

Surgical Treatment of a Giant Hydatid Liver Disease Combined with Endoscopic Sphincterotomy: A Case Report

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Abstract

A 14-year-old boy was admitted with a shoulder pain and epigastric discomfort. A palpable mass was found in the abdomen upon clinical examination. Computed tomography (CT) scan showed a giant cystic mass occupying most of the right liver lobe. At laparotomy a volume of 1050 ml fluid was aspirated and 20% NaCl solution was injected into the cavity. After evacuating the cyst contents, the endocyst was totally extracted. Visible biliary orifices were sutured with 3/0 polyglycolic acid sutures and a polyglycolic acid (PGA) sheet (Neoveil, Gunze, Osaka, Japan) was applied to the surfaces of sutured biliary orifices in the endocyst. Omentoplasty and capitonage was added to the procedure. External bile leakage of 150 ml/day did not decrease during postoperative period despite medical treatment (TPN, octreotide) and an endoscopic retrograde cholangiopancreatography (ERCP) with sphincterotomy was performed on the 28th postoperative day. It dramatically decreased after sphincterotomy and eventually stopped. Choices of treatment modalities in hepatic hydatid disease (HD) are radical operations including hepatectomy and pericystectomy and conservative methods including partial cystectomy, omentoplasty and capitonage. Although hydatid cysts of 10 cm or greater in diameter are called "giant" traditionally there are no uniformly accepted criteria that define "giant". Biliary fistulas are the most common morbidity following hydatid liver surgery. Although there are reports stating that all the biliary fistulas close spontaneously after surgical treatment, they persist in 4% - 27.5% of cases. Endoscopic sphincterotomy has been proposed in biliary fistula of more than 3 weeks' duration or with bile output exceeding 300 ml/day. Nevertheless, once conservative measures are inadequate, one should not be in delay to perform ERCP and sphincterotomy. If postoperative biliary fistula develops after surgical intervention it should first be treated conservatively. If it persists then the endoscopic procedures including ERCP and sphincterotomy becomes a necessity.

Keywords: *Giant; CT; ERCP; Endoscopic Sphincterotomy*

Introduction

Hydatidosis which is caused by the larval form of *Echinococcus granulosus* is a widely endemic disease in many parts of the world. According to the Ministry of Health database, there are 14000 cases recorded from 2001 to 2005 in Turkey [1]. The liver is involved in 50% - 70% of cases [2-5]. A 14-year-old boy with a giant echinococcal cyst occupying 2/3 of the right lobe of the liver was surgically treated using polyglycolic acid sheet (PGA). Endoscopic retrograde cholangiopancreatography (ERCP) with sphincterotomy was performed for persistent bile leakage during postoperative period. Biliary fistula healed 2 days following sphincterotomy. In this report, our case is presented to address current information about management of huge liver echinococcosis in the light of relevant literature.

Case

A 14-year-old boy was admitted to our clinic with a shoulder pain and epigastric discomfort. A large palpable mass was found in the upper abdomen upon clinical examination. Ultrasonography (US) and computed tomography (CT) scan showed a giant cystic mass occupying nearly 2/3 of right liver lobe accompanying a solidified satellite cyst at the inferior margin of the right lobe (Figure 1). CT clearly delineated reactive layer (pericystic wall) containing fibrous tissue and calcifications. No other organs were involved by the disease. An

indirect hemagglutination test was used for the serological confirmation of the diagnosis. A laparotomy under general anesthesia was performed using a liberal subcostal incision. A giant cystic mass of 15 cm in diameter occupying most of the right lobe of the liver was found. The mass pushed dome of the right diaphragm upwards and liver downwards. Packs enriched with hypertonic saline (20% NaCl) for prevention of dissemination of disease were used for isolation. A volume of 1050 ml clear fluid was aspirated and scolocidal solution (20% NaCl) was injected into the cavity. After evacuating the cyst contents and cystotomy the entire endocyst comprising both the inner germinal and outer chitinous layers was totally extracted from the ectocyst of the surrounding liver parenchyme (Figure 2). Visible biliary orifices were sutured with 3/0 polyglycolic acid sutures. In order to avoid any biliary fistulas that may happen to occur during postoperative period, a polyglycolic acid (PGA) sheet (Neoveil, Gunze, Osaka, Japan) was also applied to the surfaces of sutured biliary orifices in the endocyst using fibrin glue (Figure 3). The large cavity after enucleation was filled with omentum to prevent collection of fluid and abscess formation and a capitonage was added to the procedure at the end of the surgical procedure (Figure 4). An external catheter with multiple side-holes at the tip was also inserted into the bottom of the residual cavity for drainage. Bile leakage of 150 ml/day from the external catheter did not decrease during postoperative period despite medical treatment including total parenteral nutrition, systemic octreotide usage and an ERCP with sphincterotomy was performed on the 28th postoperative day for intractable bile leakage. Bile leakage dramatically decreased after sphincterotomy and eventually stopped 2 days after ERCP and the drainage catheter was removed. The patient is now well with no symptoms of disease and receiving regular dose of 10 mg/kg of benzimidazole compounds (albendazole/mebendazole).



