Ascariasis of the biliary tract : report of 2 cases at the General Surgery Department of Aristide Le Dantec Teaching Hospital and review of the literature

# Ascaridiose des voies biliaires: à propos de 2 cas au service de chirurgie générale du centre hospitaluniversitaire Aristide Le Dantec et revue de la littérature

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# RÉSUMÉ

L'infection à Ascaris lumbricoïdes est une helminthiase cosmopolite qui est endémique dans les pays tropicaux. Souvent asymptomatique, elle est parfois révélée par des complications qui la rendent grave. L'ascariose biliaire est l'une d'entre elles. Nous rapportons 2 cas d'ascariose de la voie biliaire principale dans le service de chirurgie générale de l'hôpital Le Dantec de Dakar (Sénégal). Il s'agissait de deux patients atteints de colique hépatique et d'une angiocholite aiguë chez lesquels le diagnostic a été posé par échographie. Le traitement était chirurgical dans les deux cas. A travers ces 2 cas, nous passons en revue les aspects diagnostiques et thérapeutiques de cette maladie rarement décrite dans nos régions, qui sont pourtant des zones d'endémie ascaridienne.

Mots-clés : Ascaris lumbricoïdes, cholécystite, angiocholite, échographie, chirurgie.

# ABSTRACT

Ascaris lumbricoïdes infection is a cosmopolitan helminthiasis that is endemic in tropical countries of the globe. Often asymptomatic, it is sometimes revealed by complications that make it serious. Biliary ascariasis is one of them. We report 2 cases of ascariasis of the common bile duct in the General Surgery Department of Hospital Le Dantec of Dakar (Senegal). These were two patients with hepatic colic and acute angiocholitis in whom the diagnosis was made by ultrasound. Treatment was surgical in both cases. Through these 2 cases we review the diagnostic and therapeutic aspects of this disease rarely described in our regions, which are however ascaridian endemic zones.

Keywords: Ascaris lumbricoïdes, cholecystitis, angiocholitis, ultrasound, surgery.

# Introduction

Ascariasis of the bile ducts is the presence of adult worms of Ascaris lumbricoïdes in the bile ducts. This is a serious complication of Ascaris lumbricoïdes infection due to the risk of hepatic colic, angiocholitis, cholecystitis, liver abscesses and pancreatitis [1]. It is frequently described in Asia but also, at a lesser extent, in our regions, and Senegal in particular, where Ascaris lumbricoïdes infection is endemic [2,3]. We report 2 cases of ascariasis of the common bile duct, revealed respectively by hepatic colic and acute angiocholitis, confirmed by ultrasound and treated surgically. The aim of our work is to refine, through a review of the literature, the epidemiological, diagnostic and therapeutic aspects of ascariasis of the biliary tract, while emphasizing the contribution of ultrasound to its diagnosis.

# Case 1

A 22-year-old woman, with no specific past medical history, was admitted to the emergency department for right hypochondrium pain and vomiting, which had started 15 days before admission. She had benefited, in a medical center, from an unspecified treatment and an abdominal ultrasound concluded to a gallbladder with a thin and regular wall, partly filled with biliary mud; intra- and extrahepatic biliary tracts were not dilated; liver was normal in size and homogeneous in echo structure. On admission, vital signs were normal and there was no jaundice. She was having pain and a right hypochondrium defense with the presence of Murphy's sign. Lab studies showed no hyperleukocytosis and liver function was normal. A treatment based on analgesics, quinolone and metronidazole was instituted, with the persistence of symptoms. After 2 days of treatment, a new abdominal ultrasound followed by an abdominal computed tomography (CT) were performed. Ultrasound (Figure 1) showed a homogeneous liver with dilation of the main biliary tract at 13 mm in which had a filiform hyper echogenic structure of 6 mm in diameter, evoking parasitizes. Abdominal CT (Figure 2) showed a distended, thin-walled gallbladder with dilation of the common bile duct and the presence of a linear endoluminal structure doubling the lateral wall. The pancreas was homogeneous. A right subcostal laparotomy was then performed on the 4th day of admission. The exploration showed a distended gallbladder with dilation of the cystic duct and the common bile duct. Anterograde cholecystectomy was performed followed by a longitudinal choledoctomy and extraction of a living Ascaris (Figure 3).





Figure 1. Abdominal ultrasound showing the triple "line sign".



Figure 2. Abdominal CT showing dilation of the gallbladder and of the common bile duct with a linear structure doubling its lateral wall (red arrow).

The closure of the bile duct was done on Kehr's drain. In postoperative care, she was put on albendazole 1 tablet of 400 mg per day for 3 days. A wound infection occurred and was treated with appropriate antibiotics and local care. Examination of the worm in parasitology confirmed that it was a female Ascaris 22 cm long and 8 mm in diameter. Kehr's drain was clamped at the 9<sup>th</sup> day before the patient's discharge on the 13<sup>th</sup> day after surgery. Cholangiography by Kehr's drain at 1 month was normal (Figure 4), allowing removal of the drain. Follow up was uneventful after 3 months.



