

Original Article

Inflammatory pseudotumor of the liver caused by a migrated fishbone misdiagnosed as cholangiocarcinoma: a case report and literature review

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Abstract: Inflammatory pseudotumor of the liver secondary to a migrated fishbone is very rare. A 51-year-old man presented to our hospital with a 3-year history of vague epigastric pain, anorexia, and weight loss. Conventional abdominal ultrasound showed a hypoechoic mass in the left lobe of the liver, and contrast-enhanced ultrasonography indicated the typical enhanced pattern of fast-in-fast-washout. Both magnetic resonance imaging and computed tomography revealed a liver mass with progressive enhancement. Unenhanced computed tomography showed homogeneous hypo-attenuation with central calcification. Hematological examination revealed leukocytosis with granulocytosis. The initial diagnosis was cholangiocarcinoma accompanying inflammation. However, the pathological results revealed chronic non-specific inflammation on day 17 after admission. The patient's symptoms recurred despite antibiotic treatment. Laparotomy was performed to establish a clear diagnosis, which revealed a foreign body (fishbone) embedded in the left lobe of the liver. Another biopsy confirmed an inflammatory lesion. The definitive diagnosis was inflammatory pseudotumor of the liver caused by a migrated fishbone. Surgeons and radiologists should be aware of liver inflammatory pseudotumor as a differential diagnosis in patients presenting with a hepatic mass, but with normal serum tumor markers and no specific history, especially in cases where there is discordance between imaging and histological results.

Keywords: Inflammatory pseudotumor, liver, foreign body, contrast-enhanced ultrasonography, computed tomography, magnetic resonance imaging

Introduction

Inflammatory pseudotumors (IPT) of the liver are very rare benign lesions of uncertain etiology. IPT of the liver caused by ingested foreign bodies via perforation of the gastrointestinal tract is extremely uncommon. To the best of our knowledge, only one previous case has been reported with accompanying computed tomography (CT) imaging features [1]. The lack of characteristic clinical and radiological features means that IPT mimicking a malignant liver neoplasm, such as metastatic cancer or cholangiocarcinoma, may be misdiagnosed and its treatment delayed [2-5]. Early diagnosis and removal of the foreign body are crucial.

We report a case of IPT of the liver induced by an ingested foreign body and initially misdiag-

nosed as a cholangiocarcinoma. We also reviewed the relevant literature and retrospectively analyzed previous imaging features, with a focus on contrast-enhanced ultrasonography (CEUS) and magnetic resonance imaging (MRI) features of liver IPT. This case highlights the fact that the diagnosis of foreign body ingestion remains a confounding problem, despite the availability of powerful imaging methods.

Case report

No ethical approval was required for this case report.

A 51-year-old man presented to the hepatic surgical unit of the Third Affiliated Hospital of Sun Yat-sen University, China, on August 10, 2015, with a 3-year history of intermittent epigastric-

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Table 1. The laboratory data on the second day of admission

Measurement	Value	Reference range
Hematological panel		
White blood cells ($\times 10^9/L$)	17.67	3.5~9.5
Neutrophilic granulocyte (%)	82.2	0.400~0.750
C-reactive protein (mg/L)	25.20	0.0~6.0
Platelets ($\times 10^9/L$)	618	100~350
Serum chemistry results		
Aspartate aminotransferase (U/L)	22	15~40
Alanine aminotransferase (U/L)	25	3~35
Alkaline phosphatase (U/L)	191.0	45~125
Glutamyltran-speptidase (U/L)	168	10~60
Total bilirubin (umol/l L)	13.80	4.0~23.9
Serum ferritin (ng/mL)	1217.8	22~232
Alpha-fetoprotein (ng/mL)	1.8	0~8.1
Carcinoembryonic antigen (CEA) (ng/ml)	0.4	0-5.0
Carbohydrate antigen 19-9 (CA 19-9) (U/ml)	11.92	0-35
Carbohydrate antigen 125 (CA-125) (U/ml)	8.6	0-35
Carbohydrate antigen 15-3 (CA 15-3) (U/ml)	4.0	0-35

