

A Study of Complications of Hydatid Cyst Disease in a Tertiary Care Center in South India

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Abstract

Introduction: Hydatid cyst disease is a zoonosis caused by *Echinococcus* spp. There is sparse literature on complications of hydatid cyst disease and the tools available to treat them. This article is a study on hydatid cyst disease in a tertiary care hospital in India.

Materials and Methods: Retrospective study on the number of treated hydatid disease between August 2015 and August 2018 and further study on complicated hydatid disease and their management.

Results: A total of 34 patients treated for hydatid disease with 20 males and 14 females. Median age was 46 years. The right lobe of the liver was involved in all the patients. 10 patients had complications with five biliary complications and five vascular complications. Two patients had cholangitis on presentation which improved on endoscopic retrograde cholangiopancreatography, three patients had biliary complications, one detected intraoperatively, and two detected due to post-operative bile leak. Among the vascular complications, the cyst was in close proximity to the right hepatic vein, of which one developed intraoperative anaphylaxis, two patients had cyst in close proximity to the inferior vena cava (IVC), and one in multiple site in abdomen in thorax and in close proximity to IVC and the right subclavian artery.

Conclusion: Hydatid cyst could cause biliary and vascular complications and the treating surgeon must be aware of these complications and must possess the necessary tools to deal with them.

Key words: Biliary complications, Epidemiology, Hydatid cyst, Vascular complications

INTRODUCTION

Hydatid cyst disease is a zoonosis caused by *Echinococcus* spp. Man is accidental intermediate host. The cyst is the larval stage of the parasite. Two main species of *Echinococcus* have been identified; *Echinococcus granulosus* and *Echinococcus multilocularis*. Recently, 10 different genotypes of *E. granulosus* G1 to G10, of which G1 is the most common one found around the world.^[1,2] Due to the subtypes, *E. granulosus* have been divided into four different species. There is extensive literature on the demographic profile of patients affected with hydatid

disease, but very few on the complications of hydatid disease and on the tools available to deal with them. We bring forth a study on complications of hydatid in a tertiary care hospital in India.

MATERIALS AND METHODS

We reviewed the records of patients who had undergone operation for hydatid cyst of the liver between August 2015 and August 2018. We collected demographic profiles of these patients - age and gender, location of the cysts, and the complications developed in these patients. Of the 34 patients, we chose patients who developed biliary or vascular complications or who had cysts in close proximity to the inferior vena cava (IVC)/hepatic veins. One patient had multiple hydatid cysts in liver, diaphragm, right lung, and in close proximity to the IVC. Subsequently, we found five patients with biliary complications and five patients with vascular complications.

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RESULTS

A total of 34 patients were identified who had undergone operative procedure of hydatid cysts of the liver. There were 20 males and 14 females [Figure 1]. The median age of the patients was 46 years [Figure 2]. All the patients had involvement of the right lobe of the liver with the left lobe involved in four cases. Multiple cysts were present in 28 patients with six patients having single cyst, all in the right lobe of liver.

Ten patients were identified with biliary or vascular complications with five in each group.

Six patients were male and four were female. All patients except one had >1 hydatid. The right lobe was involved in all the cases and segment 3 was involved in one case, which was, however, a part of multicystic disease. Segment 5 was involved in five instances, whereas segments 6, 7, and 8 were involved in four instances each. Among the biliary complications [Figure 3], two patients presented with cholangitis with cysts in the cannabidiol (CBD). Pre-operative endoscopic retrograde cholangiopancreatography (ERCP) was performed in both the cases. Biliary communication was identified intraoperatively in one

of the patients which was suture closed. Two patients developed biliary leak postoperatively, of which ERCP and sphincterotomy were done in one of the patients and the other patient improved with expectant line of management.

Among the vascular complications [Figure 4], one patient had multiple hydatidosis which involved cysts in segments 3 and 8, one cyst in the substance of the right half of diaphragm, in the aortocaval groove in the abdomen, right lung, and in close proximity to the right subclavian artery. All the cysts were marsupialized except the cyst in close proximity to the right subclavian artery, which was dealt with percutaneous aspiration-injection-reaspiration (PAIR).

