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#### Case Report

Biliary ascariasis and severe bacterial outcomes: Report of three cases from a paediatric hospital in Brazil

Breno Lima de Almeida a, Daniel Vicente Silva b, Mateus Santana do Rosário c, Bruno Simões Dias Gonçalves b, Míria Guimarães Nunes <sup>b</sup>, André Luis Chukr Mafra Ney <sup>b</sup>, Celia Silvany <sup>b</sup>, Isadora Cristina de Siqueira <sup>a</sup>  $\stackrel{\triangle}{\sim}$   $\stackrel{\boxtimes}{\sim}$ 

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# Highlights

- Biliary ascariasis can lead to infectious complications and severe outcomes.
- Three cases of biliary ascariasis with severe infectious complications involving peritonitis, hepatic abscess and sepsis were reported.
- Although often overlooked, it is important to consider ascariasis as a differential diagnosis in children presenting jaundice and abdominal pain in endemic regions.

## Abstract

#### Introduction

Biliary ascariasis, although uncommon, can lead to infectious complications and severe outcomes. This study reported three patients with biliary ascariasis and who were admitted to a paediatric hospital in Salvador, Brazil.

#### Case reports

Case 1: A 1-year-old boy, with HIV, hospitalised with diarrhoea, fever, pain, and abdominal distension. He underwent an exploratory laparotomy, which showed peritonitis secondary to a perforation of the hepatic

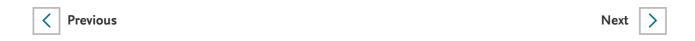
duct by ascaris. Case 2: A 3-year-old boy admitted with fever, abdominal pain and jaundice. Imaging examination was suggestive of ascaris in the intrahepatic biliary tract and a hepatic abscess. Case 3: A 7-year-old boy who was hospitalised with a history of abdominal colic, jaundice and fever, with a suggestive image of ascaris in the biliary tract and evolution to sepsis.

#### Discussion

Three cases of biliary ascariasis were reported with severe infectious complications involving peritonitis, hepatic abscess and sepsis.

#### Conclusion

In endemic regions, biliary ascariasis should be considered in cases with jaundice, abdominal pain and fever, due to its morbidity and risk of complications.



# Keywords

Biliary ascariasis; Ascaris lumbricoides; Sepsis; Liver abscess

## Introduction

Ascariasis, caused by the nematode *Ascaris lumbricoides*, is a soil-transmitted helminthiasis that affects about 1 billion people around the world, mainly children and populations living under poor conditions of basic sanitation in developing countries (de Lima Corvino and Horrall, 2019). It is estimated that this disease, which can greatly impact health and quality of life, infects 29.7 million Brazilians (Scholte et al., 2013), leading to harmful conditions such as long-term cognitive impairment and malnutrition (de Lima Corvino and Horrall, 2019).

Most *A. lumbricoides* infections are asymptomatic or present with mild symptoms such as intermittent abdominal pain; nevertheless, severe complications may occur. Individuals with high parasitic loads can present with partial or total intestinal obstruction, which sometimes requires surgical intervention (Uysal and Dokur, 2017). Ascariasis can also rarely affect ectopic locations such as the appendix, lower airways and the gallbladder (Khuroo, 1996). In biliary ascariasis, roundworms occupy the biliary tract and may cause mechanical obstruction and inflammatory reactions, resulting in biliary colic, acute cholangitis or cholecystitis (Khuroo, 1996).

In episodes of ectopic ascariasis, worms can carry bacteria that lead to infectious complications such as hepatic abscess and sepsis ((de Rocha et al., 1995)) de et al., 1995). As reports documenting bacterial infection arising from ectopic ascariasis are sporadic in the literature, there are no reliable estimates regarding prevalence. Considering that ascariasis and its associated complications continue to be considered a neglected disease, this study reported three cases of biliary ascariasis with severe secondary bacterial infections in children admitted to a paediatric hospital in the city of Salvador, Bahia-Brazil.

