Diagnostic Pathology

CASE REPORT

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Fasciolopsis buski infection of the biliary tract: a case report

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Abstract

Background *Fasciolopsis buski* is a large fluke that parasitises the human small intestine, with its infection in the biliary tract being even rarer. Given its relatively rare occurrence in recent years, the clinical diagnosis of *F. buski* infections can pose certain challenges.

Case demonstration A 59-year-old male patient with a history of consuming raw pig blood was admitted with recurrent upper abdominal pain for over 10 years. Hepatobiliary and pancreatic magnetic resonance cholangiopancreatography showed stenosis of the lower end of the common bile duct, dilatation of the intrahepatic and extrahepatic bile ducts above, and tortuous strips in the common bile duct, indicating parasitic infection. Histopathological examination further confirmed a diagnosis of parasitic infection with *F. buski* in the biliary tract. The patient was treated with praziquantel after surgery and did not exhibit recurrence during 6 months of follow-up.

Conclusions Biliary tract infection with *F. buski* is a rare parasitic disease. This case report discusses an extremely rare case of *F. buski* infection of the biliary tract caused by consuming raw pig blood. The clinical features, common diagnostic methods, imaging and pathological features, differential diagnosis, treatment, and prognosis of this disease were reviewed to facilitate an improved understanding of this rare condition.

Keywords *Fasciolopsis buski*, Parasites, Biliary tract, Pathology, Diagnosis

Background

Fasciolopsis buski, which was first described by Buski in 1843, is a large fluke parasite of the human small intestine. This worm is mainly distributed in South and Central China, India, and Southeast Asia, with humans and pigs as the primary source of infection [1]. Earlier, *F. buski* infections were a common health problem in numerous countries, but they have now become rare. Consequently, this disease has a restricted understanding due to its low incidence, easily leading to its missed diagnosis and even misdiagnosis [2]. This case report presents a case of *F. buski* infection of the biliary tract caused by the consumption of raw pig blood. Here, we reviewed the clinical features, common diagnostic methods, imaging and pathological features, differential diagnosis, treatment, and prognosis of this disease, aiming to expand the current limited knowledge of this rare parasitic condition.

Case demonstration

A 59-year-old male patient with a history of consuming raw pig blood was admitted to the hospital with recurrent upper abdominal pain for over 10 years. Ten years ago, the patient had reported upper abdominal pain and discomfort without obvious inducement and no waist and back pain, dizziness, headache, cough, expectoration,

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jaundice, pruritus, nausea, vomiting, and blurred vision. However, no specialized treatment was provided during this period. One week ago, the patient experienced recurrent upper abdominal pain and aggravated symptoms. Subsequently, the patient was admitted to our hospital for treatment. Additionally, physical examination revealed mild abdominal tenderness, rebound pain, and muscle tension but a negative Murphy's sign (–).

Blood routine examination demonstrated an eosinophil percentage of 0.10, while the absolute value of eosinophils was 0.68×10^9/L. Hepatobiliary and pancreatic magnetic resonance cholangiopancreatography (MRCP) (Fig. 1) showed that the lower end of the common bile duct had become sharp and the intrahepatic and extrahepatic bile ducts above were dilated. Furthermore, the inner diameter of the common bile duct was approximately 1.2 cm at the widest point, with irregular, tortuous strips in its lumen. Based on the findings of stenosis of the lower end of the common bile duct, dilatation of the intrahepatic and extrahepatic bile ducts above, and tortuous strip shadows in the common bile duct, parasitic infection was considered. After successful endoscopic retrograde cholangiopancreatography (ERCP), a TJF-260 electronic duodenoscope was inserted into the lower part of the duodenum, and the duodenal diverticulum was visualised. Moreover, the nipple was located below the diverticulum with an oval-shaped opening. A 0.035-Fr yellow zebra guide wire was then used to guide the selective cannulation of the bile duct with a triple-lumen needle knife. X-ray imaging further showed developed extrahepatic and intrahepatic bile ducts, whereas the gallbladder did not show any development. Additionally, the common bile duct was found to be slightly dilated with a diameter of approximately 10 mm, accompanied by a strip filling defect in the lower segment. Subsequently, a small incision was made with a knife. A stone balloon was then utilised to explore the suspected filling defect shadow, and one living parasite (approximately 10×15 mm in size) was removed with a stone basket. Histopathological examination was performed on the isolated worm.

