



# Imaging Findings in Complicated Hepato-biliary Ascariasis: A Case Report

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## **Authors' contributions**

*This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.*

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**Case Report**

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## **ABSTRACT**

**Aim:** To review the clinical presentation and demonstrate imaging findings in complicated Hepatobiliary Ascariasis (HBA) and its management.

**Presentation:** A 45 years old male diabetic patient presented with the symptoms of fever, hiccoughs, abdominal bloating, nausea, and bilious vomiting lasting for 10 days with elevated bilirubin and Alkaline phosphatase levels.

**Discussion:** HBA arises from the migration of the intestinal parasite *Ascaris lumbricoides* from the duodenum into the biliary tree causing obstruction. HBA may present as, acute cholecystitis, biliary colic, pyogenic cholangitis, pancreatitis, obstructive jaundice, hepato-lithiasis and hepatic abscess. Significant morbidity and mortality are associated with HBA complications and hence early

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diagnosis and management is of utmost significance. Majority of cases are treated conservatively. Endoscopic withdrawal of *Ascaris lumbricoides* is feasible only upon the availability of expert endoscopy services and surgical removal if unavailable.

**Conclusion:** HBA though uncommon, it can direct to serious morbidity and mortality which necessitate early diagnosis of the disease. Clinical features of Ascariasis and laboratory tests are commonly non-specific. Multiple Imaging methods aids in morphological recognition of the roundworm, potential complications of HBA and treatment follow up.

**Keywords:** *Biliary ascariasis; hepatobiliary ascariasis; oriental cholangio-hepatitis; recurrent pyogenic cholangitis; round worm.*

## 1. INTRODUCTION

Ascariasis is one of the most common gastrointestinal parasitic infections in developing countries [1]. People from rural region with poor socio-economic and unhygienic condition tend to have infested with *Ascaris lumbricoides*. Biliary Ascariasis is an uncommon disease, more prevalent in children, females, pregnant woman and in patients who underwent biliary surgery. Adult worm typically lives in small intestine and can foray into the biliary tree and pancreatic duct leading to blockade of bile and pancreatic juice. *Ascaris* causes partial obstruction and spasm of sphincter of Oddi, ultimately leading to cholangitis secondary to chemical irritation with or without bacterial infection. Complications following partial obstruction include empyema, perforation of the gall bladder and sometimes jaundice secondary to Acalculous cholecystitis [2]. The common complication associated with *Ascaris lumbricoides* infestation is biliary colic and it was evidenced in 56-98% cases. It is associated with various clinical manifestations such as nausea, vomiting, abdominal pain and urticaria [3,4]. Complicated HBA present as common bile duct (CBD) stone, cholangitis, Acalculous cholecystitis, liver abscess, and intermittent pancreatitis. Ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), magnetic resonance cholangio-pancreatography (MRCP) and endoscopic retrograde cholangio-pancreatography (ERCP) are the frequently used diagnostic imaging methods. Utmost of the patients with biliary Ascariasis can be managed medically with anti-helminthic treatment, but some cases need ERCP based extraction and surgery in the absence of endoscopy. The intention of this study is to prospectively study the imaging appearances of HBA and methods of management.

## 2. CASE REPORT

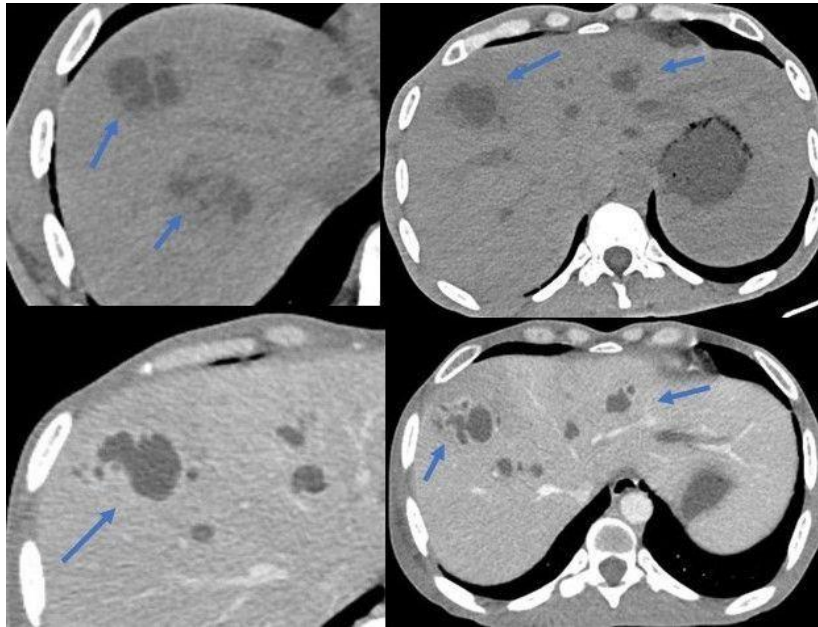
A 45 years old male diabetic patient was admitted with the symptoms of fever, hiccoughs,

abdominal bloating, nausea, and bilious vomiting lasting for 10 days. He also gave the history of passing yellow coloured urine and yellowish discolouration of the eyes. He denied a history of recent blood transfusion. There was no record of prior surgery. On physical examination, the patient had jaundice averagely nourished and built, blood pressure of 110/80 mm Hg, pulse rate of 110/minute, tenderness in the right hypochondrium and hepatomegaly.