#### Case 1

A 1.5-year-old boy with HIV was admitted to a paediatric hospital following episodes of diarrhoea, fever, abdominal pain and distension. Symptoms began 4 days prior to admission. Oral elimination of roundworms was also reported 3 days before admission. On physical examination, the boy was in moderate distress,

hypoactive, remarkably pale, and mildly dehydrated. His abdomen was distended and painful when palpated. An abdominal x-ray showed dilated loops of the small bowel with multiple air-fluid levels and a visible dark tangled mass occupying the duodenum. The patient was treated with piperazine, mebendazole, ceftriaxone, and metronidazole. After 24 hours, the patient's condition worsened and he subsequently underwent an exploratory laparotomy. Biliary peritonitis secondary to perforation of the hepatic duct by a nematode was identified. The worm was removed, the perforation was sutured, and a Kehr's T-tube was inserted for drainage. The patient was admitted to the Intensive Care Unit postoperatively, where he developed pneumonia and went into septic shock, requiring vasoactive drugs and mechanical ventilation. Following improvement he was transferred to the paediatric ward and was discharged after 47 days of hospitalisation with a referral to a specialised HIV/AIDS service.

#### Case 2

M.C.S., a 3-year-old boy, was hospitalised after 4 days of daily fever, diffuse abdominal pain, constipation, loss of appetite, choluria, and jaundice. He was in a generally poor condition, emaciated, groaning, with a dry mouth and sunken eyes. His abdomen was distended with normal bowel sounds, and his liver was palpable 4 cm from the right costal border. Abdominal ultrasonography revealed an image suggestive of roundworms in the gallbladder and intrahepatic biliary tract (Figure 1A). Computed tomography revealed mild hepatomegaly and an expansive process in the right hepatic lobe compatible with a hepatic abscess (Figure 1B). Parasitological examination of his faeces detected *Ascaris lumbricoides* eggs. The patient was treated with piperazine and mebendazole and underwent a 4-week antibiotic regimen consisting of ceftriaxone and metronidazole. He was discharged after 40 days of hospitalisation, after ultrasonography revealed no evidence of parasites in his gallbladder and resolution of the hepatic abscess.



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Figure 1.

#### Case 3

J.S.S., a 7-year-old boy, was hospitalised after 3 days of abdominal distension and pain, jaundice, loss of appetite, and daily fever. He was pale and jaundiced with disseminated petechiae. His abdomen was bulky, he had painful hepatomegaly, and his liver was palpable 4.5 cm from the right costal border and 6 cm from the xiphoid appendix. Abdominal ultrasonography detected an echogenic tubular image in the hepatocholedochal lumen extending into the cystic duct, which was compatible with nematodes within the biliary tract (Figure 1C). Parasitological stool testing evidenced eggs of *A. lumbricoides*. He was treated with mebendazole and piperazine, and started antibiotic therapy with ceftriaxone and metronidazole. The patient's clinical condition worsened, and he evolved with decreased platelet counts and acute renal failure. A diagnosis of sepsis was considered, and he was transferred to a paediatric intensive care unit where he received mechanical ventilatory support. Vitamin K and platelet transfusion were used and hydro-electrolytic

abnormalities were corrected. New ultrasonography performed 8 days after the onset of therapy demonstrated the absence of both obstruction and nematodes. He was discharged at 20 days after achieving clinical and laboratory improvement.

Clinical and laboratory characteristics of the three cases are shown in Table 1.

Table 1. Clinical and laboratory characteristics of three patients with biliary ascariasis from Salvador, Brazil.

	Case 1	Case 2	Case 3
Age (years)	1.5	3	7
Sex	Male	Male	Male
Past medical history	HIV	None	None
Laboratory tests results			
Haemoglobin (g/dl)	5.9	10.8	7.0
Haematocrit (%)	23	32	26
White blood cell count (cells/mm)	16,100	9900	15,400
Segmented neutrophils (%)	77	52	58
Band neutrophils (%)	7	0	18
Lymphocytes (%)	16	46	22
Eosinophils (%)	1	2	2
Platelet count (cells/mm)	328,000	-	27,000
AST (IU/l)	51	-	360
ALT (IU/l)	52	-	450
Urea (mg/dl)	33	_	174
Creatinine (mg/dl)	0.4	-	5.8

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	Case 1	Case 2	Case 3
Clinical presentation	Bowel obstruction/hepatic duct perforation	Cholecystitis/cholangitis	Cholecystitis/cholangitis
Abdomen image	X-ray: dilated loops of the small bowel with multiple air-fluid levels	USG: image suggestive of nematode in the gallbladder and intrahepatic biliary tract CT: hepatic abscess	USG: image suggestive of nematode in the hepato-choledochal lumen extending into the cystic duct
Complications	Peritonitis/septic shock	Liver abscess	Acute kidney injury/sepsis
Surgical intervention	Yes	No	No
PICU admission	Yes	No	Yes
Days to discharge	47	40	20

USG, ultrasonography; CT, computed tomography; AST, aspartate aminotransferase; ALT, alanine aminotransferase; PICU, paediatric intensive care unit.