Figure 1: CT showing the giant cyst (15 cm) occupying most of the right lobe of the liver.



Figure 2: Operative view of opened cyst. Note the entire endocyst was totally extracted.

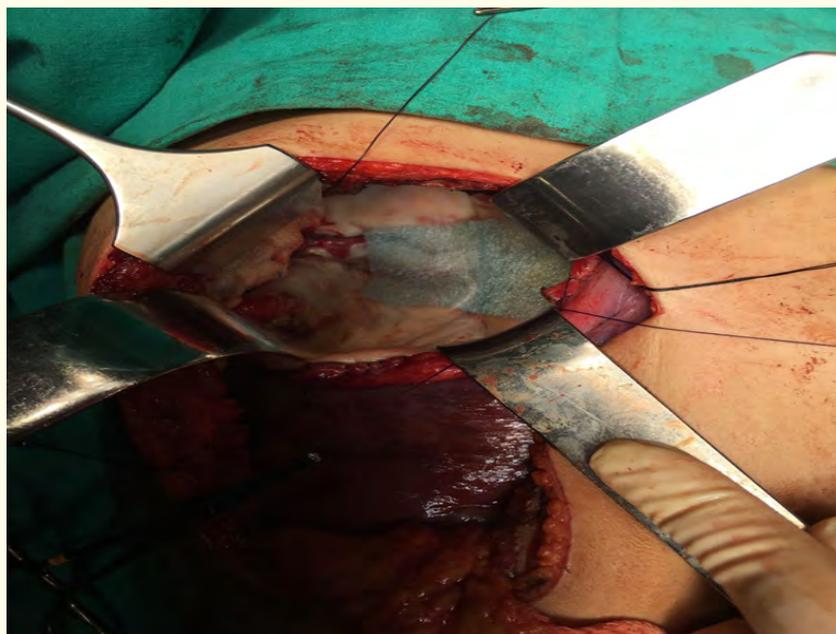


Figure 3: Operative view. Note partial cyst wall excision was performed and PGA sheet inserted into the cyst cavity.

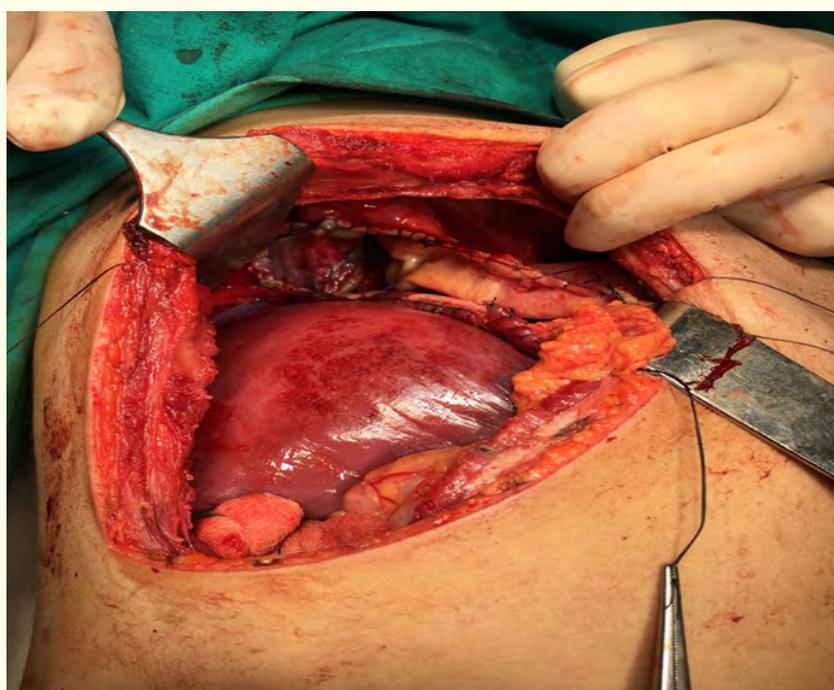


Figure 4: Operative view. The large cavity after enucleation was filled with omentum and capitonage was completed.

