

CASE REPORT AND MINI-REVIEW

HEPATOBIILIARY ASCARIASIS COMPLICATED BY OBSTRUCTIVE JAUNDICE: CASE-REPORT AND MINI-REVIEW

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ABSTRACT

Introduction. The development of obstructive jaundice caused by hepatobiliary ascariasis is quite rare. The main factors contributing to the occurrence of mechanical obstruction of bile leakage are the death of helminth in the lumen of the duct, the massive invasion of the biliary tract by ascarids, or the accompanying stenotic papillitis.

Case presentation. We describe a case of obstructive jaundice in a woman with massive ascariasis of the biliary tract and stenotic papillitis. The treatment strategy consisted in endoscopic retrograde cholangiopancreatography (ERCP) with mechanical removal of helminths from the bile ducts, papillosphincterotomy, antihelminthic drugs and symptomatic therapy.

Conclusions. Biliary ascariasis must enter into the differential diagnosis of biliary duct pathologies, especially in endemic regions for this parasitosis. ERCP may be an important therapeutic method for this disease.

Keywords: hepatobiliary ascariasis, mechanical jaundice, endoscopic retrograde cholangiopancreatography, antihelminthic agents.

RÉSUMÉ

Ascaridiase hépato-biliaire compliquée d'ictère mécanique: cas clinique et mini-revue

Introduction. Le développement d'une jaunisse obstructive causée par une ascarirose hépatobiliaire est assez rare. Les principaux facteurs contribuant à la survenue d'une obstruction mécanique de la fuite de bile sont la mort d'helminthes dans la lumière du conduit, l'invasion massive des voies biliaires par les ascarides ou la papillite sténosée qui l'accompagne.

Présentation du cas. Nous décrivons un cas de jaunisse obstructive chez une femme avec une ascarirose massive des voies biliaires et une papillite sténosée. La stratégie de traitement consistait en une cholangiopancréatographie rétrograde endoscopique (CPRE) avec élimination mécanique des helminthes des voies biliaires, une papillosphinctérotomie, des médicaments antihelminthiques et un traitement symptomatique.

Conclusions. L'ascaridiase biliaire doit entrer dans le diagnostic différentiel des pathologies des voies biliaires, notamment dans les régions endémiques pour

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INTRODUCTION

Ascariasis is one of the most common parasitic diseases, affecting approximately 25% of the world population (0.8-1.4 billion people)^{1,5}. Ascariasis is common in India, China, the African continent and Latin America⁵. The increase of the prevalence of this disease in developed countries is probably related to population migration and world traveling^{1,6}. Ascariasis is asymptomatic or only associated with subtle abdominal symptoms³. In children, the disease can lead to growth inhibition, protein and vitamin deficiency^{7,8}.

Although adult ascarids are usually found in the small intestine, they can migrate to various organs such as lungs, bladder, or biliary system. Helminth migration is influenced by factors such as fever, medication, general anesthesia, and intraoperative bowel manipulation. The disease can have different presentations, depending on the organs and systems affected¹.

The spectrum of clinical diseases includes pulmonary, intestinal (including intestinal obstruction), appendicular, hepatobiliary and pancreatic ascariasis⁷. The severity of helminth-related diseases has often been underestimated. Hepatobiliary and pancreatic ascariasis has gained attention more recently, with the development of diagnostic imaging techniques (ultrasound, computed tomography scan, etc.)⁵. Researchers became interested in biliary ascariasis in the early 1990s, when reports emerged about the development of mechanical jaundice in helminth infestation in several parts of the world^{9,10-12}. Studies in Kashmir, in the highly-endemic region, have identified the ascariasis in 36.7% of biliary and pancreatic diseases¹³.

Nowadays, the disease is recognized as a major health problem in endemic regions of the world. However, clinicians worldwide should be aware of hepatobiliary ascariasis, since the disease can be observed in non-endemic areas and can lead to severe complications^{1,5}.