# Discussion

Ascariasis remains a neglected parasitic disease mostly affecting children in tropical regions (Hotez et al., 2008). Despite the high prevalence in endemic regions, hospitalisations or severe complications are uncommon or underreported. Partial or total intestinal obstruction is the main clinical manifestation associated with paediatric hospital admissions or the need for surgery. This study reported three paediatric hospitalised cases of biliary ascariasis with severe complications.

It is estimated that 10–19% of ascariasis-related hospital admissions are due to biliary obstruction (Sanai and Al-Karawi, 2007). In these patients, biliary duct blockage can lead to jaundice, biliary colic, cholecystitis, and cholangitis. In a large study of biliary ascariasis, including 171 patients from India, the majority of cases presented as cholangitis (42%), cholecystitis (25%) or biliary colic (25%), with few cases with pancreatitis (4%) or hepatic abscess (2%) (Khuroo et al., 1990). Two of the current cases presented with biliary tract obstruction, evidenced by jaundice and abdominal pain. Although hepatic duct perforation was seen in the Case 1, it must be emphasised that few articles have reported biliary tree perforation as a possible complication of this disease (Witcombe, 1978).

Ultrasonography is commonly the first stage of screening in patients with suspected biliary ascariasis, since it offers 85.7% sensitivity in detecting ascariasis in the biliary tree (Khuroo et al., 1977) and was a helpful tool in the diagnosis of the three reported cases herein. Computed tomography is another diagnostic method to detect biliary ascariasis (de Rocha et al., 1995), and proved useful in the second case in the detection of a hepatic abscess. Endoscopic retrograde cholangiopancreatography (ERCP) is an alternative method of detection that can also be used as a therapeutic strategy to remove ascaris from pancreatic ducts and intestinal lumen (Khuroo et al., 2016). While the nematode was surgically removed in Case 1, no surgical intervention was necessary in Cases 2 and 3, since the elimination of worms was spontaneous.

In biliary ascariasis, nematodes migrate from the duodenum, carrying bacterial flora that give rise to secondary infectious complications associated with the disease. Ascaris in the biliary tree has been reported to be responsible for 14.5% of all cases of liver abscess (Das, 2014). In Case 2, the helminth was found outside the

abscess, despite its presence inside this hepatic lesion being documented in the literature (Pereira, 1995). Although hepatic abscess resolution is generally based on surgical intervention, antimicrobial therapy with ceftriaxone and metronidazole sufficiently treated the abscess presented in Case 2.

Sepsis (de Rocha et al., 1995) and peritonitis (Quintos Aranda, 1990), although generally associated with a worsening of the patient's condition, are uncommon in biliary ascariasis, and have been documented in very few articles. Less than 2% of confirmed cases of biliary ascariasis result in death (Khuroo et al., 1992). In spite of the infectious complications related herein, all patients fully recovered from the disease. In each of the present cases, treatment was based on oral anti-helminth medication and antibiotics supplemented by intravenous fluids.

The presence of biliary ascariasis represents a strong indication of high parasitic load, highlighting the need for rapid hospitalisation (Das, 2014). Although often overlooked, it is important to consider ascariasis as a differential diagnosis in children presenting with jaundice and abdominal pain, in endemic regions, as evidenced by these three cases, since secondary infection can rapidly evolve and involve life-threatening complications.

## Author contributions

ICS and CS set the conceptual design of the study; BSDG, MGN, ALCMN, CS, and ICS performed clinical care, diagnosis, and analysis of patients' status; DVS, MSR and BLA performed data collection and data analysis; BLA and ICS made the first draft of the manuscript. All authors read and approved the final manuscript.

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