Figure 3. Living Ascaris removed from the common bile duct.



Figure 4. Cholangiography by Kehr's drain showing the vacuity of common bile duct.

# Case 2

A 60-year-old woman was admitted for right hypochondrium pain and vomiting, occurring 5 days before admission. She also reported having vomited a worm. She had undergone 6 months earlier a cholecystectomy by laparoscopy for a symptomatic vesicular lithiasis. The clinical examination showed jaundice. The temperature was 38.5°C and blood pressure was 120/80 mmHg. Physical exam showed pain and defense of the right hypochondrium. There was no pruritus. Lab studies showed hyperleukocytosis. Pancreatic and liver function were normal. Abdominal ultrasound (Figure 5) concluded at aspects evoking obstructive ascariasis of the biliary tract with dilation of common bile duct and intrahepatic bile ducts. In view of this symptomatology, the following treatment was instituted: albendazole 400mg 1 tablet per day, ciprofloxacin 200mg injectable twice daily, metronidazole 500mg injectable 3 times daily and perfalgan1g injectable 3 times daily. Pain and jaundice disappeared after ten days. A subsequent control ultrasound showed the persistence of parasites in the common bile duct. A right subcostal laparotomy was then performed during the 4<sup>th</sup> week of admission. A longitudinal choledoctomy was performed with extraction of 2 dead Ascaris (Figure 6). The closure of the bile duct was done on Kehr's drain. A cholangiography by Kehr's drain, performed on the 7th day after surgery, showed a permeable biliary tract without any obstacle. The removal of the drain was carried out at the check-up at 1 month. Follow-up at 6 months was uneventful.





Figure 5. Abdominal ultrasound with the triple 'line sign' (red arrow) evoking Ascaris lumbricoïdes



Figure 6. Dead Ascaris removed from common bile duct

# **Discussion**

#### **Epidemiological aspects**

Ascaris lumbricoïdes infection is a strictly human and cosmopolitan parasitosis that is endemic in tropical countries around the world. It is in these endemic zones that Ascaris is most frequent in the biliary tract, accounting for 25% of cases of extra-intestinal localizations [4]. It is one of the leading causes of jaundice in children in some regions such as India, China, South Africa and South America [6]. In India, in Kashmir province, Khuroo et al. [7] noted that Ascaris lumbricoïdes was as common a cause of biliary pathologies as gallstones [7]. The transpapillary passage of Ascaris lumbricoïdes is favored mainly by surgical or endoscopic procedures on the biliary tract, such as endoscopic sphincterotomy, cholecystectomy, choledoctomy or even sphincteroplasty [5,8]. Indeed, cholecystectomy, according to Gonzalez et al. [8], would change the movement of fluids in the bile duct, encouraging the migration of Ascaris lumbricoïdes into the biliary tract. In their study, 30% of patients had a medical history of cholecystectomy [8]. For other authors, such as Sandouk et al. [9], these procedures would lead to a gap in the papilla, facilitating the trans papillary passage of Ascaris lumbricoïdes [5,9,10]. In the Sandouk et al study, 77.3% of patients had a medical history of sphincterotomy [9]. One of our patients (Case No. 2) had a medical history of cholecystectomy while the second (Case No. 1) had no history of biliary surgery. However, her sex and age are (young female) favorable for trans papillary migration of Ascaris [8,9]. Indeed, the largest series on ascariasis of the biliary tract showed a net predominance in women [5,8,9,11]. Progesterone, known for its inhibitory properties on smooth muscle contraction, is thought to relax the smooth muscles of the Oddi sphincter in young women, facilitating the passage of Ascaris into the biliary tract [12,13]. As it was the case in our 2 patients, Ascaris are most of the time located in the common bile duct [2,8,14]. In the Gonzalez et al. [8] study, this location is found in 81.1% of cases and in the study of Astudillo et al., in 84.6% [14]. Invasion of the cystic and gallbladder is rare due to the narrow and tortuous nature of the cystic, as well as the passage through the intrahepatic bile ducts [8,15].

#### **Clinical aspects**

Our patients had the 2 most frequent clinical findings: hepatic colic and angiocholitis [16,17]. However, these manifestations are not specific to biliary ascariasis. Various pathologies of the biliary tract (e. g. gallstones, moats) may be responsible for these symptoms [5,8]. In Case No. 2, the fact that the patient had vomited a worm helped us to thing of the possibility of biliary ascariasis. This was found in 25.3% of cases in the Sandouk *et al*'s [9] study, should be taken in account in making the diagnosis of biliary ascariasis [9]. For Case No. 1, the diagnosis was confirmed by laparotomy. This situation is still quite common in endemic areas [5,10]. This is why, in the endemic zone, for many authors, the diagnosis of biliary ascariasis must be discussed in any case of acute biliary symptomatology, before considering the possibility of other pathologies [1].