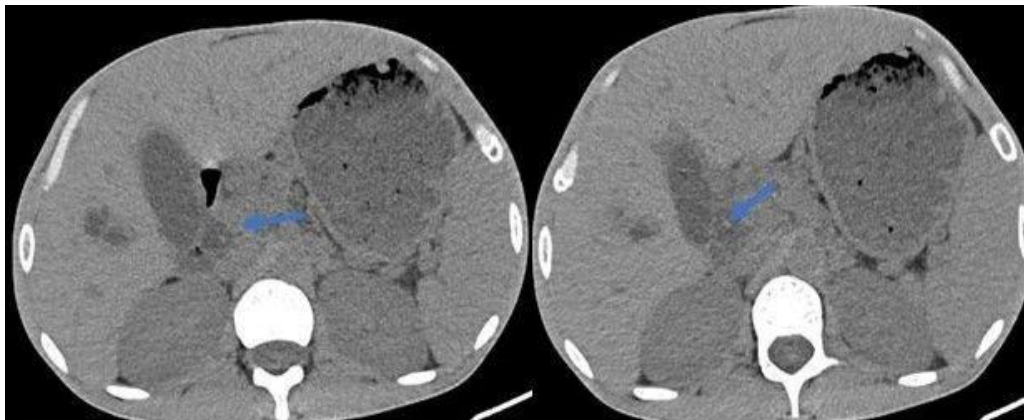
The blood investigations showed leucocytosis (white blood cells – 12900 cells/mm<sup>3</sup>) with elevated bilirubin levels and Alkaline phosphatase (total bilirubin – 3.5 mg/dl, direct bilirubin – 2.5 mg/dl, alkaline phosphatase 1001 IU/L). His Random blood sugar (639mg/dl) and Fasting blood sugar (262 mg/dl) were increased with HbA1C 14.9%. Urine was positive for sugar. Serum triglycerides (532 mg/dl) were elevated with decreased total protein (6 g /dl), albumin (2.3g/dl) and A/G ratio (0.6).

Patient initially underwent non-contrast CT of abdomen which revealed hepatomegaly with multiple focal hypodensities (5-16 millimetre) in both lobes (Fig. 1 A, B)-possibly infective foci / complex cysts. Moderate intra and extra hepatic biliary dilatation (15 millimetre) with narrowing of the ampulla of Vater- probably stricture of benign cause. An isodense dot surrounded by hypodense bile in dilated CBD noted ('Bullseye' appearance) -Suggestive of *Ascaris* (Fig. 2. A, B).

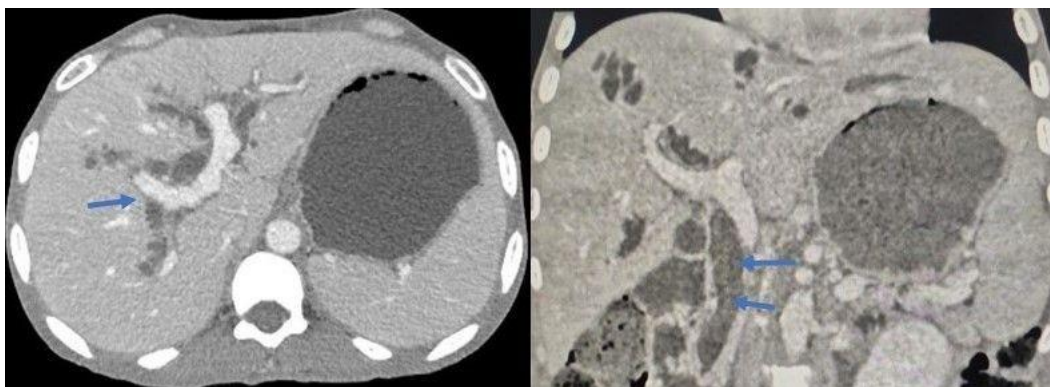
After 2 days, patient underwent contrast CT abdomen with which showed hepatomegaly and multi-septated abscess measuring 43x30x25 millimetre in right lobe segment-8. Gross dilatation (12-13 millimetre) of intrahepatic bile ducts with significant thickened walls, internal debris and sludge (Fig. 3 A, B) - suggestive of cholangitis. CBD was dilated (14mm) with internal debris, strand like opacities and smooth terminal tapering (Fig. 3 A, B). Acute oedematous pancreatitis with mild peripancreatic inflammatory changes were also present.



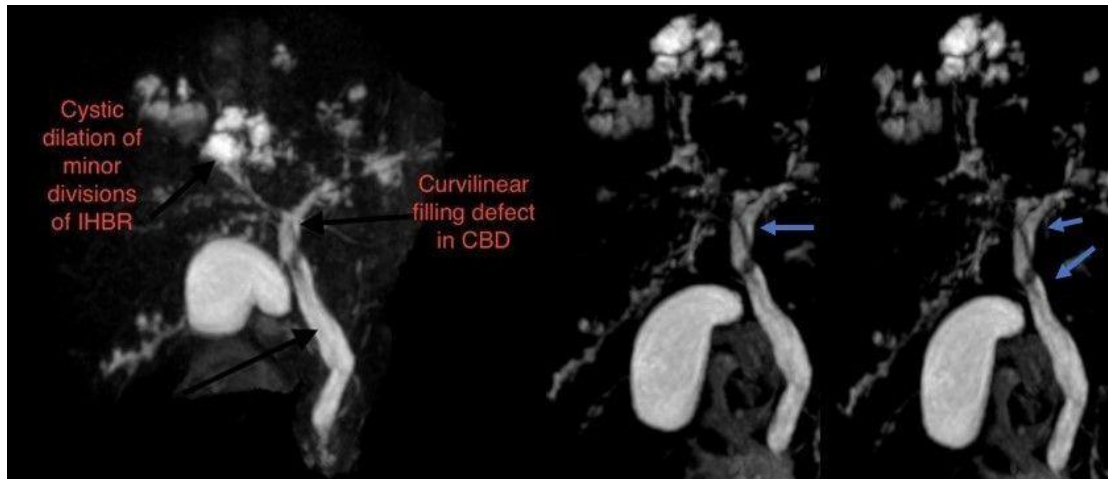
**Fig. 1. (A-D). Non contrast CT(A,B) and Contrast CT (C,D) reveals multiple hypodense abscesses in right & left lobes of liver**



