# Hydatid cyst of the liver with portal thrombosis: A case report

Nasrin Milani[1], Najme Majidi[1], and Laden Goshayeshi[2]

[1]Mashhad University of Medical Sciences
[2]Mashhad University of Medical Sciences Emam Reza Hospital

October 14, 2021

**Abstract**

The extensive involvement of the liver due to hydatid cyst and it's invasion to the portal vein and resulted portal vein thrombosis are rare complications of hydatid cyst. The diagnosis and treatment of hydatid liver cyst and its rare complication such as thrombosis in infected patients should be critically considered.

**Title: Hydatid cyst of the liver with portal thrombosis: A case report**

**Running title: Hydatid cyst and portal thrombosis**

Nasrin Milani[1], Najmeh Majidi[2], Laden Goshayeshi[3]

1. Department of Internal Medicine, School of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.ORCID: 0000-0002-4895-380X.nasrinmilani@gmail.com.
2. Department of Internal Medicine, School of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran
3. Department of Gastroenterology, School of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran. GoshayeshiL@mums.ac.ir.ORCID: 0000-0002-4184-8353.*** Corresponding authors:**

**Nasrin Milani,** MD. E-mail: *nasrinmilani@gmail.com*Tel: 098915502332 Department of Internal Medicine, School of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.

**Abstract**

The extensive involvement of the liver due to hydatid cyst and it's invasion to the portal vein and resulted portal vein thrombosis are rare complications of hydatid cyst. The diagnosis and treatment of hydatid liver cyst and its rare complication such as thrombosis in infected patients should be critically considered.

**Keywords:** Hydatid cyst, *Echinococcus* , Thrombosis, Hepatomegaly.

**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 can be health problem [2]. These larvae form cysts in the affected organs, which have an infinite membrane that slowly penetrates around and can cause side effects [3].

Liver involvement is often reported to be 50% to 70% of cases during this disease [1]. In most cases, cystobiliary involvement can lead to jaundice and rupture of the abdominal cavity[4], while other organs, such as the lungs, kidneys, and spleen, are less frequently affected [1]. This study aimed to report a rare complication

of hydatid cyst, in which the patient developed hepatic vein and port thrombosis and acute liver failure despite medical treatment, of which less than ten cases have been reported to date.

## 2. Case Report

The patient was a 66-year-old woman living in Chenran, Razavi Khorasan Province, Iran. In 2013, she was admitted to the hospital due to abdominal pain. An ultrasound examination of the abdomen showed that liver is enlarged with coarse and heterogeneous echotexture. Suspicious hyperechoic and heterogeneous ill-defined mass like lesion was 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. Right lobe was more affected. Caudate lobe was intact. A Core needle biopsy was taken from liver lesions in July 2013. The H&E stained section showed necrotic material containing multiple micro cysts covered by hyaline layered membranes with inflammation. Parasite abscess compatible with Echinococcus multilocularis were identified. At that time, the patient was not diagnosed as a candidate for surgery and received treatment with Albendazole.

In 2019, the patient was admitted again at the Imam Reza Tertiary Teaching Hospital, Mashhad University of Medical Sciences, Mashhad, Iran, with a chief complaint of abdominal pain. She suffered from new onset sever pain in the right upper quadrant (RUQ) and periumbilical with extension to right flank from the day before hospitalization. On physical examination, vital signs were stable with low-grade fever and icterus. On abdominal examination, epigastric and 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 (AST (IU/l):149), ALT (IU/l):118 Alkaline phosphatases (U/l):284, Total bilirubin:3.3, Direct bilirubin:2.1, INR:1.21). ESR = 98, and CRP = 18.9 mg/dl.

