of the liver with a retroperitoneal growth: a case report, *J. Med. Case Rep.* 6 (2012) 298.
- [18] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, Orgill DP and the SCARE group. the SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.

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