distension pain (progressively worsening for 1 month), anorexia, and weight loss of almost 7.5 kg. On hospital admission, physical examination showed a soft abdominal wall but mild tenderness in the right upper quadrant. Other vital signs were stable, with no fever, chills, asthenia, nausea, or vomiting. The patient had no history of hepatitis, and serological tests for hepatitis A, B, C, and E were normal. The results of laboratory examinations on the second day after admission, including blood counts, liver function, and serum chemistry, are shown in **Table 1**. Two stool tests for opisthorchiasis were negative and his coagulation functions were normal. Upper gastrointestinal endoscopy at the local hospital revealed chronic superficial gastritis. Chest radiography was not contributory, showing no free gas in the abdomen on the first day after admission.

Conventional ultrasound (US) on the second day after admission revealed a solid heterogeneous, ill-defined hypoechoic mass in the left lobe of the liver, with a maximum diameter of 8.9×6.8 cm. CEUS performed immediately after conventional US showed arterial phase high enhancement, portal venous phase, and delay-period low enhancement, and the typical sign of fast-in-fast-washout (**Figure 1**).

Dynamic gadolinium-enhanced MRI performed on the fifth day after admission mainly showed

hypointensity on fat-suppressed T1-weighted and hyperintensity on fat-suppressed T2-weighted images. After gadolinium injection, the arterial phase showed homogeneous slight enhancement, with further enhancement at the portal and delay phases. Dynamic-enhanced MRI revealed progressive enhancement (**Figure 2**).

The combined results of physical and laboratory examinations, CEUS, and MRI were highly suspicious for cholangiocarcinoma accompanying an inflammatory lesion. The patient received antibiotic treatment with moxifloxacin injections (0.4 g, once daily) for 11 days, and polyene phosphati-

dylcholine injection for 1 week (0.464 g, once daily) for temporary liver protection.

The patient underwent US-guided fine needle aspiration to obtain a sample for microbiological analysis. Histological results on day 17 after admission revealed densely fibrous tissue infiltrated by inflammatory cells comprising lymphocytes, plasma cells, neutrophils, and eosinophils. Immunohistochemical examination was positive for vimentin, partially positive for CD68, and negative for CK, EMA and ALK, indicating an inflammatory lesion. The patient's epigastric-distension pain ameliorated and his liver-function indexes normalized, and he was therefore discharged on day 18 after admission.

However, the patient was re-hospitalized 2 months later because of recurrent epigastric-distension pain. Contrast-enhanced CT on the second day after admission showed a large low-density mass in the left lobe of the liver, with a maximal diameter of approximately 8.6×7.0 cm, demonstrating progressive enhancement. Careful examination of the original plain CT images revealed a high-density, stick-shaped object situated in the mass, adjacent to the hepatic falciform ligament. Injection of contrast medium revealed the periphery of the lesion to be non-homogeneously enhanced and isodense in the late phase, compared with