Other etiologies of liver failure such as viral hepatitis B, C and autoimmune hepatitis, Wilson and hemochromatosis were negative. Ultrasound showed enlarged liver with heterogeneous parenchymal echo, containing echogenic masses (the largest 101×73 in 5 segment of liver), intrahepatic bile duct dilatation, common bile duct = 7; normal gallbladder; echogenic foci suggesting thrombosis in porta hepatic and slow flow in inferior vena cava (IVC), especially intrahepatic; large spleen (140×140); collateral vessels in the umbilicus, liver, and spleen;

Doppler ultrasound showed the hepatic arteries of the left branch of the vein of the port were narrow with the partial venous flow; involvement of the left port vein wall by parenchymal lesions of the IVC fluvial liver was highly slow and reciprocating in suprahepatic and subhepatic: IVC thrombosis and susceptibility to thrombosis; narrowing of the right hepatic vein caused by thrombosis as well as the compressive effect of the involved liver parenchyma; and collateral vessels around the spleen hill and evidence of splenorenal shunt.

The findings obtained from contrast and Triphasic CT of the abdomen showed huge hepatomegaly (Its span in midclavicular 200 mm); 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; IVC, intrahepatic and hepatic veins were invisible ; portal vein and its right and left branches in quite narrow status; dilated intrahepatic bile ducts; free fluid in the abdominal and pelvic cavities (figure 1)

Due to acute hepatic and portal vein thrombosis, and hydatid cyst treatment with heparin and Albendazol initiated, the liver enzyme and bilirubin decreased and clinical condition improved. The patient's laboratory tests are shown in Table (1).

Due to thrombosis and extensive liver involvement in*Echinococcus* , the patient received treatment with ceftriaxone and albendazole 4oo mg twice a day, heparin infusion, and then warfarin. The patient was discharged in good condition and is currently waiting for liver transplantation.

## 3. Discussion

The literature review showed that *Echinococcus multilocularis* and*E. granulosus* were prevalent in Canidae in the area where the patient was 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 nonspecific symptoms, such as nausea, vomiting, and abdominal pain.

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 of the 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, prehypertension) and Budd-Chiari syndrome or urethral obstruction in rare cases [5, 6]. Vascular erosions, such as hepatic or vena cava veins, can also be other rare complications of hydatid cyst[5].

The causes of portal vein thrombosis and cavernous transformation are numerous, which can often be due to cirrhosis , infection and hypercoagulopathy and can be a rare complication of hydatid cyst disease[7, 8]. Only three cases were reported of hydatid cyst invasion of the portal vein[9]. Hydatid cysts located in the region around the port can lead to thrombosis of the portal vein and cavernous [10]. Its manifestations can be abdominal pain, fever, portal hypertension, or anaphylactic shock. The preferred treatment in these cases is surgery in addition to albendazole[9].

Patients with hydatid cyst and thrombosis require the combination therapy of liver cyst and thrombosis or portal hypertension. Such treatments as albendazole, endoscopic retrograde cholangiography, and surgery to cure these patients were performed in cases reported from various countries, including Turkey, Spain, and Greece[7, 8, 11]. 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 [8, 10].

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 transplanted. Therefore, orthotopic liver transplantation can be considered a treatment option due to its beneficial results and the low possibility of recurrence[12] .

**Conclusion**Although thrombosis and vascular problems in hydatid cyst are rare, this problem should always be considered and diagnostic tests related to these complications should be performed. It is recommended that the treatment consist of the combined treatment of hydatid cyst, thrombosis, and acute liver failure; moreover, surgery should be considered as an option if necessary.

