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Case Report

Hydatid Cyst of the Liver with Portal Thrombosis: A Case Report

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ABSTRACT

The infection caused by Echinococcus can affect various organs of the body, especially the liver. A 66-year-old woman with a 5-year history of hepatic hydatid cyst was referred to the hospital with the symptoms of new-onset severe abdominal pain without abdominal tenderness and distension. The results of Doppler ultrasound and triphasic computed tomography scan of the abdomen and pelvis showed right and left hepatic venous thrombosis with portal thrombosis and giant hydatid liver cysts. The patient received treatment with anticoagulant and albendazole. The extensive involvement of the liver due to hydatid cyst, the invasion of hydatid cyst to the portal vein, and the resulting portal vein thrombosis are rare complications of hydatid cyst that have been reported in less than ten cases to date. The diagnosis and treatment of hydatid liver cyst and its rare complications in infected patients, such as thrombosis, should be critically considered.

Keywords: Hydatid cyst, Echinococcus, Thrombosis, Hepatomegaly

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INTRODUCTION

Hydatid cyst is caused by a zoonotic infection with the larva stage (metastasis) of Echinococcus granulosus(1). This disease is developed in humans through Canidae, including dogs and foxes, and is more common in some parts of the world, such as in the North Khorasan Province, Iran, where several cases have been reported and can be a health problem(2). These larvae form cysts in the affected organs have an infinite membrane that slowly penetrates around and can cause side effects, including invasion to adjacent organs, metastasis to distant organs, and destructive tissue growth(3,4).

The liver involvement rate has often been reported to be at 50-70% of cases during this disease(1). In most cases, cystobiliary involvement can lead to jaundice and rupture of the abdominal cavity(5), while other organs, such as the lungs, kidneys, and spleen, are less frequently affected(1).

Cysts in the liver can cause problems in 40% of cases and create complications, such as infection, rupture, cholangitis, obstructive jaundice, and pancreatitis. The pressure effect of the mass on the bile ducts and portal and hepatic veins or on the inferior vena cava (IVC) causes cholestasis and high blood pressure in the port (e.g., post hypertension and prehypertension) and Budd-Chiari syndrome (BCS) or urethral obstruction in rare cases(6,7). Regarding this, portal vein thrombosis and cavernous transformation can be caused by numerous factors (which can often be due to cirrhosis, infection, and hypercoagulopathy), and they can be a rare complication of a hydatid cyst disease(8,9).

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This study aimed to report a rare complication of hydatid cyst, in which the patient developed portal thrombosis and hepatic vein and massive liver involvement without cirrhosis despite medical treatment. Such complications have been reported in less than ten cases so far.

CASE REPORT

The patient was a 66-year-old woman living in Chenaran, Razavi Khorasan Province, Iran. She was admitted to the hospital because of abdominal pain in 2013. The results of an ultrasound examination of the abdomen showed that the liver was enlarged with coarse and heterogeneous echotexture without any changes of cirrhosis. Suspicious hyperechoic and heterogeneous ill-defined masses, such as a lesion, were noted. In the next examination, computed tomography (CT) scan of the abdomen and pelvis showed an enlarged liver with abnormal ill-defined hypo-hetro density in both liver lobes. It was revealed that the right lobe was more affected, and the caudate lobe was intact. A core needle biopsy was taken from liver lesions in July 2013. The hematoxylin and eosin-stained sections showed necrotic material containing multiple micro cysts covered by hyaline layered membranes with inflammation. Parasite abscesses compatible with Echinococcus multilocularis were identified. At that time, the patient was not diagnosed as a candidate for surgery. The patient was well treated during this period with albendazole from 2013 to 2019. Due to the patient's lack of regular follow-ups, it was not possible to check the response to treatment.

In 2019, the patient was admitted again to the Imam Reza Tertiary Teaching Hospital, Mashhad University of Medical Sciences, Mashhad, Iran, with a chief complaint of abdominal pain. She suffered from a new-onset severe pain in the right upper quadrant (RUQ) and periumbilical with extension to the right flank from the day before hospitalization. The physical examination upon admission to the hospital showed that vital signs were stable with a low-grade fever of 37.9ºC, a pulse of 100/min, normal blood pressure of 120/80 mm Hg, and icterus.