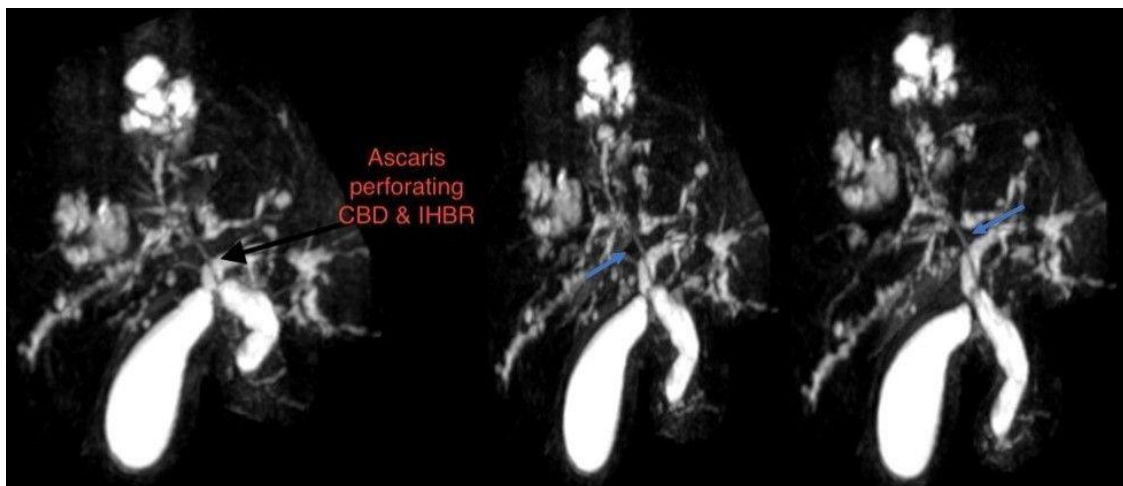
**Fig. 2. (A, B). Non contrast CT images reveal isodense dot surrounded by hypodense bile in dilated CBD ('Bulls-eye' appearance)**



**Fig. 3. (A,B). Contrast enhanced CT axial (A) & reconstructed coronal (B), images shows dilated intrahepatic bile ducts, common bile duct with strand like opacities in common bile duct**



**Fig. 4. (A-C). MRCP images reveal curvilinear hypointense filling defects in CBD suggestive of Ascariasis and cystic dilatation of minor divisions of intrahepatic bile ducts / microabscesses**



**Fig. 5. (A-C). MRCP 3-D maximum intensity projection images reveal linear isointense translocating (perforating) Ascaris in CBD and entering intrahepatic ducts ( Bridge sign) with multiple strictures & dilatations of intrahepatic bile ducts (Beaded appearance)**

The upper gastrointestinal endoscopy showed lax lower oesophageal sphincter, pangastritis and duodenitis. After 8 days of initial treatment, patient was referred for MRCP.

The MRCP showed- Dilated (8-10 millimetre) CBD with linear internal filling defect 5 millimetre in diameter for a length of 70-80 millimetre extending from left hepatic duct to distal CBD (Fig. 4)-possibly biliary Ascariasis. Multiple small cystic dilatations of the branching intrahepatic ducts /micro abscesses (Fig. 4).

Translocating (perforating) Ascaris in CBD and entering intrahepatic ducts is seen as a linear isointense structure connecting hyperintense common bile duct & intrahepatic ducts (likened to

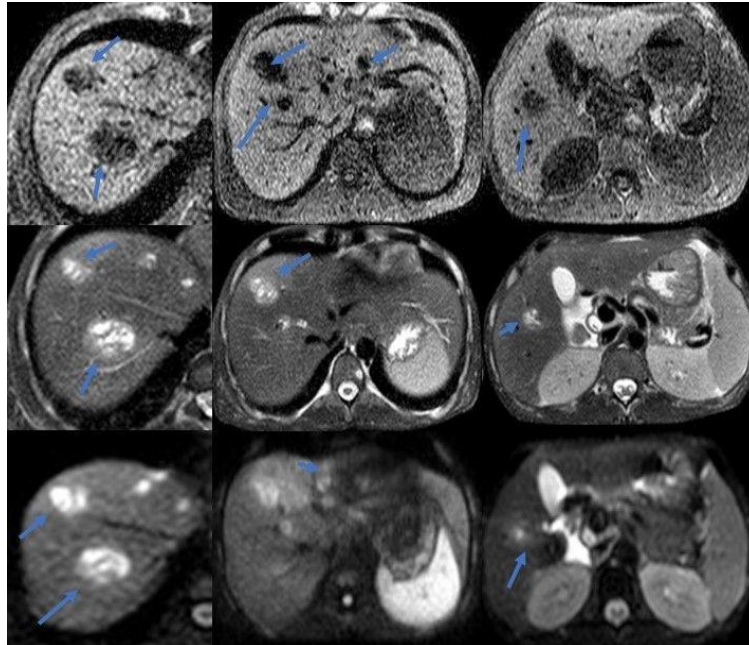
a bridge connecting the water canals-Bridge Sign-Coined by author) (Fig. 5). Dilated intrahepatic biliary radicles with multiple strictures and dilatations ('Beaded appearance') (Fig. 5).