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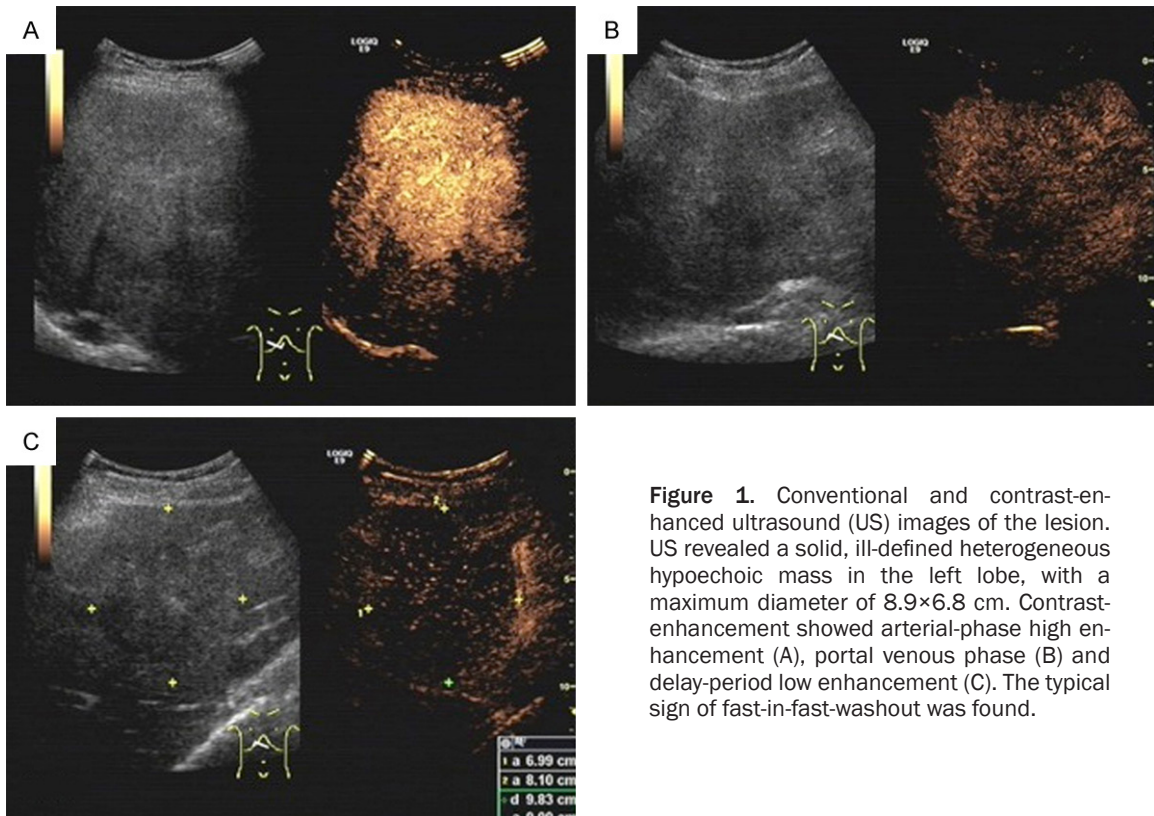


Figure 1. Conventional and contrast-enhanced ultrasound (US) images of the lesion. US revealed a solid, ill-defined heterogeneous hypoechoic mass in the left lobe, with a maximum diameter of 8.9×6.8 cm. Contrast-enhancement showed arterial-phase high enhancement (A), portal venous phase (B) and delay-period low enhancement (C). The typical sign of fast-in-fast-washout was found.

the surrounding normal liver tissue. The biliary duct was not dilated, and there was no shrinkage of the adjacent liver capsule (**Figure 3**). The possibility of cholangiocarcinoma could not be ruled out on the basis of these additional findings.

Laparotomy was eventually performed to arrive at a clear diagnosis and to decide on the appropriate treatment. Inflammation and adhesions between the omentum and gallbladder, gastric antrum, and hepatic hilar were detected during surgery, and a foreign body (later recognized as a fishbone) was found embedded in the left lobe of the liver. No obvious signs of gastrointestinal-tract perforation were found. A cholecystectomy was performed, the fishbone was removed, and a tumor biopsy confirmed it as an inflammatory lesion (**Figure 4**). The definitive diagnosis was thus of an IPT of the liver. The patient was symptom-free 2 months postoperatively, and a subsequent CT scan confirmed obvious regression of the liver lesion.

Discussion

IPTs of the liver are rare clinical and pathological entities, first described in 1953 by Pack and

Baker in a patient after right hepatic lobectomy [6]. Schmid reported that the incidence of IPT was relatively high in individuals from Southeast Asia (54.7%) [7], with a male preponderance. Histologically, these tumors are characterized by a well-circumscribed mass composed of densely hyalinized collagenous tissue infiltrated by a variety of cells, including plasma cells, enigmatic plump spindle cells, monocytes, and lymphocytes [8].

However, the etiopathogenesis of these tumors is uncertain. Horiuchi et al. suggested that they may have a septic origin from an aberrant inflammatory reaction to microorganisms migrating from the large bowel [9], while further studies are needed to verify the suggestion of a causative role of Epstein-Barr virus [10]. IPTs of the liver are clinically classified into different types according to their etiology [11].