On abdominal examination, epigastric and right upper quadrant (RUQ) tenderness were observed. She had hepatomegaly and splenomegaly without ascites. Laboratory tests revealed anemia without thrombocytopenia, elevated liver enzymes with hepatocellular pattern with hyperbilirubinemia (aspartate aminotransferase (IU/L):149), alanine aminotransferase (IU/L):118, alkaline phosphatases (U/L):284, total bilirubin:3.3, direct bilirubin:2.1, international normalized ratio:1.21, erythrocyte sedimentation rate:98, and C-reactive protein:18.9 mg/dl. Table 1 demonstrates initial and subsequent laboratory parameters.

Negative results were obtained for other etiologies of this hepatocellular pattern in liver function tests, such as viral hepatitis B, hepatitis C, autoimmune hepatitis, Wilson's disease, and hemochromatosis. Cholangitis was ruled out. Ultrasonography showed enlarged liver with heterogeneous parenchymal echo, containing echogenic masses (the largest 101*73 in five segments of the liver), intrahepatic bile duct dilatation, common bile duct of 7 millimeter; normal gallbladder; echogenic foci suggesting thrombosis in porta hepatic and slow flow in Inferior Vena Cava (IVC), especially intrahepatic; large spleen (140*140); and collateral vessels in the umbilicus, liver, and spleen.

Based on the results of the Doppler ultrasound, the left branch of the port vein was narrow with the partial venous flow; the left port vein wall was involved with parenchymal lesions of the liver; IVC flow was very slow and reciprocating in suprahepatic and subhepatic; thrombosis of IVC was developed; the right hepatic vein was narrowed due to thrombosis and the compressive effect of the involved liver parenchyma; collateral vessels were developed around the spleen hill, and evidence of splenorenal shunt was observed.

The findings obtained from contrast and triphasic CT of the abdomen showed huge hepatomegaly (with a span of 200 mm in midclavicular); disseminated internal calcified areas of the liver with pressure effect on the peripheral organs and its branches and their displacement to the opposite; hypertrophy caudate lobe of liver; the invisibility of IVC, intrahepatic, and hepatic veins; quite a narrow status of the portal vein and its right and left branches; dilated intrahepatic bile ducts; and free fluid in the abdominal and pelvic cavities. (figure 1)

Upper gastrointestinal endoscopy revealed the esophageal varices. (figure 1)

Table 1: Courses of the patient's tests

Variables	1st day	2st day	3st day	4st day	5st day	6st day
WBC (1,000/µL)	6.4	6.3	5.3	5.2	6.1	6
Neutrophils (%)	69	75	68	58	63	50
Lymphocyte (%)	91	18.8	20	26	24	28
Mixed (%)	9.4	5.4	11.4	15	12	21
Hemoglobin (g/dL)	11.5	11.7	11.9	11	11.3	10.5
Platelet count (1000/µL)	400	377	355	327	350	275
MCV (fL)	90	86	87	88	87	90
RDW (fL)	18	18.4	18.2	18.1	18.1	21
Prothrombin time (INR)	1.21	1.29	0.95	1.1	1.5	1.58
Partial thromboplastin time (PTT)	40	30	>180	42	31	38
pH (nmol/L)	7.39					
pCo2 (mmHg)	32					
$HCO3^-$ (mEq/L)	19.9					
Troponin (ng/mL)	Negative					
Total protein (g/dL)	7.5					
Blood sugar (mg/dL)	96					
Urea (mg/dL)	26			27		
Creatinine (mg/dL)	0.6			0.6		
Sodium (mEq/L)	138			138		
Potassium (mEq/L)	4.4			4.3		
Aspartate aminotransferase (IU/L)	149	135		73		42
Alanine aminotransferase (IU/L)	118	126		70		19
Alkaline phosphatase (U/L)	284	280		160		200
Total bilirubin (mg/dL)	3.3	3.4		2.2		1.5
Direct bilirubin (mg/dL)	2.1	2.2		1.5		1.1
Amylase(U/L)	67					
Albumin(mg/dL)	3.8					
LDH(U/L)	368	458				

WBC: White blood cells; RDW: Red blood cell distribution width; MCV: Mean corpuscular volume; INR: International normalized ratio; LDH: Lactate dehydrogenase.