## Conflicts 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.*Clinical microbiology reviews* . Jan 2004;17(1):107-35. doi:10.1128/cmr.17.1.107-135.2004

2. Heidari Z, Sharbatkhori M, Mobedi I, et al. Echinococcus multilocularis and Echinococcus granulosus in canines in North-Khorasan Province, northeastern Iran, identified using morphology and genetic characterization of mitochondrial DNA. *Parasites & Vectors* . 2019/12/27 2019;12(1):606. doi:10.1186/s13071-019-3859-z

3. Czermak BV, Unsinn KM, Gotwald T, et al. Echinococcus multilocularis revisited. *AJR American journal of roentgenology* . May 2001;176(5):1207-12. doi:10.2214/ajr.176.5.1761207

4. Kayaalp C, Bzeizi K, Demirbag AE, Akoglu M. Biliary complications after hydatid liver surgery: incidence and risk factors. *Journal of gastrointestinal surgery : official journal of the Society for Surgery of the Alimentary Tract* . Sep-Oct 2002;6(5):706-12. doi:10.1016/s1091-255x(02)00046-x

5. Emre A, Arioğul O, Alper A, Okten A, Uras A, Yalçin S. Hydatid cysts of liver and portal hypertension. *HPB surgery : a world journal of hepatic, pancreatic and biliary surgery* . Apr 1990;2(2):129-33. doi:10.1155/1990/54578

6. Kehila M, Said R, Katar S, et al. [Spontaneous peroperative rupture of the inferior vena cava in a hydatid cyst of the liver]. *La Tunisie medicale* . Jun-Jul 1985;63(6-7):399-402. La rupture per-opératoire spontanée de la veine cave inférieure dans un kyste hydatique du foie.

7. Kayacetin E, Hidayetoglu T. Hydatid cyst of the liver causing a cavernous transformation in the portal vein and complicated by intrabiliary and intraperitoneal rupture. *Journal of gastroenterology and hepatology* . Oct 2004;19(10):1223-4. doi:10.1111/j.1440-1746.2004.03584.x

8. 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. 2006;51(4):173.

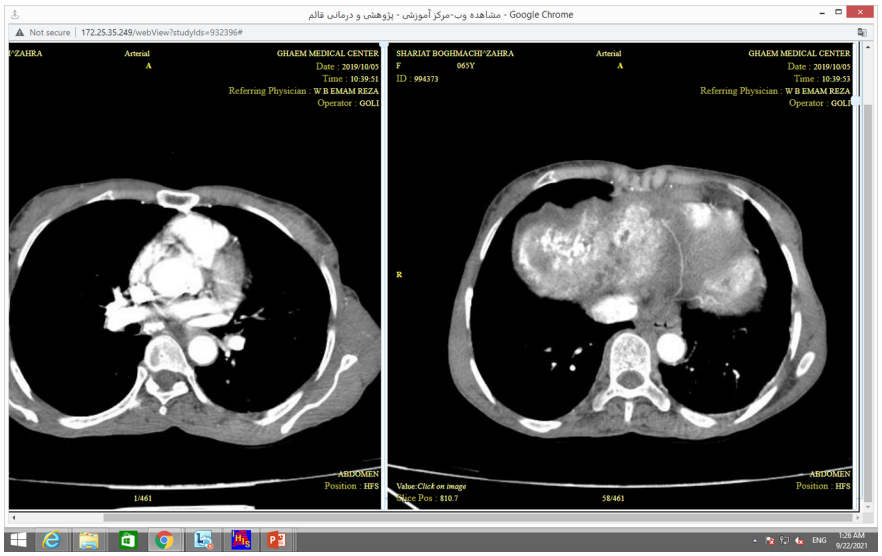
9. Zubiaurre Lizarralde L, Oyarzabal Pérez I, Ruiz Montesinos I, Guisasola Gorrotxategi E. [Invasion of the portal vein by a hydatid cyst. Review of the literature]. *Gastroenterologia y hepatologia* . Aug-Sep 2006;29(7):405-8. Fístula de quiste hepático hidatídico a vena porta: revisión de la literatura. doi:10.1157/13091454