The MRI of abdomen showed multiple abscesses of varying size with internal linear filling defects (Ascariasis) in both lobes of liver (Fig. 6), sludge in gall bladder, thickened walls of intrahepatic bile ducts with internal sludge & pericholangitic oedema (Fig. 7) - features likely to represent oriental cholangiohepatitis / recurrent pyogenic cholangiohepatitis (RPC). Dilated CBD (14 millimetre) with internal curvilinear filling defects (Ascariasis) in the lumen which appear as isointense dot surrounded by hypointense bile on

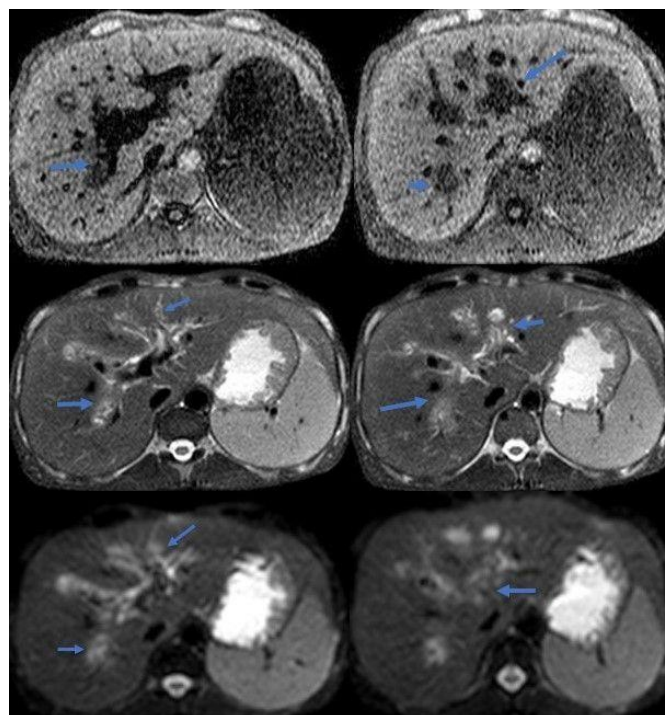


T1 weighted axial images and hypointense dot with surrounding hyperintense bile on T2 weighted axial images ('Eye-glass appearance')

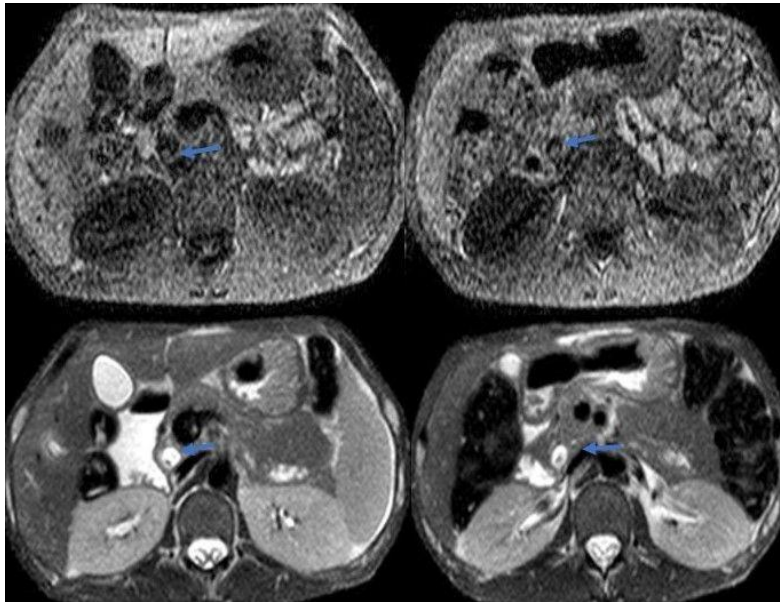
(Fig. 8)- suggestive of Ascariasis. Acute oedematous pancreatitis with peripancreatic inflammatory changes was noted in addition.



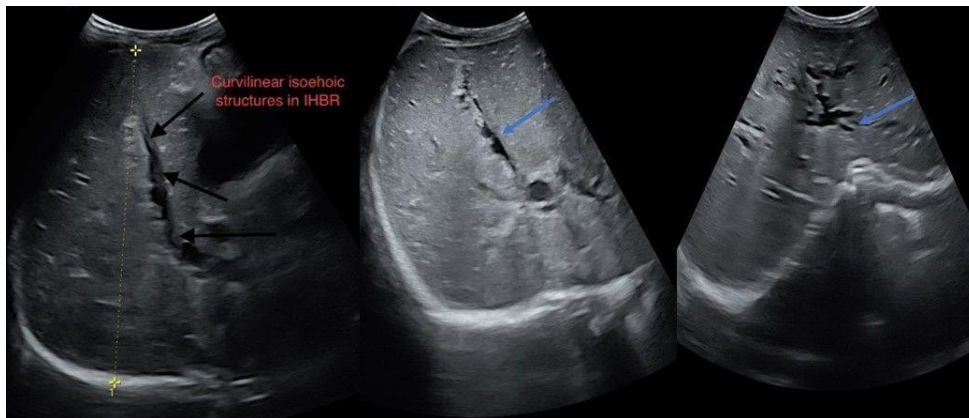
**Fig. 6. (A-I). MRI reveal multiple irregular abscesses of varying size in both lobes of liver, which appear hypointense on axial T1W(A-C), hyperintense on axial T2W(D-F) with restricted diffusion on diffusion weighted imaging (G-I) with filling defects (round worms) within the abscesses**