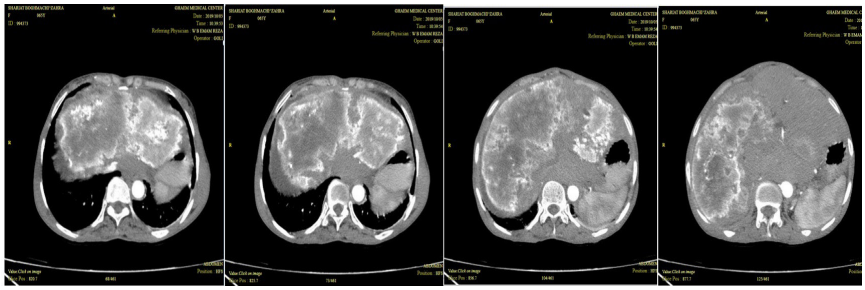

Fig.1: Widely spread hydatid cyst, hepatomegaly, and port thrombosis (second admission)

Treatment with heparin and albendazole was initiated due to portal vein thrombosis and hydatid cyst, which resulted in a decrease in the liver enzyme and bilirubin and improvement in clinical condition.

Due to thrombosis and extensive liver involvement in Echinococcus, the patient received treatment with ceftriaxone and albendazole 400 mg twice a day, heparin infusion for 5 days overlapping with warfarin, and then only warfarin until now. The patient was discharged in good condition with anticoagulant therapy and albendazole therapy. The patient is in follow-up, and she is currently waiting for liver transplantation.

DISCUSSION

The literature review showed that Echinococcus multilocularis and E. granulosus were prevalent in Canidae in the areas where the patients were living, and there were numerous reports of individuals being infected in that region(2). Although E. granulomatous infection most commonly affects the liver, this involvement is generally asymptomatic, and if the cyst grows, it can lead to hepatomegaly, which can have non-specific symptoms, such as nausea, vomiting, and abdominal pain. Cysts in the liver can cause problems. The pressure effect of the mass on the bile ducts and portal and hepatic veins or on the IVC causes cholestasis and high blood pressure in the port (post hypertension and prehypertension) or urethral obstruction in rare cases(6,7). Portal vein thrombosis as a complication is quite rare, and its manifestations can be abdominal pain, fever, portal hypertension, or anaphylactic shock(10).

Hydatid cysts rarely cause secondary BCS because of some reasons such as the occlusion of the hepatic veins and IVC by the invasion of the cyst from neighboring organs, the infestation of the great vessel wall directly by parasites in blood circulation, and compression(11). Vascular erosions, such as hepatic or vena cava veins, can also be other rare complications of hydatid cyst(6). Only three cases have been reported for the hydatid cyst invasion of the portal vein(10). Additionally, hydatid cysts located in the region around the port can lead to the thrombosis of the portal vein and cavernous(12).

Based on the findings of a study, the preferred treatment in cases with hydatid cysts located in the region around the port is surgery and albendazole since the effects of hydatid cysts in this region can lead to thrombosis of the portal vein and cavernous(10). Patients with hydatid cyst and thrombosis require the combination therapy of liver cyst and thrombosis or portal hypertension. Treatments such as albendazole, endoscopic retrograde cholangiopancreatography, and surgery to cure these patients were performed in cases reported from various countries, including Turkey, Spain, and Greece(8,9,13). In a previous study conducted on six patients with thrombosis with hydatid cyst, two cases underwent surgery, and the rest of them were subjected to endoscopic retrograde cholangiography(9,12). Moreover, endoscopic retrograde cholangiopancreatography and catheter-directed thrombolytic therapy were used for one case that was successfully cured(14).

In another research, seven patients were investigated who were infected with Echinococcus with extensive involvement of the liver lobes and liver helium and eventually underwent a liver transplantation. Therefore, orthotopic liver transplantation can be considered a treatment option due to its beneficial results and the low possibility of recurrence(15).