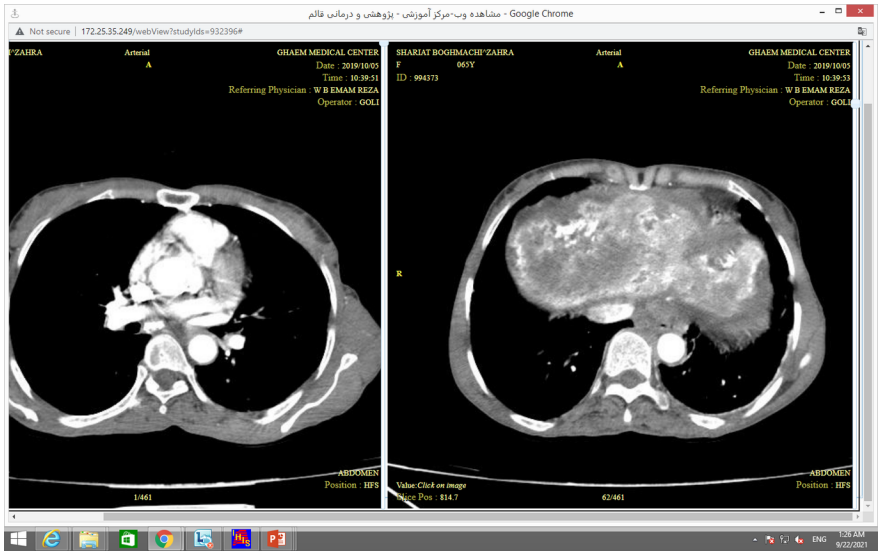
10. Kirmizi S, Kayaalp C, Yilmaz S. Hydatid liver cyst causing portal vein thrombosis and cavernous transformation: a case report and literature review. *Gastroenterology and hepatology from bed to bench* . Fall 2016;9(4):331-335.

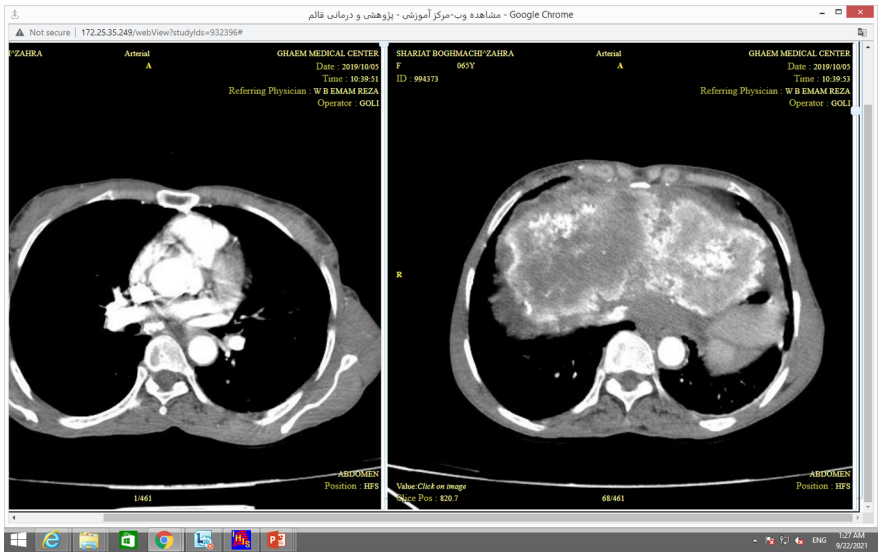
11. Moisan FR, Galindo JL, Jarufe NP. An unusual cause of portal cavernomatosis. *Annals of hepatology* . Nov-Dec 2012;11(6):944-8.