#### Paraclinic aspects

With sensitivity and a positive predictive value of 98% and 91% respectively, ultrasound is the method of choice in the detection of the presence of Ascaris in the biliary tract [5,6,18]. In fact, it is a simple, non-invasive, accurate and inexpensive method that is rapidly available in our countries limited in means. Thus, this method should be widely used as a first-line diagnostic method in our developing countries where Ascaris lumbricoïdes infection is endemic. In longitudinal ultrasonography section, Ascaris is seen as a linear or curved, tubular echogenic image made of 3 thin, parallel, hyper echogenic lines, without acoustic shadowing. This appearance is known as 'triple line' sign [5,7,18]. This image, corresponding to the walls of the worm and its digestive tract, is mobile with slow, pendula, multidirectional movements (the zig-zag sign [19]) if the worm is alive [5,7]. On transverse section, Ascaris appears like a round hyper echogenic structure with a hypo echogenic center [5,7]. These typical and pathognomonic images help easily to make the differential diagnosis with gallstones and other biliary parasites such as moats [6,7]. For one of our patients (Case No. 1), ultrasound was carried out twice. The negativity of the 1st ultrasound could be linked either to the inexperience of the radiologist, or to the movement back of the worm into the duodenum which can occur according to Das *et al.* [18] between the  $1^{st}$  day and the  $2^{nd}$  week [18]. While a subsequent analysis of the second ultrasound confirmed the ascaridian nature of the bile duct obstruction. It showed the typical 'triple line' sign (Figure 1) characteristic of Ascaris lumbricoïdes, making the abdominal CT done later superfluous. This is why, in our regions, the ultrasound aspects of Ascaris but also of other helminths with hepatic tropism (moats, amoebas) must be well known by practitioners in order to avoid diagnostic and therapeutic delays and wanderings, as well as unnecessary check-ups that are detrimental to patients. But ultrasound is not only a method of choice in the diagnosis; it helps also to monitor the spontaneous exit of the worm from bile duct [7,18]. However, abdominal ultrasound has limitations, especially in cases of location at the lower bile duct. In these cases, abdominal CT or nuclear magnetic resonance imaging (MRI) may be indicated [18,20]. Abdominal CT, usually performed especially in cases of persistent diagnostic doubt, is less sensitive than ultrasound for Ascaris detection, but allows a better assessment of the severity of pancreatitis, angiocholitis, liver damage and elimination of another associated lesion [9,15,18]. On non-contrast CT, Ascaris appears as hyper attenuating tubular structure, surrounded by less attenuating bile and does not rise after injection of contrast [15]. MRI is highly effective in evaluating pathologies of the biliary tract, particularly for detecting ascariasis of the biliary tract, enabling diagnosis to be made in cases where ultrasound has not been contributive [20]. As for endoscopic retrograde cholangiopancreatography, the most sensitive method for identifying the parasite, it shows Ascaris as a mobile or fixed linear filling defect within the biliary tract and offers therapeutic possibilities [15]. However, it is an invasive, expensive method that is not available in our developing countries and is now mainly used for endoscopic removal of Ascaris in already diagnosed case [18].

#### Therapeutic aspects

Treatment of biliary ascariasis may be conservative, endoscopic or surgical [21]. The conservative treatment is based on anthelmintic, mainly benzimidazoles (albendazole, mebendazole, flubendazole) [21]. It is simple, effective, inexpensive, and that is why many authors such as Pasaribu *et al.* [22] recommend it in the first line as soon as the diagnosis is made [22]. However, it is systematically accompanied by ultrasound monitoring of bile ducts vacuity [23]. In case No. 2 we started by this conservative treatment and the ultrasound monitoring of the biliary tract clearance led us to surgery. In the other case (case No. 1), the treatment was immediately surgical, as part of an exploratory laparotomy. Surgical treatment is well codified for ascaridiasis of the common bile duct. It combines cholecystectomy, extraction of the worm by choledocotomy and drainage by T tube. The use of the T tube (Kehr's drain) is recommended. It makes it possible to check the vacuity of the bile ducts and to treat effectively the possible early recurrences by instillation of physiological serum through the drain allowing the worm to be driven out of the bile duct [14,24]. The classic approach route is a right under costal laparotomy. However, laparoscopic approach in experienced hands reduces parietal injury, postoperative pain, morbidity, and length of hospital stay [14,21]. The extraction of the parasite, whether endoscopic or surgical, is always associated with the antiparasitic treatment [15]. Regardless of whether the treatment is conservative, endoscopic or surgical, the course of treatment is usually favorable with low morbidity and mortality [1,8,9,14].

### Conclusion

Ascariasis of the biliary tract is a condition rarely described in our West African countries, which are endemic areas of Ascaris lumbricoïdes infection. However, we must think about it in any case of any acute biliary symptomatology. Abdominal ultrasound is the key tool of the diagnosis. Surgery remains a reliable therapeutic option in our regions.

Déclaration d'intérêts : les auteurs ne déclarent aucun conflit d'intérêt en rapport avec cet article.

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