The article describes a case of hepatobiliary ascariasis complicated by the development of mechanical jaundice and presents the pathogenesis, clinical manifestations, diagnosis, and possible treatment options, based on the literature review.

cette parasitose. La CPRE peut être une méthode thérapeutique importante pour cette maladie.

Mots-clés: ascariose hépato-biliaire, ictère mécanique, cholangio-pancréatographie rétrograde endoscopique, antihelminthiques.

CASE REPORT

A 70-year-old woman was hospitalized in the surgical department of Ternopil City Emergency Hospital, Ukraine, with symptoms and signs of mechanical jaundice: epigastric pain, general weakness, nausea, moderate skin yellowing, icteric sclerae. The history showed that she has been ill for 3 weeks, and the disease started with epigastric discomfort, and general weakness. Gradually, the intensity of complaints increased. The physical examination revealed skin yellowing, moderate pain on palpation in the epigastrium and right hypochondrium, without signs of peritoneal irritation. Laboratory tests revealed: moderate leukocytosis, eosinophilia (20%), increased total bilirubin 58.63 $\mu\text{mol/L}$, increased urine amylase activity by 2 times. Ultrasound examination showed an expanded choledoch to 1.5-2.0 cm, with hyperechoic shadows in the lumen. The patient underwent endoscopic retrograde cholangiopancreatography (ERCP), that revealed signs of stenosing papillitis, expanded intrahepatic duct, choledoch 20 mm, contrasting two linear filling defects. A sphincterotomy was performed. Three helminths up to 15 cm in length were removed from the lumen of the choledoch (Fig. 1,2). The control cholangiography didn't reveal defects of filling. Ultrasound examination revealed linear hyperechoic images in the choledoch (Fig. 3). ERCP was repeated, because of suspected helminths in extrahepatic pathways. Two helminths were removed from the lumen of the biliary tract (Fig. 4). Antibacterial therapy, antispasmodics, infusion detoxification therapy, aprotinin, albendazole 400 mg once, symptomatic treatment were prescribed. Six days after, the patient was discharged from the hospital in a satisfactory condition, with the recommendation to be monitored by a gastroenterologist and parasitologist.

DISCUSSION

Many reports of biliary ascariasis development originate from the Far East, India, Latin America and parts of the Middle East (Saudi Arabia^{1,13}, Syria¹⁴, etc)^{1,5}.

Anecdotal reports from developed countries describe the development of biliary tract lesions caused by roundworms¹. The infection is rare in major European cities; however, some rural areas have

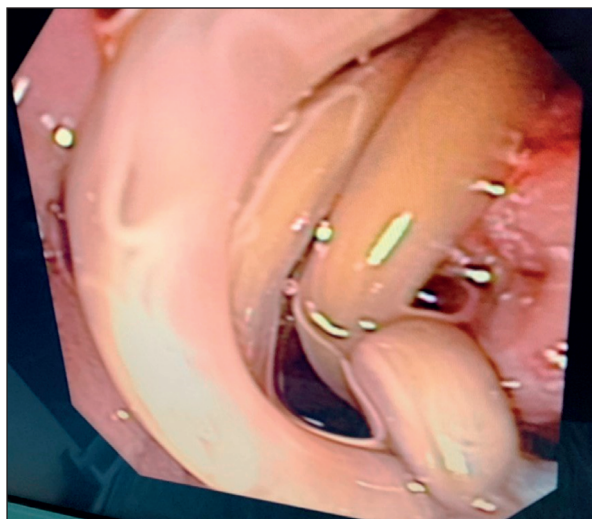


Figure 1. Helminths in the lumen of the choledochus during endoscopic retrograde cholangio-pancreatography.



Figure 2. Ascarids removed during the first endoscopic retrograde cholangio-pancreatography.