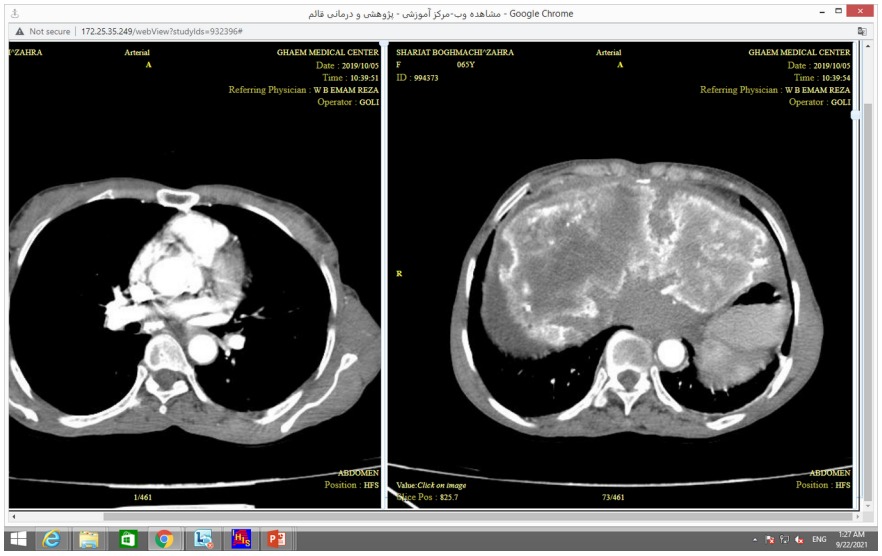
12. Li F, Yang M, Li B, et al. Initial clinical results of orthotopic liver transplantation for hepatic alveolar echinococcosis. *Liver transplantation : official publication of the American Association for the Study of Liver Diseases and the International Liver Transplantation Society* . Jun 2007;13(6):924-6. doi:10.1002/lt.21187

