

Laparoscopic resection of benign schwannoma in the hepatoduodenal ligament: A case report and review of the literature

LIANG TAO^{1*}, SHAOYAN XU^{2*}, ZHIGANG REN², YITING LU¹, XIAOWU KONG¹,
XIAOHUI WENG¹, ZHIJIE XIE³ and ZHIMING HU³

¹Department of General Surgery, The Third People's Hospital of Haining, Jiaxing, Zhejiang 314408;

²Department of Hepatobiliary and Pancreatic Surgery, The First Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou, Zhejiang 310003; ³Department of Hepatobiliary and Pancreatic Surgery, Zhejiang Provincial People's Hospital, Hangzhou, Zhejiang 310014, P.R. China

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Abstract. Schwannomas are mesenchymal neoplasms with a low malignant potential, which arise from Schwann cells. The tumors can occur in most parts of the body; however, the head, neck and flexor surfaces of the extremities are the most common locations. Schwannomas occurring in the hepatoduodenal ligament are extremely rare. To the best of our knowledge, only two cases of schwannoma in the hepatoduodenal ligament have been reported in the literature, and treatment of such cases by laparoscopic surgery has not yet been reported. The present study reports a case of schwannoma in the hepatoduodenal ligament in a 50-year-old male patient. Physical and laboratory examinations showed no abnormal results. Ultrasound and computed tomography failed to definitively diagnose the mass and identify its location. During laparoscopic surgery, a mass was identified in the hepatoduodenal ligament and was completely removed. The gross specimen was a 4.5x2.5x2.5-cm localized mass, yellowish-white in color. Microscopic examination revealed that the tumor was mainly composed of spindle-shaped cells and no atypical cells were identified. Immunohistochemical staining showed a strong positive S-100 protein reaction, whereas cluster of differentiation 34 and epithelial membrane antigen were negative. The final diagnosis of the lesion was benign schwannoma of the hepatoduodenal ligament. The patient was followed-up for 7 months and, at the time of writing, was healthy and without any complications. The aim of the present study was to describe a rare case of hepatoduodenal ligament schwannoma in a

50-year-old male patient, and present a review of the literature. To the best of our knowledge, this is the first case of hepatoduodenal ligament schwannoma treated by laparoscopic surgery.

Introduction

Schwannomas are neurogenic tumors that originate from Schwann cells, the cells of the peripheral nervous system (1). Schwannomas have been shown to occur in all ethnic groups and at all ages worldwide, with >90% of the tumors being benign (2). Schwannomas account for ~5% of benign soft-tissue neoplasms (3). Schwannomas may develop in any part of the body, but the most common sites include the head, neck and flexor surfaces of the extremities (4). Generally, schwannomas have a single place of origin, but 10% originate from multiple locations (5). Surgery is the most common treatment for schwannomas. Benign schwannomas rarely recur subsequent to complete surgical resection. Long-term survival can be expected in the majority of patients (4). Peripheral nerve sheath tumors account for 2-6% of all gastrointestinal stromal tumors; however, schwannomas occurring in the hepatoduodenal ligament are extremely rare. To the best of our knowledge, only two such cases have been reported in the literature (6,7). One patient was female and the other patient was male, aged 62 and 29 years old, respectively (6,7). As schwannomas of the hepatoduodenal ligament are normally asymptomatic and often discovered incidentally, preoperative diagnosis is considerably challenging. The two patients each accepted more than two imaging examinations prior to treatment by laparotomy. The lesions were solitary and found in the hepatoduodenal ligament in the two patients. Finally, the patients were definitively diagnosed with schwannoma by pathological examination (6,7). Following complete tumor excision, patients with benign schwannomas generally have a good prognosis. The present study describes the case of a hepatoduodenal ligament schwannoma in a 50-year-old male patient, and presents a review of the literature. To the best of our knowledge, this was the first time laparoscopic surgery was used for the treatment of a benign schwannoma of the hepatoduodenal ligament.