Figure 3. Linear hyperechoic images in the projection of the choledoch during the control ultrasound examination

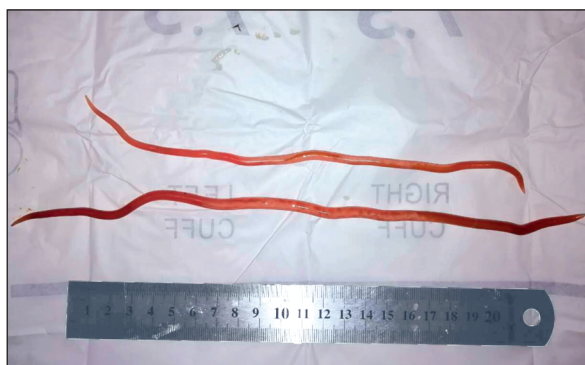


Figure 4. Ascarids removed during repeated endoscopic cholangio-pancreatography.

a high incidence of ascariasis, reaching 52% in some cases^{7,15}. In the United States, about 4 million people are infected with ascarids. Most of the infected patients are immigrants from developing countries^{5,15,16}.

So far, the accurate epidemiological data on the actual incidence of biliary ascariasis are absent due to the the lack of diagnostic equipment¹. Large-scale population migration and increased frequency of travel have contributed to the increase in the incidence of ascariasis. Poverty, overpopulation, and pollution of water resources have increased the spread of the disease in endemic areas¹.

The natural environment of *Ascaris* is the small intestine. Hepato-biliary ascariasis is initiated by the proximal movement of parasites to the duodenum (duodenal ascariasis)⁵. *Ascaris lumbricoides* has a natural tendency to migrate. Massive helminth infestation or intestinal infections of viral, bacterial, or parasitic origin can affect intestinal motility, which causes the migration of ascarids into the biliary tree¹⁷. In the case presented, there was also a massive

helminth invasion of the intestine, that has not been diagnosed preoperatively, because the patient has not sought medical examination for a long time.

In patients with duodenal ascariasis, helminths often enter the bile duct (choledochus ascariasis) via papilla Vateri. Ascarids can also move into the intrahepatic ducts (hepatic ascariasis). More rarely, ascarids enter the gallbladder (ascariasis of the gallbladder)⁵. Sometimes, helminths may enter the pancreatic ducts or cause outflow disorder of pancreatic juice, which may manifest in the form of acute pancreatitis¹⁸. *Ascaris* remains in the lumen of the ducts in a small percentage (2.4%)⁵. The ascaris mobility in the ducts is maintained usually for up to 10 days and during this time the helminth may escape from the ducts. Often, ascaris become trapped inside the bile ducts, die and cause the development of mechanical jaundice, cholangitis, gallstones, and other complications^{5,19,20}. In the case presented, the lumen of the biliary tract contained a large number of ascaris. Probably, the stenosing papillitis was the

reason for helminths' remain in the lumen of the ducts, which made it difficult for the ascarids to enter the lumen of the intestine.

Hepatobiliary ascariasis is mostly an adult disease (mean age 35 years, with an age range of 4-70 years), predominantly female (female/male ratio 3:1)^{1,5,16}. Although ascarid invasion is more common in children, lesions of the biliary system at this age appear in rare cases²¹. This may be due to the smaller size of the ampoule orifice. Hepatobiliary ascariasis is commonly observed in pregnant women, possibly because of hormone-induced relaxation and dilatation of the ampoule opening, which facilitate the ascarid entering into the ducts^{1,5}. Earlier biliary tract surgery (cholecystectomy, choledocholithotomy, sphincteroplasty, endoscopic sphincterotomy, etc) can also be a risk factor for the ascarids entering into the biliary tract^{5,12,17,22}. Yet, prolonged fasting, as reported in a recent study, can cause ascarid migration and development of hepatobiliary helminthiasis¹⁴.

According to the literature, the most common (56 to 98%) symptom of hepatobiliary ascariasis is bile colic besides nonspecific clinical manifestations such as nausea, vomiting, abdominal pain, and urticaria^{1,5,16,23}. Also, cholecystitis, pancreatitis, cholangitis, mechanical jaundice, and liver abscesses may be noted^{1,14,16}.