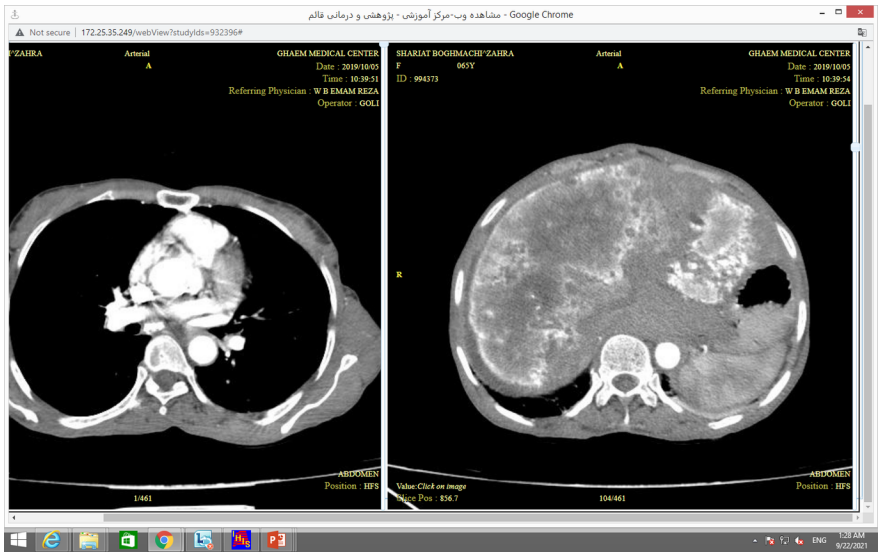
**Figures:**

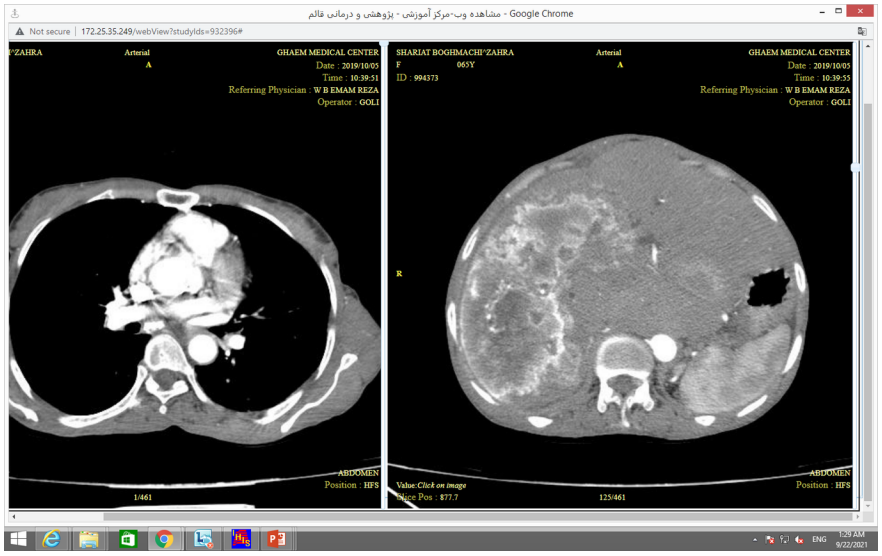


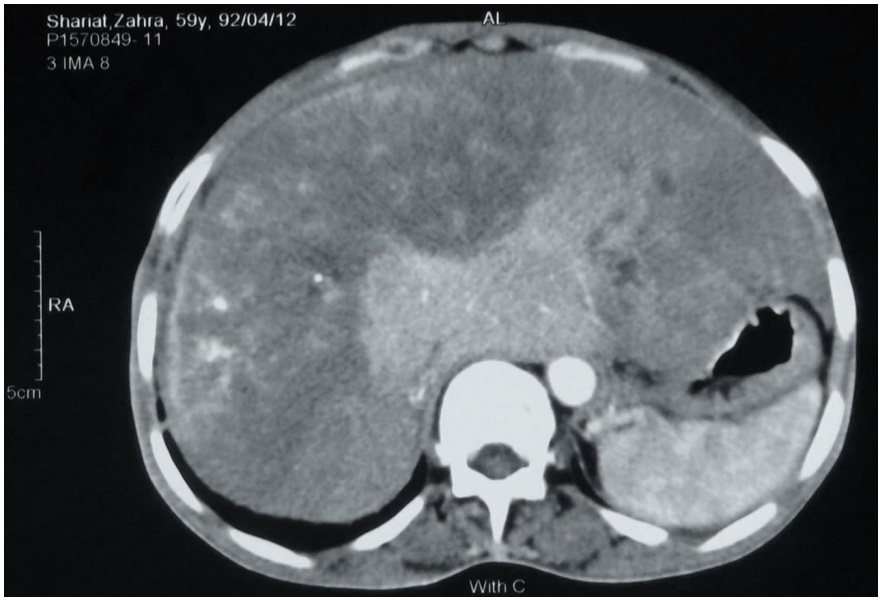









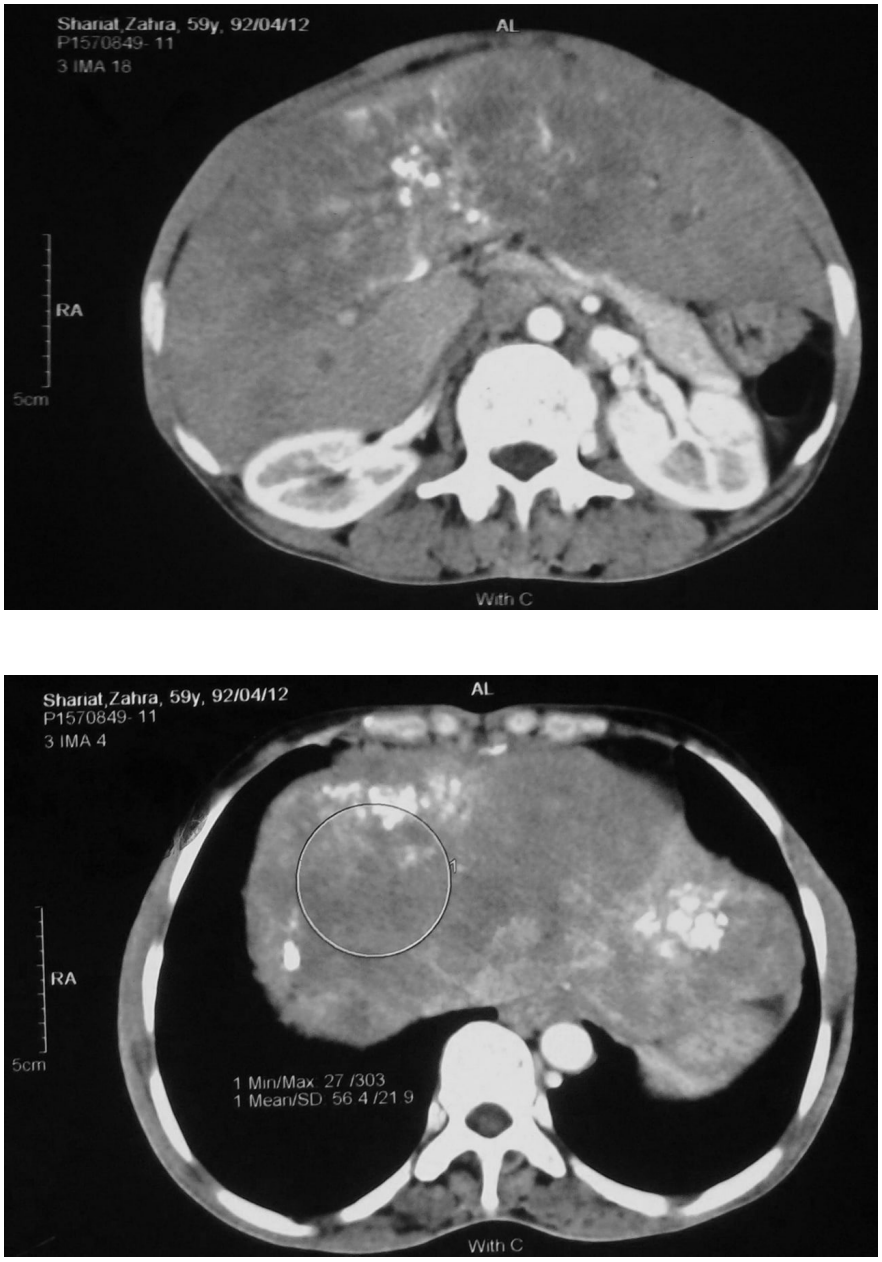




**Figure1.** Widely spread hydatid cyst, hepatomegaly, and port thrombosis (Second admission)

**Table:**

**Table 1** . Courses of the patient's tests

| | First day | Second day | Third day | Fourth day | Fifth day | Sixth 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 |

| | First day | Second day | Third day | Fourth day | Fifth day | Sixth day |
|---|---|---|---|---|---|---|
| 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 | | | | | |
| $pCo_2$ (mmHg ) | 32 | | | | | |
| $HCO_3^-$(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 | | |
| AST (IU/l) | 149 | 135 | | 73 | | 42 |
| ALT (IU/l) | 118 | 126 | | 70 | | 19 |
| Alkaline phosphatase (U/l) | 284 | 280 | | 160 | | 200 |
| Total bilirubin | 3.3 | 3.4 | | 2.2 | | 1.5 |
| Direct bilirubin | 2.1 | 2.2 | | 1.5 | | 1.1 |
| Amylase | 67 | | | | | |
| LDH | 368 | 458 | | | | |

WBC: White blood cells; RDW: Red blood cell distribution width; MCV: Mean corpuscular volume; LDH: Lactate dehydrogenase; Alt: Alanine aminotransferase; AST: Aspartate aminotransferase.

**Hosted file**

`1400.7.7 pic_.docx` available at https://authorea.com/users/422611/articles/541692-hydatid-cyst-of-the-liver-with-portal-thrombosis-a-case-report