IPTs of the liver caused by perforation of the gastrointestinal tract as a result of ingested foreign bodies are extremely uncommon, especially secondary to the migration of a fishbone [1, 12], as in the current patient. During surgery, inflammation and adhesions were found between the gastric antrum and hepatic hilar,

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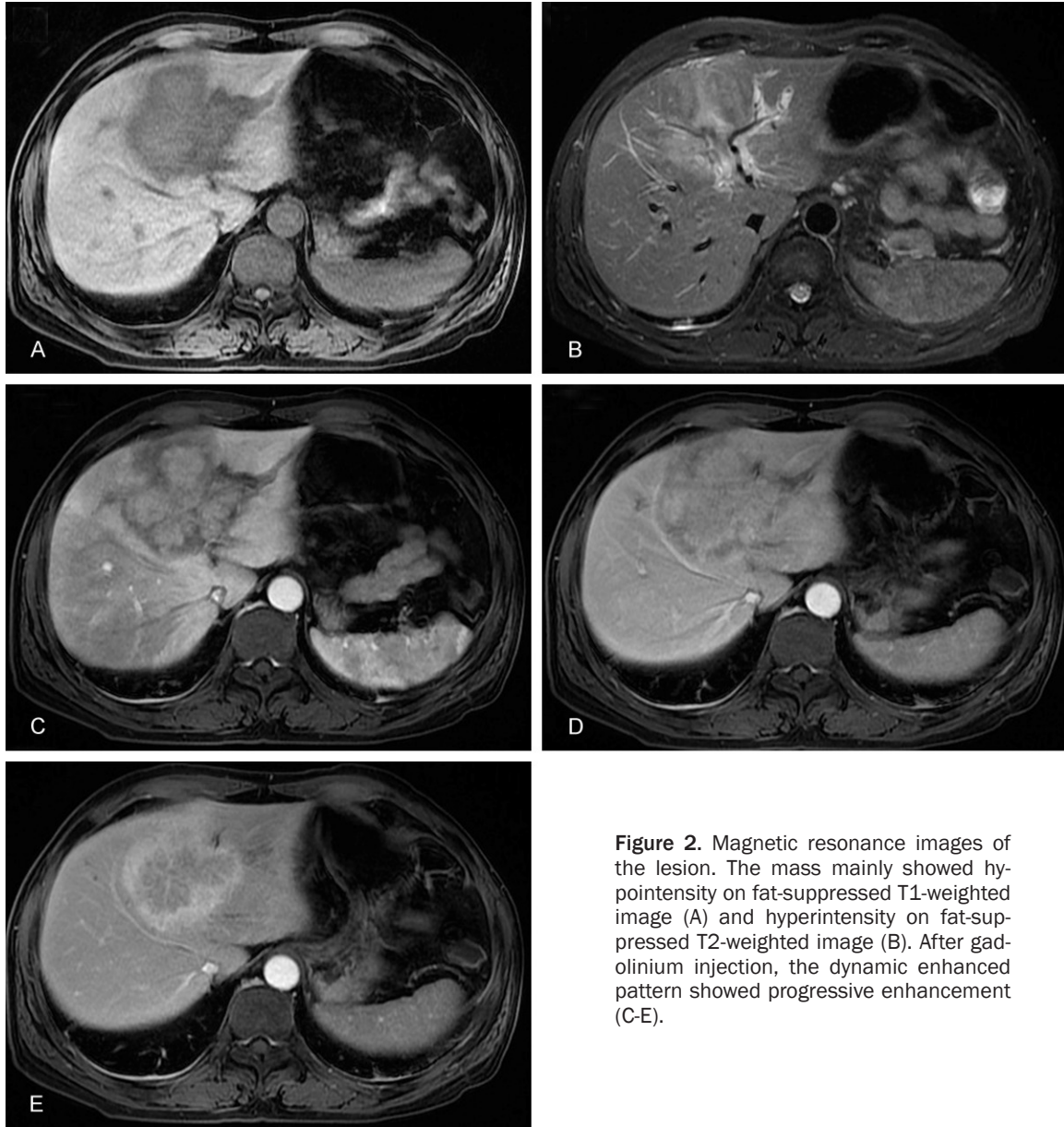


Figure 2. Magnetic resonance images of the lesion. The mass mainly showed hypointensity on fat-suppressed T1-weighted image (A) and hyperintensity on fat-suppressed T2-weighted image (B). After gadolinium injection, the dynamic enhanced pattern showed progressive enhancement (C-E).

suggesting that the fishbone was most likely to have reached the liver by penetration through the wall of the gastric antrum.