The diagnosis depends on the visual detection of helminths in the biliary tract. This is sometimes difficult, because most of the ascarids move in and out of the ducts for 7 days¹⁵. Ultrasonography is a highly sensitive and specific imaging technique for detecting the ascarids in the biliary system and assessing their mobility^{7,15,24,25}. Characteristic signs of hepatobiliary ascariasis at ultrasonography are numerous, long, linear, parallel band echogenicity, usually without acoustic shadowing^{3,13}. The ultrasonography disadvantage is that during the study it is impossible to detect helminths in the duodenum or ampullary aperture. The sensitivity of this method in the diagnosis of hepatobiliary ascariasis is slightly higher than 50-80%^{1,7,26}. In our case, ultrasound was useful for the diagnosis and has led to the repeating of ERCP for total cleaning of the biliary tree of helminths.

ERCP is useful for hepatobiliary ascariasis for both diagnostic and therapeutic purposes^{7,11,27}. For the visualization of helminths in the biliary system, it is also possible to perform computed tomography (CT) scan⁷.

Laboratory tests are mainly useless in the diagnosis of hepatobiliary ascariasis. However, the evaluation of blood, liver and kidney functions and serum amylase helps to identify and evaluate the severity of complications⁵. Peripheral eosinophilia is common, due to larval invasion of the blood^{7,28}. Peripheral eosinophilia is present in most patients¹⁷.

The identification of ascarid eggs in the feces has little diagnostic value in endemic areas, since the ascariasis incidence in such regions can range from 30% to 90%⁵. Although anti-ascariasis antibodies develop in infected individuals, they are of little value due to their significant cross-reactivity with other helminths⁷.

Hospitalization is necessary for all these patients. The principles of treatment of biliary ascariasis are simple and definite^{7,29}:

- Treatment of cholangitis or cholecystitis by conservative means.
- Oral administration of antihelmintic agents.
- Endoscopic and surgical treatment.

Obstructive jaundice and intestinal obstruction in patients with hepatobiliary ascariasis are usually indicative of surgery^{7,8}.

Treatment of hepatobiliary ascariasis consists in the treatment of various clinical syndromes by appropriate means. After confirmation of the diagnosis, it is necessary to start the administration of oral antihelmintic agents¹⁷. Paralyzed ascarids are excreted usually by the peristaltic activity of the intestine^{5,26,27}. Most patients have a positive response to conservative treatment and subsequent deworming³. Many antihelmintic preparations have been developed for the effective treatment of ascariasis. The most effective antihelmintics include pyrantel pamoate, mebendazole, albendazole⁷ (Table 1).

In our patient, we used a single administration of albendazole (at a dose of 400 mg) for the deworming, followed by monitoring by a parasitologist.

The antihelmintic drugs introduction directly into the bile ducts, such as piperazine citrate, is not advised, as this may interfere with the removal of live worms from the ducts^{5,7,15,21}.

Sometimes, ascaris in the biliary tract is not easy to cure with antihelmintic agents, as these drugs are poorly excreted in the bile³². When conservative treatment is ineffective, surgery is required, and persistent eosinophilia should alert the physician of this possibility^{7,32}.

ERCP in hepatobiliary ascariasis is necessary in one-quarter of cases in the presence of persistent clinical symptoms³, being both a diagnostic and therapeutic intervention^{17,33}. It involves the helminths detection and removal, where possible, or placement of a stent and subsequent removal of ascarids in the second session^{3,34}. Endoscopic treatment should be performed in the following cases:

- if the intensive treatment and use of antihelmintic agents do not alleviate the patient's symptoms^{5,7,17,35};
- if the helminth does not leave the lumen of the bile duct up to 3 weeks of observation using antihelmintic agents^{5,7};