CONCLUSION

Although thrombosis and vascular problems in hydatid cysts are rare, these problems should always be considered, and diagnostic tests related to these complications should be performed. It is recommended that the treatment consists of the combined treatment of hydatid cyst and thrombosis, and if necessary, surgery is considered as an option.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

Authors' contribution

N.Mi. and N.M. collected patient information. N.Mi. interpreted the patient's information based on radiological findings. N.Mi. and L.Gh. participated in writing the article, and all the authors read and approved the final article.

ETHICAL CONSIDERATIONS

This study was reviewed by the Ethics Committee of Mashhad University of Medical Sciences (IR.MUMS.REC.1399.644). The patient expressed her satisfaction with the publication of the article.

Data Availability

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

REFERENCES

1. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev. 2004;17(1):107-35.
2. Heidari Z, Sharbatkhori M, Mobedi I, Mirhendi SH, Nikmanesh B, Sharifdini M, et al. Echinococcus multilocularis and Echinococcus granulosus in canines in North-Khorasan Province, northeastern Iran, identified using morphology and genetic characterization of mitochondrial DNA. Parasit Vectors. 2019;12(1):606.
3. Czermak BV, Unsinn KM, Gotwald T, Waldenberger P, Freund MC, Bale RJ, et al. Echinococcus multilocularis revisited. AJR Am J Roentgenol. 2001;176(5):1207-12.
4. Qu B, Guo L, Sheng G, Yu F, Chen G, Wang Y, et al. Management of Advanced Hepatic Alveolar Echinococcosis: Report of 42 Cases. Am J Trop Med Hyg. 2017;96(3):680-5.
5. Kayaalp C, Bzeizi K, Demirbag AE, Akoglu M. Biliary complications after hydatid liver surgery: incidence and risk factors. J Gastrointest Surg. 2002;6(5):706-12.
6. Emre A, Arioğul O, Alper A, Okten A, Uras A, Yalçin S. Hydatid cysts of liver and portal hypertension. HPB Surg . 1990;2(2):129-33.
7. Kehila M, Said R, Katar S, Ben Hamida R, Khalfallah A, Jerbi A, et al. [Spontaneous peroperative rupture of the inferior vena cava in a hydatid cyst of the liver]. La Tunisie Med 1985;63(6-7):399-402.
8. Kayacetin E, Hidayetoglu T. Hydatid cyst of the liver causing a cavernous transformation in the portal vein and complicated by intrabiliary and intraperitoneal rupture. J Gastroenterol Hepatol.2004;19(10):1223-4.
9. Spanou E, Tzanetou K, Foutris A, Panetsos G, Giannou S, Archimandritis AJAMH. Hydatid disease of the liver causing cavernous transformation of portal vein and portal hypertension. Acta Microbiologica Hellenica 2006;51(4):173-9.
10. Zubiaurre Lizarralde L, Oyarzabal Pérez I, Ruiz Montesinos I, Guisasola Gorrotxategi E. [Invasion of the portal vein by a hydatid cyst. Review of the literature]. Gastroenterologia Hepatologia. 2006;29(7):405-8.
11. Sabzi F, Faraji R. Multiple Complications by Hydatid Cyst-induced Budd Chiary Syndrome: A Case Report. Iran J Parasitol. 2017;12(1):148-51.
12. Kirmizi S, Kayaalp C, Yilmaz S. Hydatid liver cyst causing portal vein thrombosis and cavernous transformation: a case report and literature review. Gastroenterol Hepatol Bed Bench. 2016;9(4):331-5.
13. Moisan FR, Galindo JL, Jarufe NP. An unusual cause of portal cavernomatosis. Ann Hepatol 2012;11(6):944-8.
14. Rasheed K. Hydatid cyst of liver complicated with budd-Chiari syndrome and portal vein thrombosis. N Am J Med Sci . 2013;5(3):242-4.
15. Li F, Yang M, Li B, Yan L, Zen Y, Wen T, et al. Initial clinical results of orthotopic liver transplantation for hepatic alveolar echinococcosis. Liver Transpl . 2007;13(6):924-6.