Correspondence to: Dr Zhiming Hu, Department of Hepatobiliary and Pancreatic Surgery, Zhejiang Provincial People's Hospital, 158 Shangtang Road, Hangzhou, Zhejiang 310014, P.R. China
E-mail: xsy200888@126.com

*Contributed equally

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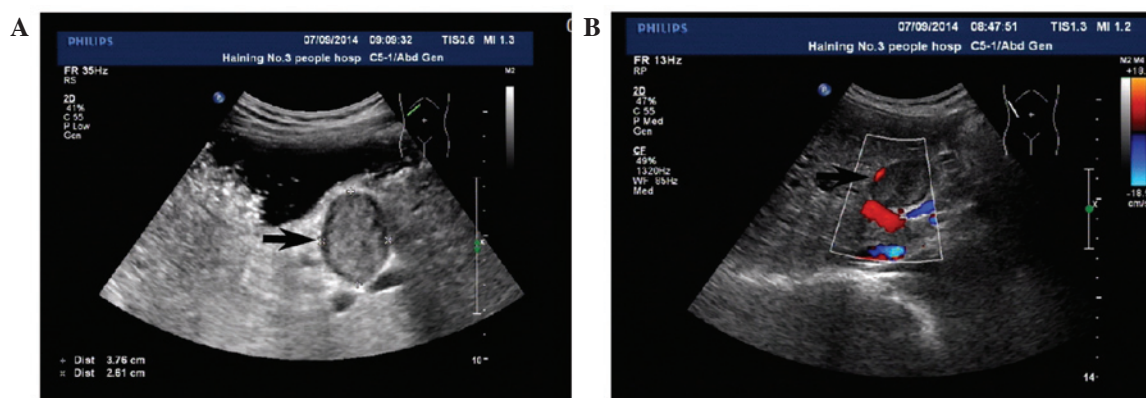


Figure 1. US and Color Doppler US. (A) Conventional US showing a mass (arrow) measuring 3.8x2.6 cm in the space between the porta hepatis and the stomach. (B) Color Doppler US showing no blood supply in the mass (arrow). US, ultrasound.

Case report

A 50-year-old male patient was referred to the Department of General Surgery, The Third People's Hospital of Haining (Jiaxing, Zhejiang, China), presenting with pain in the right abdomen following trauma, on July 11, 2014. Physical examination revealed a soft and flat abdomen, and no enlarged lymph nodes were identified in the examinable sites. The patient's family had no history of any specific disease; however, 3 years prior to admission, a mass was incidentally detected in the hepatogastric ligament of the patient by ultrasound (US; Philips IU22; Philips, Amsterdam, Netherlands), during a health examination. As no abnormal symptoms had presented at the time, the patient did not receive any treatment. During the period of hospitalization in July 2014, an US revealed a well-defined hypodense lesion in the space between the porta hepatis and the stomach, which measured 3.6x2.4 cm (Fig. 1A). Color Doppler US (Philips IU22) showed no flow signals within the mass (Fig. 1B).

Subsequently, an abdominal computed tomography (CT) scan (GE Brightspeed Elite; Hangwei Tongyong Electric Medicine System Co., Ltd., Beijing, China) was performed to examine the region. The non-enhanced CT scan showed a well-defined round soft-tissue mass without internal calcification or liquefaction in the space between the porta hepatis and the stomach. The mass measured 3.8x3.0x2.5 cm, had a uniform density and was surrounded by a clear tunica (Fig. 2A). On the contrast-enhanced CT scan, the mass showed no evident enhancement in the arterial phase (Fig. 2B). Based on the findings of the CT scan, the diagnosis of gastric stromal tumor was primarily considered. However, since the mass had been growing slowly for 3 years and possessed a clear tunica, the mass was then considered to be a benign tumor, which may be removed.

Laboratory results were as follows: Red blood cells, $4.38 \times 10^{12}/l$ (normal values, $4.30-5.80 \times 10^{12}/l$); white blood cells, $5.60 \times 10^9/l$ (normal values, $4.00-10.00 \times 10^9/l$); hemoglobin, 131 g/l (normal values, 130-175 g/l); lactate dehydrogenase, 146 international units (IU)/l (normal values, 80-240 IU/l); total protein, 58.10 g/l (normal values, 65.00-85.00 g/l); total bilirubin, 10.80 $\mu\text{mol/l}$ (normal values, 0.00-21.00 $\mu\text{mol/l}$); conjugated bilirubin, 5.70 $\mu\text{mol/l}$ (normal values, 0.00-10.00 $\mu\text{mol/l}$); bile acids, 2.40 $\mu\text{mol/l}$ (normal values, 0.00-12.00 $\mu\text{mol/l}$); aspartate aminotransferase, 18 IU/l (normal values, 15-45 IU/l); alanine transaminase, 16 IU/l (normal values, 9-60 IU/l);

γ -glutamyl transpeptidase, 34 U/l (normal values, 10-60 IU/l); alkaline phosphatase, 67 U/l (normal values, 45-125 IU/l); amylase, 77 U/l (normal values, <100 IU/l); antimitochondrial antibody, negative; α -fetoprotein, <20 ng/ml (normal values, <20 ng/ml); carcinoembryonic antigen, 0.67 ng/ml (normal values, <5.50 ng/ml); carbohydrate antigen 19-9, 4.93 U/ml (normal values, <37.00 U/ml); cancer antigen 125, <4.00 U/ml (normal values, <35.00 U/ml).