Discussion

Treatment of hydatid cyst has not changed much over the years and surgery remains the cornerstone of the management of hepatic hydatid disease [6-10]. Choices of treatment modalities can be divided into two basic groups: radical or conservative. Hepatectomy and pericystectomy are radical operations and carry a certain perioperative risk. But it is stated that biliary leakage and recurrence are rare in radical surgical interventions [11-13]. Conservative methods including partial cystectomy with external drainage, omentoplasty and capitonage are easier to perform but carry a high incidence of postoperative biliary leakage and recurrence [4,13]. The PAIR procedure that is also a conservative treatment modality of liver hydatid disease refers to puncture of the cyst, aspiration, injection of scolocidal agent into the cyst and reaspiration. It has been proposed as an alternative approach to surgery. Conservative surgical approach including cystotomy, evacuation of the cyst and its contents, suture ligation of biliary fistulas, omentoplasty and capitonage was performed in our case. Besides PGA was applied to the biliary leakage surfaces in the endocyst after suture ligation of the fistulas in order to prevent subsequent fistula formation.

Although hydatid cyst of 10 cm or greater in diameter are called "giant" cysts traditionally there are no uniformly accepted criteria that define "giant" liver hydatid disease [14]. Only a few cases of huge liver HD have been reported so far and these are adult patients [15-18]. Literature on this issue is more scarce in pediatric patients [19]. Presence of large cysts in children can be more dangerous than in adults as they may produce pressure effect to nearby structures and destroy the parenchyme of the developing organ. With a diameter of 15 cm, the cyst in our case was accepted to be giant and timely-aggressive surgical treatment was performed.

Biliary fistulas are the most common morbidity following hydatid liver surgery with an incidence of 13%-37% [20,21-25]. It is defined as any drain output consistent with a biliary appearance with a high bilirubin count. Intracystic pressure is 30 - 80 cmH₂O [24-26]. According to the LaPlace Law, expansion of the cyst further increases the intracystic pressure therefore flow is toward the biliary system. Once the cyst has been drained pressure dynamics reversed and bile flows into the residual cavity rather than through the biliary system via papilla of Vater [27]. Despite measures were taken for the prevention of future probable biliary leakage in our case during the surgical intervention including suture ligation and PGA application on the surface of probable leakage sites in the endocyst, biliary leakage unfortunately occurred in our patient during postoperative period.

Although there are reports stating that all the biliary fistulas close spontaneously after surgical treatment of liver HD, they persist in 4% - 27.5% of cases [28 29]. ERCP with endoscopic sphincterotomy has been proposed in biliary fistula of more than 3 weeks' duration or with output exceeding 300 ml/day but unfortunately these reports come from adult series [4,30]. Literature on this subject with regard to children is scarce [28]. It was demonstrated that a postoperative biliary drainage volume < 102 ml to be the only predictor of spontaneous closure of biliary fistula following liver HD surgery [31]. As evidenced by the literature, ERCP is a complicated procedure with high rates of complications in pediatric age. Moreover, the execution of ERCP requires highly specialized medical staff and necessary equipment, which is not easy to find in all pediatric surgery centers. It is important to select the category of patients to offer this treatment. Patients with giant HD with prolonged bile leakage unresponsive to conservative treatment modalities may be candidates of this treatment choice and should be sent to specialized centers capable of managing the possible complications as well. Nevertheless, once conservative measures to control the post-surgical biliary leakage in children with huge liver HD are inadequate, the clinician dealing with these children should not hesitate to perform ERCP and sphincterotomy.

In conclusion, management of giant liver hydatid disease may constitute a great challenge to attending surgeon. Despite preventive measures, if postoperative biliary fistula develops after surgical intervention it should first be treated by conservative methods. If the bile leakage persists then the endoscopic procedures including ERCP and sphincterotomy becomes necessity for treatment of these patients.

Conclusion

Management of giant liver hydatid disease may constitute a great challenge to attending surgeon. Despite preventive measures, if postoperative biliary fistula develops after surgical intervention it should first be treated by conservative methods. If the bile leakage persists then the endoscopic procedures including ERCP and sphincterotomy becomes necessity for treatment of these patients.