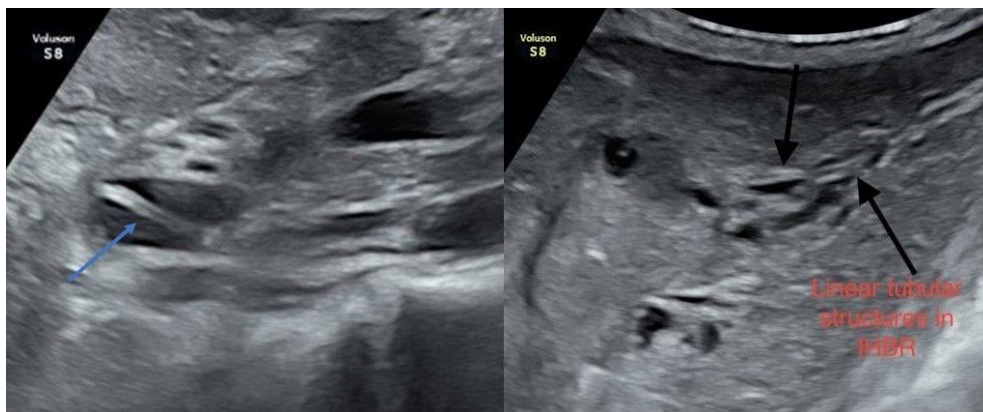
**Fig. 7. (A-F). MRI axial T1W (A, B), T2W (C, D) & diffusion weighted imaging (E, F) images show thickened walls of intrahepatic bile ducts with pericholangitic oedema suggestive of Cholangio-hepatitis**



**Fig. 8. (A-D).** MRI -T1W axial images (A, B) show isointense dot surrounded by hypointense bile and T2W axial images (C, D) show hypointense dot with surrounding hyperintense bile within dilated CBD (Eye-glass appearance)- suggestive of Ascaris



**Fig. 9. (A-C).**Ultrasound images show hepatomegaly, dilated intrahepatic bile ducts with multiple strictures and internal curvilinear isoechoic structures (Ascaris)



**Fig. 10. (A,B).** Ultrasound shows dilated CBD and left hepatic duct with intraluminal tube like structure (Inner tube sign)



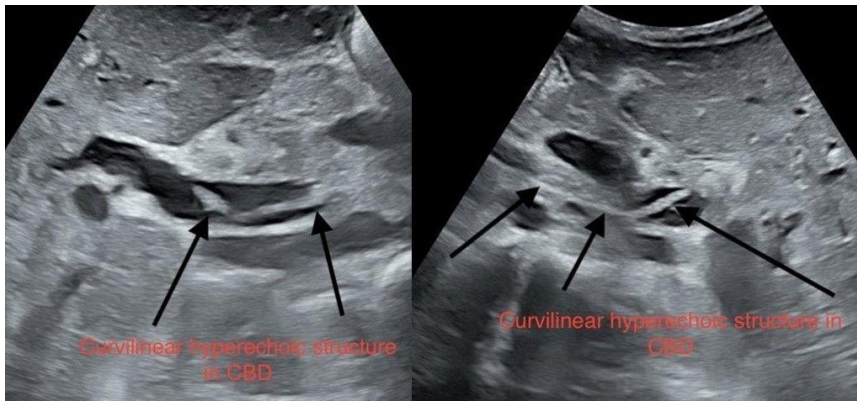


Fig. 11. (A,B). Ultrasound reveals curvilinear non-shadowing strife within the dilated CBD (Strife sign)

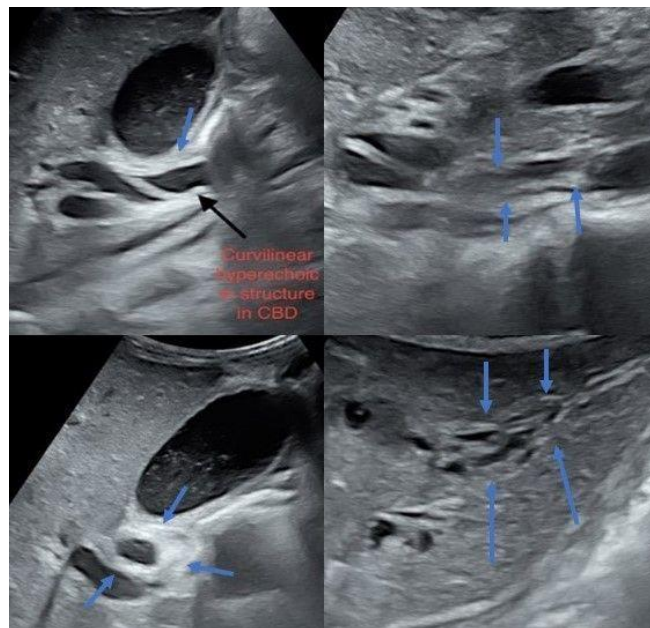


Fig. 12. (A-D). Ultrasound shows numerous elongated linear overlapping echogenic structure (arrows) due to coiling of a single or multiple worms in longitudinal section in CBD & left hepatic duct (Spaghetti sign)

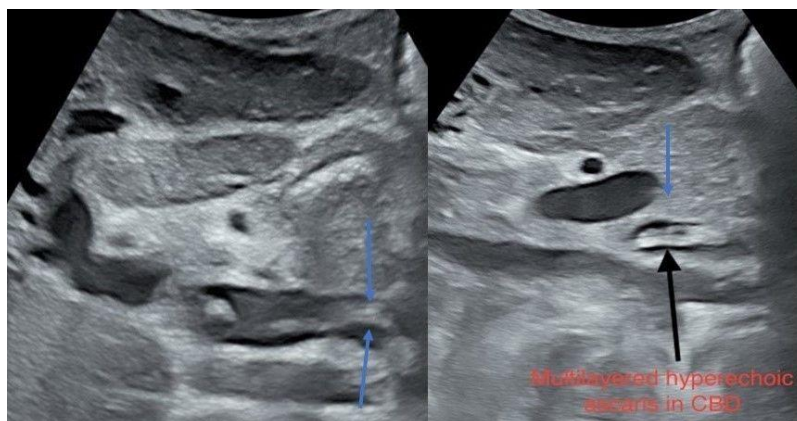


Fig. 13. (A,B). Ultrasound shows trilayered structure in CBD in longitudinal section (Triple sign)