Histopathological examination revealed a flat, oval lamellar worm measuring $18 \times 11 \times 2$ mm, as demonstrated in the macroscopic view of the worm in Fig. 2.

Microscopy images (Figs. 3, 4, 5, 6 and 7) showed that the worm had a thick and clearly striated cuticle, with a loose subcutaneous layer and distinct intestinal branches in different sections below the cuticle. The worm body exhibited a reticular excretory system. Furthermore, the central portion of the worm had a well-developed reproductive organ containing a large number of eggs. Evenly scattered vitelline glands were also observed around the worm body.

Combining these imaging findings with the pathological features, a pathological diagnosis of a parasitic infection of the biliary tract was established, with the parasites identified as the adult worms of *F. buski*.

Consequently, the patient underwent regular antiparasitic treatment with praziquantel after surgery. The patient was followed up for 6 months, and no recurrence was reported during this period.

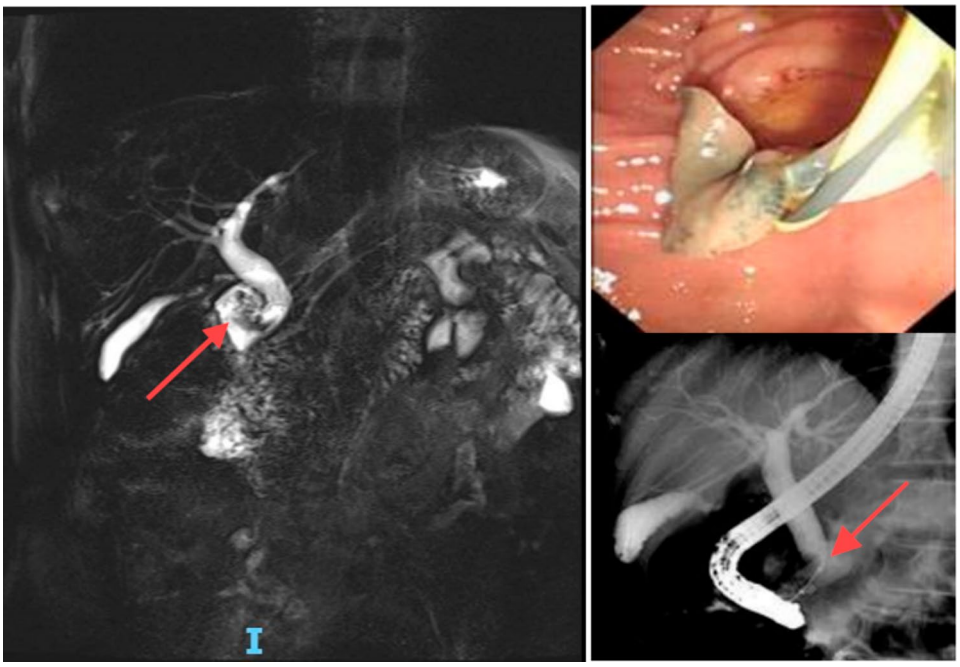

Fig. 1 MRCP and ERCP reveal that the lower end of the common bile duct has become sharp and irregular tortuous streaks are visible in the lumen, indicating the presence of living parasites

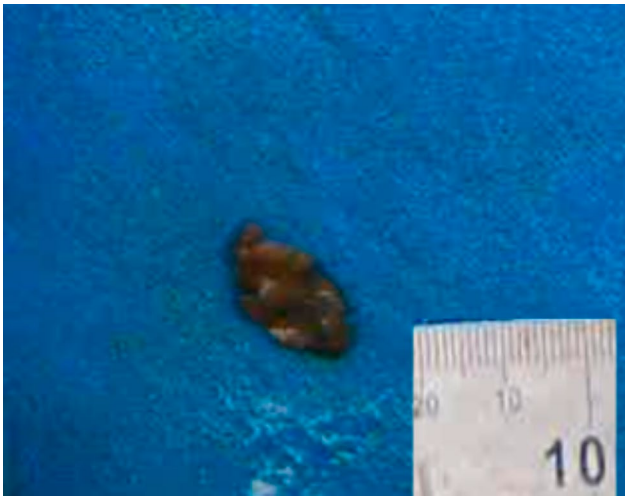

Fig. 2 Gross examination shows an oval-shaped, flat lamelliform worm measuring 18×11×2 mm in diameter