Bibliography

1. Yazar S., et al. "Cystic echinococcosis in Turkey from 2001-2005". *Türkiye Parazitoloji Dergisi* 32.3 (2008): 208-220.
2. Sayek I., et al. "Surgical treatment of hydatid disease of the liver". *Archives of Surgery* 115 (1980): 847-850.
3. Akkiz H., et al. "Endoscopic management of biliary hydatid disease". *Canadian Journal of Surgery* 39.4 (1996): 287-292.
4. Skroubis G., et al. "Significance of bile leaks complicating conservative surgery for liver hydatidosis". *World Journal of Surgery* 26.6 (2002): 704-708.
5. Al Karawi MA., et al. "Endoscopic management of biliary hydatid disease: report of six cases". *Endoscopy* 23.5 (1991): 278-281.
6. Symeonidis T., et al. "Complicated liver echinococcosis: 30 years of experience from an endemic area". *Scandinavian Journal of Surgery* 102.3 (2013): 171-177.
7. Voros D., et al. "Treatment of hydatid liver disease". *Surgical Infections (Larchmt)* 8 (2007): 621-627.
8. Halezeroglu S., et al. "Surgical management for hydatid disease". *Thoracic Surgery Clinics* 22.3 (2012): 375-385.
9. Benkabbou A., et al. "Changing paradigms in the surgical management of cystic liver hydatidosis improve the postoperative outcomes". *Surgery* 159.4 (2016): 1170-1180.
10. Gomez I., et al. "Review of the treatment of liver hydatid cysts". *World Journal of Gastroenterology* 21.1 (2015): 124-131.
11. Giordano G., et al. "Surgical treatment of hydatid cyst of the liver: pericystectomy or resection. Personal experience". *International Journal of Surgery and Surgical Sciences* 6 (1996): 113-117.
12. Moreno Gonzalez E., et al. "Results of surgical treatment of hepatic hydatidosis: current therapeutic modifications". *World Journal of Surgery* 15.2 (1991): 254-263.

13. Kayaalp C., et al. "Importance of cyst content in hydatid liver surgery". *Archives of Surgery* 137.2 (2002): 159-163.
14. Usluer O., et al. "Surgical management of pulmonary hydatid cyst". *Texas Heart Institute Journal* 37.4 (2010): 429-434.
15. Bonfrate L., et al. "Unexpected discovery of massive liver echinococcosis. A clinical, morphological and functional diagnosis". *Annals of Hepatology* 12.4 (2013): 634-641.
16. Salemis NS. "Giant hydatid liver cyst: management of residual cavity". *Annals of Hepatology* 7.2 (2003): 174-176.
17. Ettorre GM., et al. "Giant hydatid cyst of the liver with a retroperitoneal growth: a case report". *Journal of Medical Case Reports* 6 (2012): 298.
18. Sahin DA., et al. "Huge hydatid cysts that arise from the liver growing exophytically". *Canadian Journal of Surgery* 50.4 (2007): 301-303.
19. Goyal VD., et al. "Single-stage management of large pulmonary and hepatic hydatid cysts in pediatric age group: report of two cases". *Lung India* 31.3 (2014): 267-269.
20. Langer JC., et al. "Diagnosis and management of hydatid disease of the liver". *Annals of Surgery* 199.4 (1984): 412-417.
21. Kayaalp C., et al. "Distribution of hydatid cysts into the liver with reference to cystobiliary communications and cavity-related complications". *American Journal of Surgery* 185.2 (2003): 175-179.
22. Alper A., et al. "Choledocoduodenostomy for intrabiliary rupture of hydatid cysts of liver". *British Journal of Surgery* 74.4 (1987): 243-245.
23. Bedirli A., et al. "Surgical management of spontaneous intrabiliary rupture of hydatid liver cysts". *Surgery Today* 32.7 (2002): 594-597.
24. Ozaslan E and Bayraktar Y. "Endoscopic therapy in the management of hepatobiliary hydatid disease". *Journal of Clinical Gastroenterology* 35.2 (2002): 160-174.
25. Milicevic H. "Hydatid disease". In: Blumgart L, Fong Y, editors. *Surgery of the liver and biliary tract*, 2nd ed. Philadelphia: WB Saunders Company (2000): 1167-1204.
26. Yalın R., et al. "Significance of intracystic pressure in abdominal hydatid disease". *British Journal of Surgery* 79.11 (1992): 1182-1183.
27. Vignote ML., et al. "Endoscopic sphincterotomy in hepatic hydatid disease open to the biliary tree". *British Journal of Surgery* 77.1 (1990): 30-31.
28. Tiryaki HT., et al. "Is the biliary complication really seen lesser in children with hydatid liver disease?" *Turkish Journal of Pediatrics* 3.4 (2009): 34-39.
29. Dolay K., et al. "Endoscopic sphincterotomy in the management of postoperative biliary fistula. A complication of hepatic hydatid disease". *Surgical Endoscopy* 16.6 (2002): 985-988.
30. Gharbi HA., et al. "Ultrasound examination of the hydatid liver". *Radiology* 139.2 (1981): 459-463.
31. Zeybek N., et al. "Biliary fistula after treatment for hydatid disease of the liver: when to intervene". *World Journal of Gastroenterology* 19.3 (2013): 355-361.

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