Similarly, there were two other patients that had cysts in proximity to the IVC that was marsupialized. These patients did not exhibit any features of IVC obstruction preoperatively or postoperatively.

Two patients had cysts in the right lobe of the liver that was in close proximity to the right hepatic vein. While one was dealt successfully, the other patient developed an anaphylactic shock intraoperatively due to an injury to the right hepatic vein. The anaphylactic shock was treated intraoperatively, and the vein was repaired. The patient

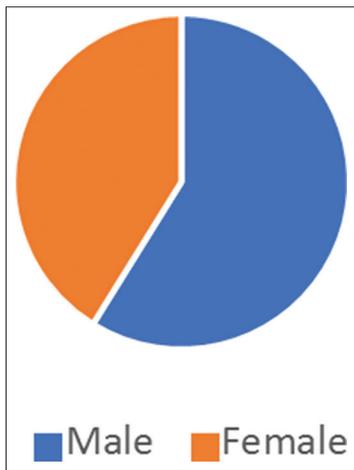


Figure 1: Gender distribution of hydatid cyst

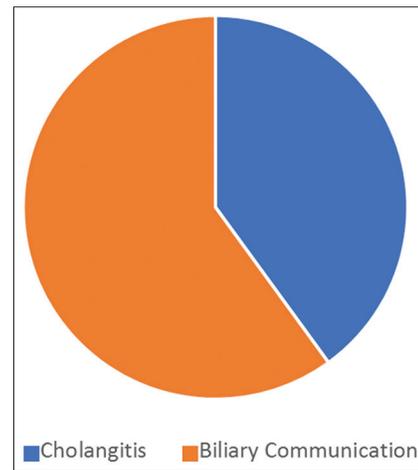


Figure 3: Biliary complications of hydatid disease

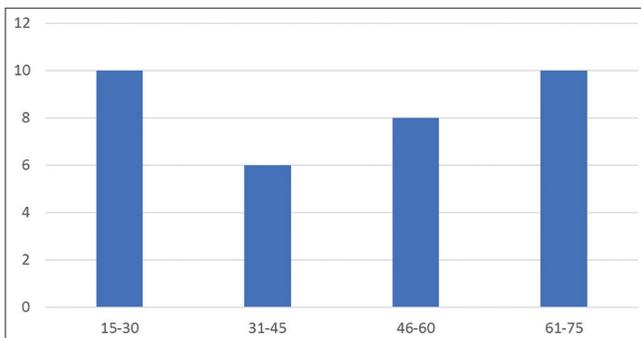


Figure 2: Age distribution of hydatid cysts

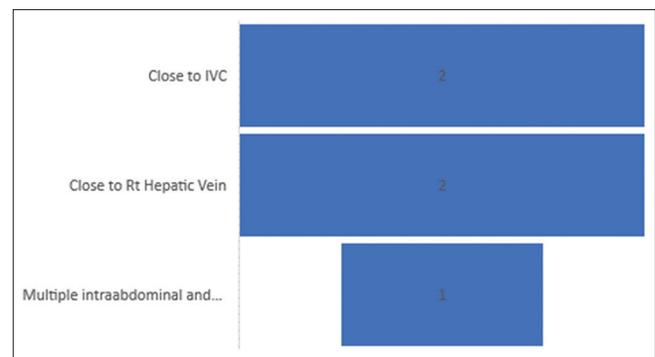


Figure 4: Vascular complications of hydatid disease

recovered well. It was later found out that the patient was non-compliant with pre-operative albendazole treatment.

DISCUSSION

Among the 34 cases of hydatid treated in our hospital, we found 10 cases that caused either biliary or vascular complications. The hydatid disease pathology is caused by two main types - alveolar *Echinococcosis* (AE) and cystic *Echinococcosis* (CE types) with AE type caused by *Echinococcus multilocularis*. CE type is more common pathology^[3] and we did not come across AE-type infection in our series. Liver is the most common organ involved followed by the lung, whereas in some species lung is more commonly involved.^[2] The right lobe of the liver is more commonly involved than the left^[4] as in our series as well. The reason why hydatid cysts and liver abscesses are more common in the right lobe is not clearly lobe. The reason could probably be due to larger bulk of the right lobe, greater width, and more linear lie of the right branch of portal vein and differential blood supply between the two lobes.^[5] Hydatid cysts have also been described in variety of common and unusual locations. Gun *et al.*^[6] in their series reported hydatid cysts in spleen followed by bones, central nervous system, soft tissue, the kidney, and the gallbladder with spleen being the most common unusual site. We had one case in our series which had cysts in both lobes of the liver, in the aortocaval groove, in the substance of the right dome of the diaphragm, and in the right lung and adherent to the right subclavian artery.