Fig. 3 Low magnification images indicate that the worm body is divided into segments, with the two segments at the upper left representing the peripheral region and the two middle and lower segments designated as the central region

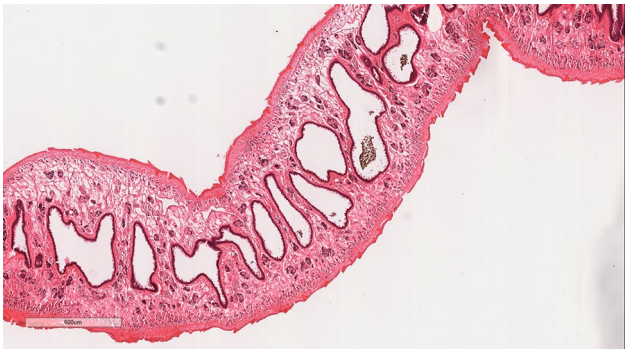

Fig. 4 Microscopic assessment demonstrates irregular intestinal branches, along with excreta in the surrounding area of the worm body

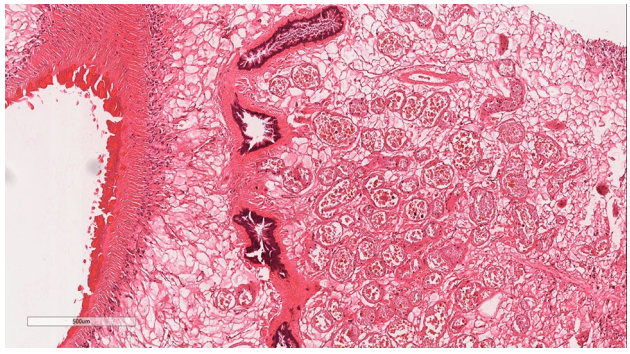

Fig. 5 The worm body exhibits a thick and clearly striated cuticle with a loose subcutaneous layer below

Fig. 6 Well-developed intestinal branches and reproductive organs are observed in the worm

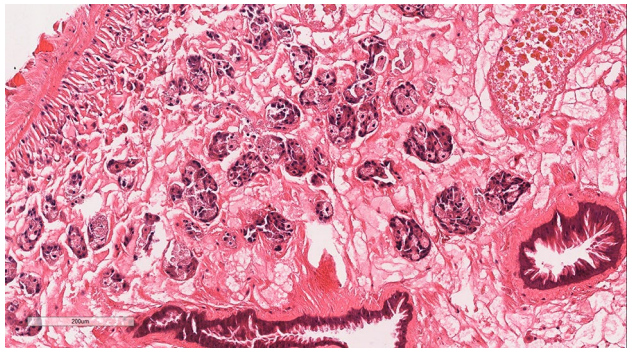

Fig. 7 Evenly distributed vitelline glands are detected around the worm body

Discussion

F. buski has a high infection rate in children and adolescents from 5 to 20 years of age. In China, *F. buski* is distributed across 19 provinces, cities, and districts, except in the southwest, northwest, and northeast regions. This large parasitic fluke is especially prevalent in the middle and lower reaches of the Yangtze River where aquatic plants such as water chestnut and lotus are abundant [3]. In this case report, we reviewed the clinical features, common diagnostic methods, imaging and pathological features, differential diagnosis, treatment, and prognosis

of *F. buski* infection to improve the understanding of this rare parasitic disease.

Life cycle of *F. Buski*

F. buski is the largest trematode that infects the human body. After 1 to 3 months of growth and development, these worms transform into adults before laying their eggs [4]. The final host of *F. buski* is humans or pigs, with aquatic plants acting as vectors [5, 6]. After the host consumes the metacercariae, the posterior cercariae escape under the action of digestive juice and bile and attach to the mucosa of the duodenum or the upper part of the jejunum to absorb nutrition, where they develop into adult worms after approximately 1 to 3 months [7].