Less than 1% of ingested foreign bodies lead to gastrointestinal perforation [13-15], usually sharp objects such as fish and chicken bones, toothpicks, sewing needles, and dental plates [16]. Perforation usually occurs at the level of the stomach or duodenum [13, 14]. The ingested object then frequently lodges in the left lobe of the liver after migrating from the intestinal lumen [14, 17]. In these respects, our patient was similar to other reported cases.

However, most previous cases of fishbone migration to the liver have presented with hepatic abscesses [13-17], while the current patient just presented with an inflammatory pseudotumor, which represents an extremely rare manifestation.

Most previously reported patients with IPT of the liver presented with non-specific symptoms, such as abdominal pain, anorexia, and weight loss, as in the present case. Critically, most patients do not remember swallowing a foreign body, and the lack of this important information makes an accurate diagnosis diffi-

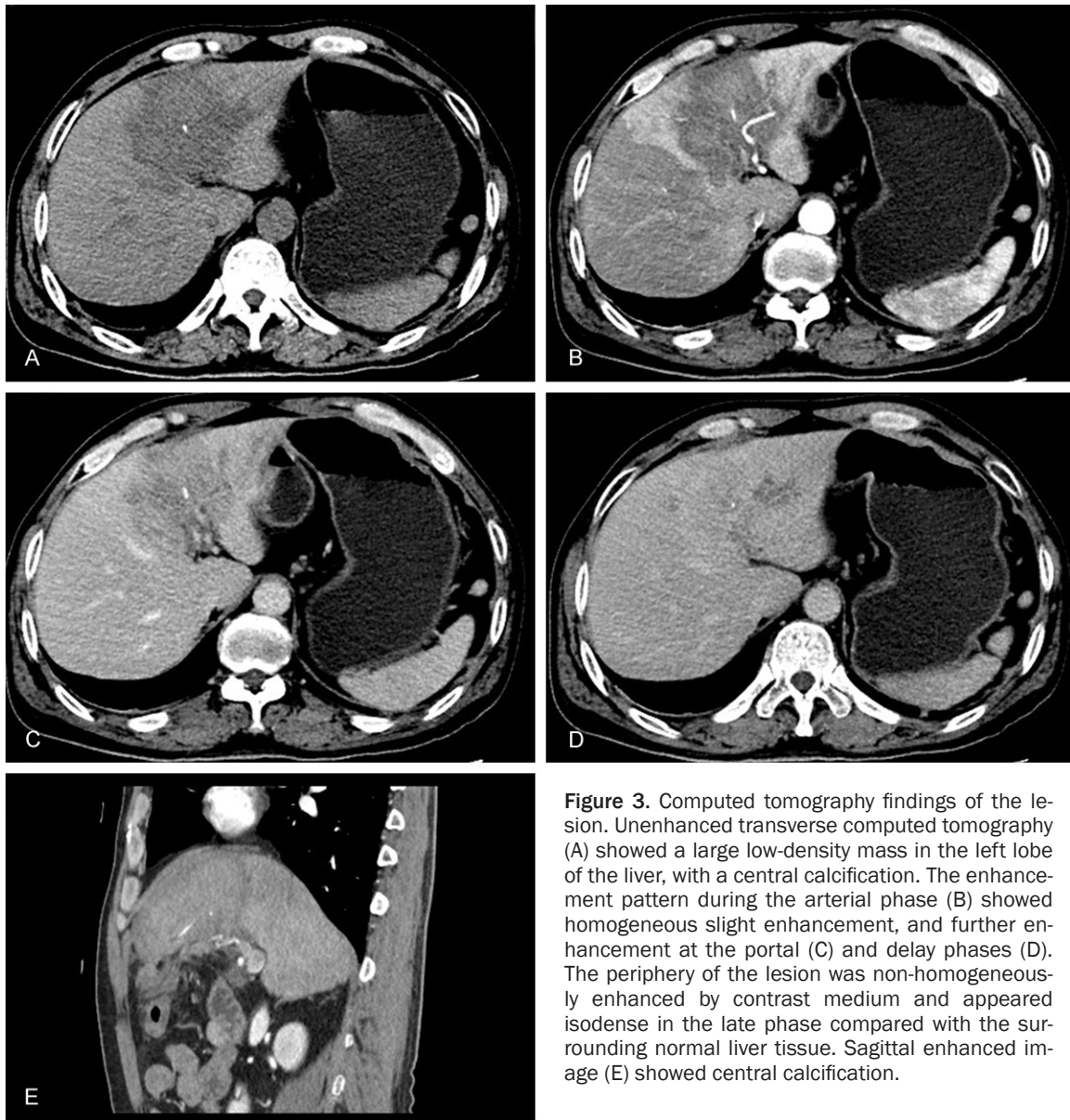


Figure 3. Computed tomography findings of the lesion. Unenhanced transverse computed tomography (A) showed a large low-density mass in the left lobe of the liver, with a central calcification. The enhancement pattern during the arterial phase (B) showed homogeneous slight enhancement, and further enhancement at the portal (C) and delay phases (D). The periphery of the lesion was non-homogeneously enhanced by contrast medium and appeared isodense in the late phase compared with the surrounding normal liver tissue. Sagittal enhanced image (E) showed central calcification.

cult. Furthermore, many foreign bodies are not radiopaque, are too small to be seen on radiographs, or the small perforations have already healed; the presence of free intraperitoneal gas is therefore uncommon and plain radiographic tests are of limited assistance [18].

US and CT are the preferred techniques when there is a clinical suspicion of this rare diagnosis. In most cases, the objects are hypoechoic on US and have low-density characteristics on CT. Although CEUS has been shown to be an accurate method for diagnosing focal liver lesions in large multicenter studies, with simi-

lar performances to contrast-enhanced CT and MRI [19, 20], CEUS in our patient showed typical fast-in-fast-washout, suggesting a probable cholangiocarcinoma [21]. Contrast-enhanced CT and MRI showed a large mass in the left lobe of the liver with progressive enhancement, which was also a typical imaging feature of cholangiocarcinoma [22]. The preoperative diagnosis was thus highly suggestive of liver cholangiocarcinoma. However, histological results revealed an inflammatory lesion. We ultimately performed a laparotomy to confirm the diagnosis, which was IPT secondary to migration of a fish bone.