Among the biliary complications, two cases presented with cholangitis which on investigation were found to have hydatid in the CBD. The etiology would be a fistulous tract developing between the biliary tracts.^[7] These patients were treated with ERCP preoperatively. It is well documented that cholangitis caused by hydatid cysts in the CBD would improve clinical outcome and cause evacuation of hydatid from the CBD.^[7,8] Sharma *et al.*^[7] had carried out endoscopic drainage in 28 patients who had developed cholangitis due to cystobiliary fistula. Endoscopy and nasobiliary drainage were performed in eight patients while ERCP and stenting were performed in 20 patients. All the patients showed amelioration of symptoms and healing of fistula. After the pre-operative ERCP, these two patients did not show any evidence of post-operative bile leak. Similarly, ERCP and drainage also help in bringing down post-operative bile leak from unsuspecting biliary communication.^[4,9] We had one patient with post-operative bile like that subsided with ERCP. In a randomized controlled trial conducted by El-Gendi *et al.*,^[9] prophylactic endoscopic sphincterotomy in patients undergoing surgery for hydatid cyst of the liver showed significant decrease post-operative bile leak

and shorter duration of hospital stay. Similarly, there was significant volume of bile output in those patients who had a bile leak and there was a lesser need for post-operative endoscopic sphincterotomy.

Vascular complications of hydatid could be difficult to manage. The CE type usually is not densely adherent to major vessels and is not difficult to treat.^[10] The presence of an AE-type cyst near a major vessel may warrant a conservative surgical excision or a radical excision and reconstruction of the vessel if it causes obstruction of the vessel.^[11] There have been reports of Budd-Chiari syndrome caused by obstruction of IVC due to hydatid.^[11,12] Ramia *et al.*^[13] in their series reported operating on 32 patients with hepatic hydatid cyst that was in contact with IVC. They proposed a classification bases on the size of the cyst, length of contact with the IVC, and degree of contact with the IVC. Radical surgery was performed in 20 of the 32 cases. No IVC resection was performed. They reported a morbidity of 28% and mortality of 3%. We had three cases where the cysts were near the IVC. Fortunately, these cysts were of CE type and we were able to deal with them without much eventuality.

We had two patients who had cysts in close proximity to the right hepatic vein. During the procedure, the vein was injured in one of the patients and he went into anaphylactic shock. The patient was revived on table and the vein repaired. There have been case reports of anaphylaxis due to intravascular rupture of hydatid cyst.^[14,15] Pre-operative treatment of hydatid with the imidazole group of drugs makes the cyst less tense and, therefore, reduces the risk of accidental rupture to cause anaphylactic shock.^[3] We later discovered that the patient in question was irregular in his pre-operative treatment of albendazole.

PAIR that is percutaneous aspiration-injection-reaspiration is a method where the hydatid cysts are localized under ultrasound guidance, contents aspirated, the cyst injected with scolicidal agents, and reaspirated.^[16] There have been reports^[16,17] that this method gives good results with low recurrence rates but may cause some allergic reactions with no serious anaphylaxis. In a study conducted by Wang *et al.*,^[18] percutaneous puncture, aspiration, drainage, and curettage was conducted in 302 patients with hepatic and abdominal hydatid cysts. They achieved a success rate of 99% with only one patient developing non-fatal allergy. On follow-up after 6 months, 97.2% of cysts had disappeared and at 5-year follow-up, 77% of the cysts had disappeared. We used PAIR only in one patient when the cyst was adherent to the right subclavian artery and was non-amenable to surgery. We did not encounter any complication during this procedure.

CONCLUSION

Hydatid disease most commonly affects the liver in humans and could also present in unusual locations. The lesion could cause biliary and vascular complications and the treating surgeon must be aware of these complications and must possess the necessary tools to deal with them.

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