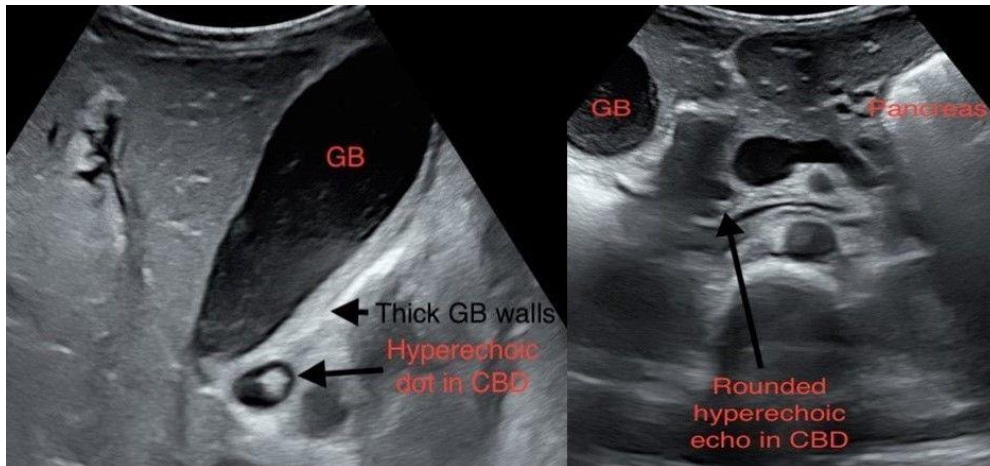


Fig. 14. (A,B). Ultrasound shows hyperechoic nonshadowing dot surrounded by anechoic bile in CBD on transverse section (Bull's eye sign)



Fig. 15. (A-C). Ultrasound pictures show multiple irregular hypoechoic abscesses in right & left lobes of liver, largest measuring 3.7x3.2 centimeters

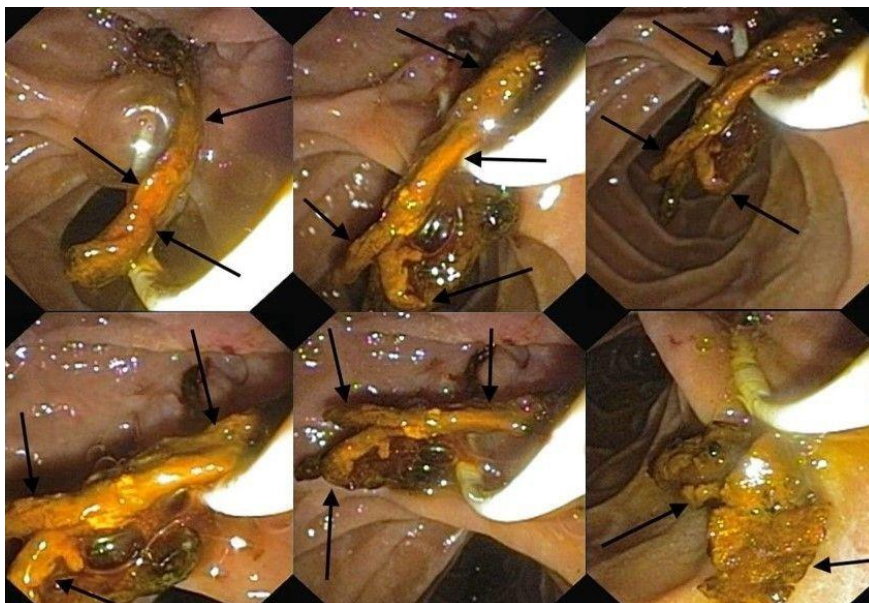


Fig. 16. (A-F). Endoscopic images reveal coiled dead round worms & sludge extracted through ampulla of Vater



The Correlative Ultrasonography revealed dilated (14 millimetre) CBD and dilated intrahepatic bile ducts with internal curvilinear echogenic structures (Fig. 9-14) - suggestive of Ascariasis. Hepatomegaly with reactionary abscesses largest measuring 3.7x3.2 centimetre noted in the right lobe of liver (Fig. 15).

**Differential diagnosis;** in about 50% of cases of recurrent pyogenic cholangitis (oriental cholangio-hepatitis) are infected with *Clonorchis Sinensis* (liver fluke). To be diagnosed, a person needs to have mature liver flukes whose eggs have passed into the stools to be detected by stool test.

The biliary obstruction by cholelithiasis / choledocholithiasis or benign biliary strictures—usually biliary dilatation seen proximal to the obstruction with absence of hepato-lithiasis.

The cases of biliary obstruction by malignant tumours such as cholangio-carcinoma, carcinoma of ampulla of Vater or pancreatic head carcinoma- dilatation of proximal biliary tree is seen along with the malignant mass.

In the primary sclerosing cholangitis, thickening of biliary walls with characteristic “beaded” appearance due to multiple strictures and dilatation with absence of hepato-lithiasis is seen.

In congenital Caroli’s disease, intrahepatic saccular segmental ductal dilation with biliary stasis, cholangitis, intrahepatic abscesses and stone formation noted. Extra hepatic involvement is rare.

Upon comprehensive physical examination, biochemical tests, non-contrast and contrast CT abdomen, MRCP, MRI abdomen and correlative abdominal ultrasound, we successfully diagnosed and treated complicated HBA with RPC.