Table 1. Antihelminthics drugs ^{7,30,31}.

| Medicinal preparation (dosage) | Efficacy against <i>Ascaris lumbricoides</i> | Contraindication | Mechanism of action |
|---|--|------------------------------|---|
| Single dosing of medication | | | |
| Pyrantel pamoate (11 mg/kg, maximum 1 g) | 90-100% | Pregnancy, age under 2 years | Depolarizing neuromuscular blocking agent causes release of acetylcholine & inhibition of cholinesterase leading to muscular paralysis |
| Albendazole (400 mg or 200 mg for children under 2 years) | 100% | Pregnancy | Inhibiting activity of fumarate reductase in parasite, inhibits glucose uptake |
| Levamisole (2-5 mg/kg) | 90% | Pregnancy, renal compromise | Nicotinic receptor agonist causes spastic paralysis of helminth muscles |
| Multiple dosing of medication | | | |
| Mebendazole (100 mg/kg for 3 days) | 100% | Pregnancy, age under 2 years | Immobilization by inhibition the glucose uptake and acetylcholine esterase |
| Piperazini citras (75 mg/kg for 2 days) | 90-100% | Epilepsy | GABA mimetic causes sluggish, reversible paralysis of helminth muscles |
| Tiabendazole (25 mg twice daily for 2 days) | No data | Pregnancy, age under 2 years | Selective interaction with β -tubulin causes paralysis, impairs reproduction and adversely affects oocytes, inhibiting activity of fumarate reductase |

- in case of the development of mechanical jaundice^{7,8,17};
- in the presence of cholangitis with bile strictures or with worms in the gallbladder^{7,36};
- in acute pyogenic cholangitis⁷.

In most patients, helminths can be removed from the biliary ducts, and such patients have rapid regression of symptoms of hepatobiliary and pancreatic system lesions⁵. The risk of complications of endoscopic procedures in such cases is low (6%)²⁸. Given this, endoscopic removal of helminths by traps, dormia baskets or biopsy forceps has become the preferred procedure for biliary ascariasis in recent times^{7,36}.

Due to the development of mechanical jaundice, our patient underwent urgent ERCP. There were no difficulties in removing the worms, but it was impossible to diagnose all the worms during the first procedure. Therefore, ERCP as a diagnostic method, in this case, had low diagnostic value. Perhaps this is due to the massive invasion of ascarid bile ducts. Therefore, in our opinion, in cases of biliary ascariasis, a thorough study of all the ducts, both intraoperatively and postoperatively, is necessary.

Open surgery is indicated only in cases where endoscopic intervention does not allow the complete removal of helminths, especially when the number of ascarids is extremely high¹⁷.

CONCLUSIONS

A serious complication of hepatobiliary ascariasis is the development of mechanical jaundice. The complication develops in the case of ascarids exit

difficulty from the bile ducts, the helminths death in the lumen of the ducts, or during a massive invasion of parasites. Therefore, we consider that the most optimal treatment is to conduct ERCP, to remove helminths from the lumen of the ducts, and to carry out antihelminthic and symptomatic therapy. The careful bile ducts cleaning from the ascarids is extremely important, with mandatory follow-up, including dynamic ultrasound after ERCP. This disease is essential to remember when making the differential diagnosis of pathological changes in the biliary tract.

Author Contributions:

V.B. was responsible for the diagnostic procedures, clinical diagnosis, and treatment decisions. V.B. and A.C. wrote the manuscript. V.B. and A.C. were responsible for the data acquisition. A.C. were responsible for the collection and assembly of the articles/published data, and their inclusion and interpretation in this review. All authors contributed to the critical revision of the manuscript for valuable intellectual content. All authors have read and agreed to the published version of the manuscript.

Compliance with Ethics Requirements:

„The authors declare no conflict of interest regarding this article“

„The authors declare that all the procedures and experiments of this study respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well as the national law. Informed consent was obtained from the patient included in the study“

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