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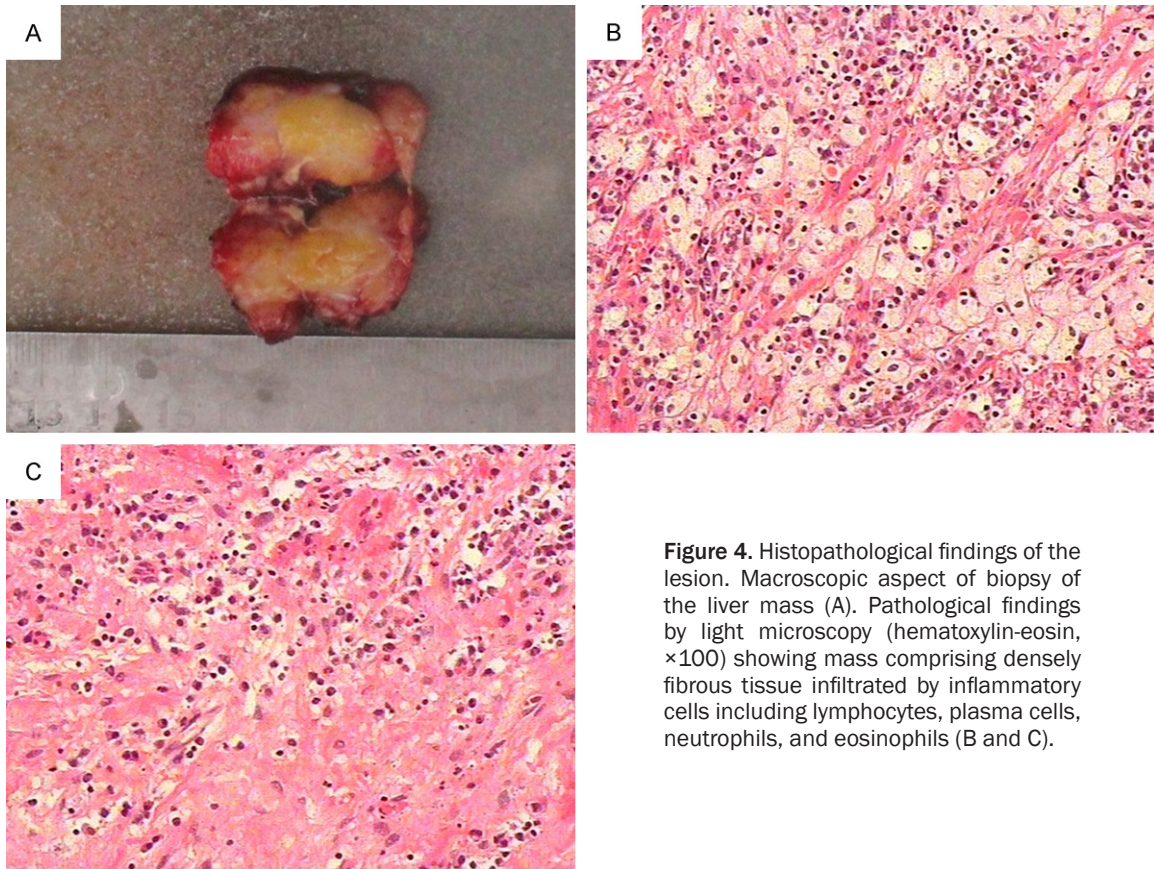


Figure 4. Histopathological findings of the lesion. Macroscopic aspect of the liver mass (A). Pathological findings by light microscopy (hematoxylin-eosin, $\times 100$) showing mass comprising densely fibrous tissue infiltrated by inflammatory cells including lymphocytes, plasma cells, neutrophils, and eosinophils (B and C).

The therapeutic management of IPTs of the liver varies according to their etiology. Conservative management, such as with antibiotics, anti-inflammatories, and even corticosteroids, can be used until resolution [11]. However, in the case of IPTs secondary to the migration of a foreign body, removal of the foreign body is crucial, and surgical resection may be indicated if unresolved symptoms persist despite conservative treatment, or if the possibility of malignancy cannot be ruled out [11], as in the current case.

Retrospectively, the patient revealed leukocytosis with granulocytosis, CT showed contrast-enhancement of the periphery of the lesion, which appeared isodense in the late phase, and there was no biliary duct dilatation or shrinkage of the adjacent liver capsule. These features pointed to the possibility of IPT as a differential diagnosis.

In conclusion, IPT of the liver secondary to a migrated foreign body is a rare entity, the definitive diagnosis of which often requires histopathological confirmation. Clinical features and

the results of imaging and laboratory examinations may not aid its diagnosis, and it may be misdiagnosed preoperatively as a malignant tumor. Delayed diagnosis or misdiagnosis is thus a major concern, despite extensive patient workup, because early diagnosis and removal of the foreign body is crucial. Obtaining a correct diagnosis relies on increased awareness of surgeons and radiologists, and the need to include liver IPT due to a migrated foreign body as a possible differential diagnosis in patients with a hepatic mass, normal serum tumor markers, and no specific history, especially in the event of apparently conflicting imaging and histological results. Finally, adequate history taking and improved communication between members of the multi-disciplinary team are also needed to improve the accurate diagnosis of this rare condition.

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Disclosure of conflict of interest

None.

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