Clinical features of *F. Buski*

Adult *F. buski* worms have large bodies, well-developed suckers, and a strong adsorption capacity, which can cause more pronounced intestinal mechanical damage than that induced by other intestinal trematodes. In large numbers, these worms can cover the intestinal wall and hinder intestinal absorption and digestion, while their metabolites can cause allergic reactions after absorption [8]. The adsorbed mucosa may become inflamed and present with bleeding, oedema, necrosis, shedding, and even ulcerations. Furthermore, neutrophils, lymphocytes, and eosinophils infiltrate the lesion site, along with an increased secretion of intestinal mucosa. Abdominal pain, diarrhoea, malnutrition, digestive disorders, decreased serum albumin, and various vitamin deficiencies often occur due to the presence of a large number of worms [9]. Infected individuals may also experience diarrhoea and constipation alternately, which can even lead to intestinal obstruction. Children with severe infection may present with emaciation, anaemia, oedema, ascites, mental retardation, and developmental disorders. A few individuals with recurrent infections may also collapse due to exhaustion and even die [8].

Diagnostic methods and applications

The diagnostic methods of human *F. buski* infection mainly include etiological diagnostic approaches such as the direct smear method [10], precipitation method, saturated saline floating method, and modified Kato–Katz thick smear method [11], while immunological examinations encompass intradermal test, ELISA [12], dot immunofiltration assay [13], and indirect haemagglutination test [14].

Additionally, gastroscopy may be of certain significance in the early detection of fascioliasis because the early clinical symptoms of gingerworm disease are not specific. Therefore, gastroscopy can help identify gingerworm bodies through direct visual evaluation, facilitating the timely detection of gingerworm infection and guidance for correct treatment decisions [6, 15].

Application of diagnostic methods

The implementation of diagnostic methods for human gingerworm infection depends on the developmental stage of the worm invading the human body [10].

In the early disease stage, the larvae migrate into the digestive tract, and the adults are not mature enough to lay eggs. Immunological examination is usually the primary method at this stage. To date, the circulating antigen of *Schistosoma japonicum* has been widely investigated as a diagnostic indicator. This antigen has been proven to have good serological sensitivity and specificity and can reflect the disease course and infection intensity. Some studies have also suggested gastroscopy for the diagnosis of gingerworm disease based on the early symptoms and absorption of the parasites to the descending duodenum.

In the middle and late disease stages, a large number of eggs accumulate in the intestine, allowing the diagnosis through stool egg examination.

Moreover, a previous study [16] that conducted a comparative sequence analysis suggested that *F. buski* organisms from China and India might represent different taxa, while those from Vietnam and China could be the same species.

Imaging of lesions in the hepatic bile duct

Mechanical stimulation by the suckers and spines of these parasitic worms in the hepatobiliary duct mainly causes the proliferation of bile duct epithelium and inflammatory changes in the bile duct wall, along with secondary infection leading to cholangitis. In the cases where the lesion was mild, the bile duct showed localized dilatation. In severe lesions, the worms might block the bile duct and induce cholestasis, which would cause thickening of the bile duct wall and branches, lumen dilatation, and extensive bleeding of the bile duct that ultimately result in anaemia. Adult worms can also produce an inflammatory reaction in the gallbladder, which can be detected through a B-ultrasound examination that shows dilatation of the bile ducts and hypertrophy of the gallbladder wall [3].

Biliary parasitic diseases are a clinically challenging diagnosis, and some patients require ERCP, followed by pathological biopsy after parasite removal. ERCP is a treatment option that can be employed for both ascariasis and fascioliasis. Therefore, ERCP plays a crucial role in the preoperative management and treatment of parasitic infections of the biliary system.

Pathological features

Gross characteristics: *F. buski* is an intestinal trematode characterised by a long oval and dorsoventral flat shape with a narrow anterior and wide posterior. This worm generally measures 20–75 mm in length, 8–20 mm in width, and 0.5–3 mm in thickness and is the largest human trematode. The oral sucker is proximally anterior, whereas the ventral sucker is close to the posterior of the oral sucker and is funnel-shaped, muscular, and 4–5 times larger than the oral sucker. The pharynx and oesophagus are short, while the intestinal branches extend posteriorly in a curved, wavy manner to the end of the worm body. The worms have two highly branched coralliform testes that are located anteroposteriorly in the posterior half of the worm body. The worms also have ovarian branches, with the uterus coiled between the ovary and ventral sucker. Moreover, the worms have Lowe's duct but lack seminal vesicles. Vitelline glands are developed and distributed along both sides of the worm. The genital cavity is located at the anterior edge of the ventral sucker and contains female and male genital pores [3, 17].

Microscopic examination has revealed that the body surface of the worm has a thick, well-striated cuticle and a loose subcutaneous layer below which a curved excreta-containing intestinal tract is visible. Vitelline glands are scattered evenly around the worm body. Reproductive organs with a large number of eggs are clearly visible in the central portion of the worm body [18].