The cholangitis and liver abscesses were treated conservatively. The patient was dewormed with Albendazole 400 milligram for 3 days and antibiotics for 15 days. Multiple coiled dead Ascariasis and mass of sludge were extracted from the ampulla of Vater endoscopically (Fig. 16). The patient has been followed up in the post endoscopy period. He showed a significant improvement following endoscopic removal of the worms and remained asymptomatic after three weeks of follow up.

### 3. DISCUSSION

Intestinal Ascariasis is a disease of low and middle socioeconomic countries. Biliary Ascariasis is one of the complications of *Ascaris lumbricoides* infestation [5]. *Ascaris* enters the faeco-oral path through contaminated food or water with mature ova. It resides in the jejunum of small intestine and the ingestion of embryonated eggs results in infection [6]. Further, the larvae would reach the duodenum upon gastric juice stimulation. The resultant larvae would further traverse to caecum and insinuate the epithelium by reaching portal vein, hepato-biliary system [7] or in some cases, they spread via the lymphatic system and targets lungs. In the lungs, they can also travel into the alveolar spaces and ascend the larynx and hypopharynx before being swallowed. They undergo maturation throughout this journey resulting in adult worms that reside and mate in the jejunum. A large number of eggs pass through the faeces to complete the cycle. Because of the excessive worm load or distal intestinal obstruction by the worms, it migrates to the duodenum [8].

In utmost of the cases, biliary Ascariasis is asymptomatic. *Ascaris* may reside in the bile duct, or traverse to gall bladder or hepatic parenchyma. Biliary Ascariasis is an uncommon disease, more common in ladies and it is due to progesterone induced sphincter of Oddi relaxation. Utmost of the reports of biliary Ascariasis show the former biliary procedure similar as cholecystectomy, endoscopic sphincterotomy, and choledochoduodenostomy [9].

HBA produces a variety of presentation similar as obstructive jaundice, cholangitis, cholelithiasis, Choledocholithiasis, cholecystitis, pancreatitis, liver abscess, and bile duct perforation [10].

Asiatic or Oriental Cholangio-Hepatitis (OCH) or recurrent pyogenic cholangitis (RPC) is characterised by biliary sludge, chronic secondary bacterial infection and intrahepatic bile duct stones, was first detected in Hong Kong during 1954 and is prevalent in Korea, Taiwan, South East Asia and South China [11].

Fever, pain abdomen, and jaundice (Charcot’s triad) may be seen in the case with biliary Ascariasis and cholangitis [12]. Round worm passage into the biliary ducts initiates the

progression of RPC into destructive cholangiopathy [13]. The occurrence of pyogenic cholangitis is witnessed in 16-25% cases [14]. Stones are common in 90% of cases with RPC, 50% cases within CBD or common hepatic ducts and 15% cases within gall bladder [15]. About 50% of cases with RPC are infected with *Clonorchis Sinensis* (liver fluke) and 12.9% cases have substantiation of Ascariasis [16]. Cases with longstanding, untreated RPC are at threat for developing secondary biliary cirrhosis.

In chronic cases, it may be affected by hepatic abscesses and scarring.

Pancreato-biliary Ascariasis leads to acute pancreatitis in 4-36% of cases. Acute pancreatitis has been attributed to most of the mortality associated with biliary Ascariasis [17].

Since the mortality is high, early identification of the intact roundworm within the abscess is important, and these patients constantly require surgical therapy [18].

Liver function tests may be altered in diversified patterns. Serum alkaline phosphatase is the most constantly increased biochemical parameter in advance. In the presence or absence of pancreatitis, mild to significant increase in the serum amylase can be noticed. Modest neutrophil leucocytosis may be rarely witnessed. After migrant phase of Ascariasis, a rise in the eosinophil count and serum IgE levels, can also be expected and can be of some diagnostic significance.

Detection of the adult worms migrated via the anus or other body orifice or recognition of its eggs in the vomitus, sputum, stool, or small bowel aspirate upon microscopic observation, leads to compulsory diagnosis of Ascariasis. Screening for roundworm ova or fragments can be effectively done by analysis of bile obtained via a nasobiliary or percutaneous transhepatic cholangiography drain.

In utmost of the series ultrasound has high accuracy for biliary Ascariasis diagnosis and to track the progress of the worm present in the biliary tree and to monitor the effect of therapy.

Various ultrasound appearance of the worm has been explained and are as follows:

a) **Inner tube sign or double tube or four lines sign [19]:** on a high frequency probe,

roundworm may be seen as tubular thick echogenic strand with the digestive tracts as central longitudinal anechoic tube in the biliary tree.

b) **Stripe sign [19]:** thin non-shadowing stripe without inner tube within the biliary tree.

c) **Dilated bile ducts [19]:** dilated intrahepatic and extra hepatic ducts in addition to the above-mentioned signs.

d) **Spaghetti sign:** in longitudinal section, numerous linear and long overlapping echogenic structure due to curling of a single or multiple worms may be seen [20].

e) **Pseudo-tumour sign [20]:** in transverse section, coiling of a single or multiple worms seen as a mass.

f) **Triple line sign [20]:** sometimes in addition to 2 echogenic walls of *Ascaris*, internal luminal line may be seen on magnified view or high-resolution linear array transducer with low frequency transducer.

g) **Amorphous appearance:** amorphous echogenic filling defect in the biliary tract due to degraded worm, making the diagnosis difficult.

h) **Linear calcified structures [20]:** calcified dead worms may appear as several linear calcified structures within the bile ducts.

i) **Bull's eye sign** - hyperechoic non-shadowing dot surrounded by anechoic bile in CBD on transverse section.

j) **Mobile structure [21]:** mobility of the worm in the biliary tree, evidently establish the diagnosis.

k) **Railway tract sign [22]:** *Ascaris* may form parallel filling defects within the ducts.