Differential diagnosis

The eggs of *F. buski* can be easily morphologically distinguished from those of *Fasciola gigantica* and *Fasciola hepatica*.

① *F. gigantica* eggs: *F. gigantica* is a common human trematode that is mainly parasitic in the small intestine, with its eggs only found in the faeces. The eggs of *F. gigantica* were extremely large, with a size of 160–190 μm × 70–90 μm [19]. Thus, the eggs of *F. gigantica* can be distinguished from those of *F. buski* based on their size.

② *F. hepatica* eggs: The size of *F. hepatica* eggs range from 130 to 140 μm × 80–85 μm to 130–150 μm × 63–90 μm. *F. hepatica* parasitizes the hepatobiliary system, and its eggs are found in the bile and faeces of its host. These worms are known to infect humans occasionally. Some researchers have highlighted that the yolk granules of *F. hepatica* eggs are uniformly distributed in the yolk cells of the egg, whereas the yolk granules of *F. hepatica* eggs are concentrated around the yolk nucleus [20].

The adult worms of *F. buski* are primarily distinguished from those of *F. hepatica*. *F. hepatica* belongs to the *Fasciola* family and is one of the largest trematode species. The adult *F. hepatica* worms are long and narrow, with a conical proboscis at the anterior end of the body. Furthermore, the ventral sucker is small and less prominent and located at the level of the base of the cephalic cone. The intestinal branch has been observed to have numerous lateral branches. Additionally, these worms have two testes with extremely thin branches that cover approximately half of the worm body area, while the ovaries are small and thinly branched.

Treatment and prevention

Praziquantel is the first choice of treatment for ginger fascioliasis due to its advantages of high efficiency, low toxicity, easy use, low cost, and mild side effects [20, 21]. The measures for preventing *F. buski* infection include controlling the infection source, blocking the transmission route, and early diagnosis. Moreover, specific measures encompass strengthening the faecal management system, preventing the eggs from entering the water by restricting the transmission route, eliminating the intermediate host snails, and implementing critical measures such as avoiding consuming raw aquatic fruits such as water caltrops without washing and boiling in water [1].

In our case report, the patient had experienced recurrent upper abdominal pain as the primary symptom. Initially, the clinician considered inflammation or tumour as the primary cause and did not consider parasitic infection until hepatobiliary and pancreatic MRCP revealed tortuous streaks in the common bile duct. Therefore, imaging examination is vital in the initial diagnosis and treatment of this parasitic disease. Subsequently, the clinician carefully reviewed the patient's life history and learned that the patient had a history of consuming raw pig blood, which further supported the possibility of parasitic infection. In contrast to other cases reported in the literature, this extremely rare case of *F. buski* infection was caused by consuming raw pig blood. Therefore, along with aquatic plants such as water nuts as transmission vectors, the blood of the final host pig, which contains the eggs or adult worms, can also act as an intermediate host for human infection with *F. buski*. This finding emphasises the need to pay attention to dietary hygiene and to avoid consuming raw food to reduce the likelihood of parasitic infection. Although the diagnosis of *F. buski* infection may initially rely on the clinical history, past life history, preliminary imaging screening, and early diagnostic tests, the final diagnosis depends on the pathological examination results.

Conclusion

Biliary tract infection with *F. buski* is an extremely rare parasitic disease. This case report describes an extremely rare case of *F. buski* infection of the biliary tract caused by consuming raw pig blood. The clinical features, common diagnostic methods, imaging and pathological

features, differential diagnosis, treatment, and prognosis of *F. buski* infection were reviewed to enhance the current knowledge of this rare condition.

Abbreviations

F.buski	Fasciolopsis buski
MRCP	magnetic resonance cholangiopancreatography

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Author contributions
Writing–original draft: S L and XX T, Writing & editing: S L, JJ W. XX T and T X prepared all figures.All the authors have read & approved the final manuscript.

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Data availability
No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate
This case report was approved by the Ethics Committee of the Affiliated Hospital of Zunyi Medical University. Written informed consent was obtained from the patient and the patient's family for publication of this clinical case report.

Consent for publication
Written informed consent was obtained from the patient and the patient's family for publication of this case report and any accompanying images.

Competing interests
The authors declare no competing interests.

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