The endoscopic ultrasound has demonstrated sensitivity close to 100% when used together with ERCP in biliary Ascariasis diagnosis.

For the diagnosis of biliary diseases, MRI can provide a veritably specific imaging modality for featuring the worm morphology [23]. T2 weighted axial images show the worm as hypointense dot surrounded by hyperintense signal & isointense on T1 weighted surrounded by hypointense signal within the biliary tree (Eye glass appearance) [23]. MRI is also effective than CT

imaging, since it aids in coronal imaging, improved delineation of fluid surrounding the worm in the CBD and using MRCP 3D maximum intensity projection (MIP) descriptions of the biliary tract can be obtained.

MRCP is the gold standard method, replacing ERCP which is reserved for therapeutic purposes. MRCP can display status of intrahepatic bile ducts, associated strictures and stones along with revealing the disease load in the biliary tract. Intraductal worms are found as linear hypointense filling defects [24]. Translocating (perforating) *Ascaris* in CBD and entering intrahepatic ducts is seen as a linear isointense structure connecting hyperintense CBD & intrahepatic ducts on 3D maximum intensity projection MRCP images ( likened to a bridge connecting the water canals -Bridge sign). Serious Ascariasis load (13-40 worms) is allied with more severe clinical ailment, accounting for 10,000-20,000 mortality cases / year globally [24].

The CT scan is benefits the diagnosis, as it shows the worms within the bile ducts and to portray the degree of probable ailments like cholangitis and hepatic abscess. On non-contrast CT, the roundworms are seen as fairly hyper attenuating tubular structures bordered by less inactivating bile. On transverse section, the worm within the inflated CBD can be seen as 'bullseye' appearance [23]. The entry of worms in gall bladder is easily visualised as curled tubular structures [25]. Contrast enhanced CT abdomen demonstrated dilated intrahepatic bile ducts, CBD and bulky pancreas due to pancreatitis.

In proven cases, owing to inadequate ductal illumination, intravenous cholangiography has a yield lower than 50% [26].

As *Ascaris* infrequently enter the gall bladder, oral cholecystography is indeed less helpful. By the routine availability of ERCP, the indication for percutaneous trans-hepatic cholangiography (PTC) is limited as it has no real advantage.

In ERCP, cholangiograms disclose a wide range of abnormalities such as dilatation, motile tubular structures, irregularity and strictures of the biliary tree. The round worms appear as smooth linear/ parallel filling defects and loops passing the hepatic ducts diagonally. ERCP alone can diagnose 53-58% of biliary Ascariasis and when combined with ultrasound, the sensitivity reaches almost 100% [4]. If a case fails to respond to

conservative treatment; or the live or dead worm is witnessed within the pancreato-biliary tree, then ERCP can be considered as therapeutic intervention. Dead or calcified parasites within the intrahepatic ducts are difficult to remove [27]. In CBD Ascariasis, ERCP extraction has been reported with 90% of successful cases. Incipiently, roundworms inside the pancreatic duct are reluctant towards endoscopic extraction leading to poor prognosis [28].

Although CBD and intrahepatic duct perforation was witnessed in our case, few articles have reported HBA as a major complication of CBD and Intrahepatic duct perforation.

Ascariasis elimination by oral Mebendazole or Pyrantel pamoate was achieved in 90% cases [29]. Pyrantel pamoate is normally used and it causes spastic paralysis in *Ascaris*, whereas Piperazine citrate causes flaccid paralysis leading to worm clearance within 3 days. *Ascaris* in gall bladder is eradicated by cholecystectomy [30]. Anthelmintic treatment targets only the adult worms rather than larvae. Thus, 2-3 months of repeated stool microscopy and ultrasound with appropriate treatment methods must be carried out [31].

In HBA, anti-helminthic treatment should be started only after complete removal of round worms from the bile duct. If antihelminths started before its removal, it leads to the death of worms and further biliary complications. The dead worm remains spark a large inflammatory reaction, which has the potential to cause fibrosis and strictures [32].

Single or numerous parasites in CBD without hepatic invasion react well to conservative treatment [33]. Worms in CBD generally stagnate, die, get macerated or form a nidus for stone formation. In complicated HBA such as cholangitis, CBD stone, dead worms, and a liver abscess may need an intervention [33]. Balloon extraction of the roundworm, endoscopic sphincterotomy and bile duct clearance, along with pharmacotherapy are the methods of management. If endoscopy is not possible, then proceed with either open surgery or laparoscopic surgery [34].

#### 4. CONCLUSION

HBA though uncommon, it can direct to serious morbidity and mortality which necessitate early diagnosis of the disease. Clinical features of



Ascariasis and laboratory tests are commonly non-specific.

- Morphological recognition of roundworm by multiple imaging methods aids in the analysis and treatment follow up. Ultrasound is the first modality of option for diagnosis and follow up of HBA. CT scan is precious in presenting the degree of possible complications of biliary Ascariasis. MRI and MRCP with 3-D maximum intensity projection of the biliary tree are effective non-invasive methods to show biliary Ascariasis. ERCP can be used for endoscopic exclusion of worms in a formerly diagnosed case.
- Long-term effects of HBA comprise recurrent pyogenic cholangitis, hepatic abscesses development and pancreatitis, thus a high index of notion and early intervention should be considered.
- The vast majority of cases respond to conservative anti-helminthic treatment. In non-responders or severely infected cases, endoscopy or surgical exclusion of the worm is essential.

## CONSENT

As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

## ETHICAL APPROVAL

As per international standards or university standards written ethical approval has been collected and preserved by the author(s).

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

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