Subsequent to obtaining patient consent, laparoscopic surgery was performed (Laparoscope operating system, 22202011V110; Karl Storz GmbH & Co. KG, Tuttlingen, Germany). The patient was placed in a horizontal position. Following the successful induction of anesthesia, the skin of the surgery field was conventionally sterilized, and sterile drapes were whisked onto the patient's body. Subsequently, a ~2.0 cm long incision was made in the superior border of the umbilicus. A CO₂ pneumoperitoneum was set up with 15 mmHg intra-abdominal pressure using a veress needle (26120J; Karl Storz GmbH & Co. KG, Tuttlingen, Germany), and a 1.1 cm Trocar puncture (30103MP; Karl Storz GmbH & Co. KG) was made to insert a laparoscopic lens (26003BA; Karl Storz GmbH & Co. KG). Under direct vision, 3 holes were made beneath the xiphoid process and left and right upper abdomen, and the ultrasonic scalpel (GEN04; Johnson & Johnson Medical (China) Ltd., Shanghai, China), dissecting forceps (33321ML; Karl Storz GmbH & Co. KG), scratch-free grasping forceps (33321R; Karl Storz GmbH & Co. KG) were inserted. During laparoscopic exploration, a mass surrounded by a fibrous capsule was located in the hepatoduodenal ligament, medial to the gallbladder and near the upper antrum (Fig. 3A and B). No invasion of the surrounding tissue was observed and the biliary ducts were not dilated. The mass was completely resected. The capsule was then removed and the gross specimen was described as a 4.5x2.5x2.5-cm sized localized mass, yellowish-white in color (Fig. 3C and D). Following postoperative pathological analysis of an intraoperative frozen section, the mass was diagnosed as a schwannoma. Postoperatively, the patient had a smooth recovery and left the hospital 3 days later.

The tissue section was analyzed at the Department of Pathology, Zhejiang Provincial People's Hospital (Hangzhou, Zhejiang, China). Microscopically, the tumor mainly consisted of spindle-shaped cells, and no atypical cells or signs of malignancy were observed (Fig. 4A). Immunohistochemical

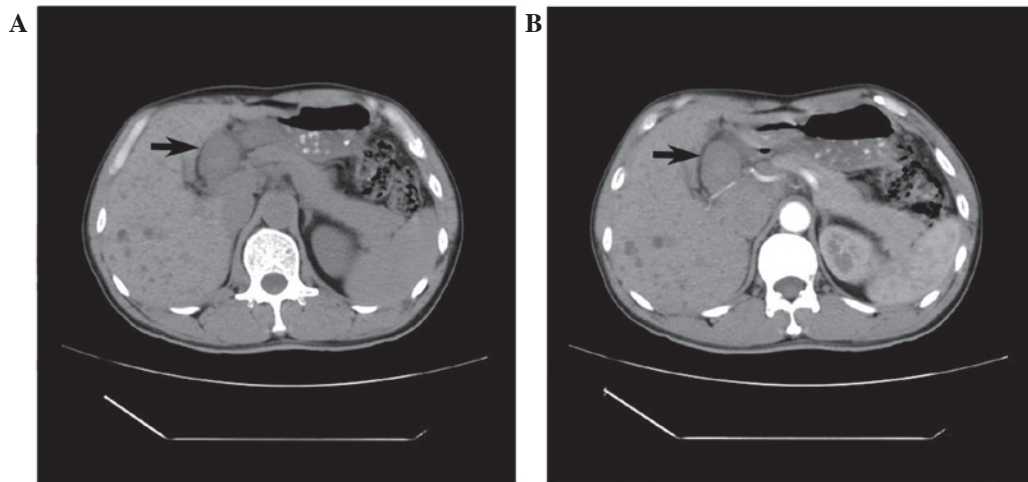


Figure 2. CT scans. (A) Non-enhanced CT scan showing a uniform density mass in the space between the porta hepatis and the stomach (arrow); (B) Enhanced CT scan showing no obvious enhancement in the arterial dominant phase (arrow). CT, computed tomography.

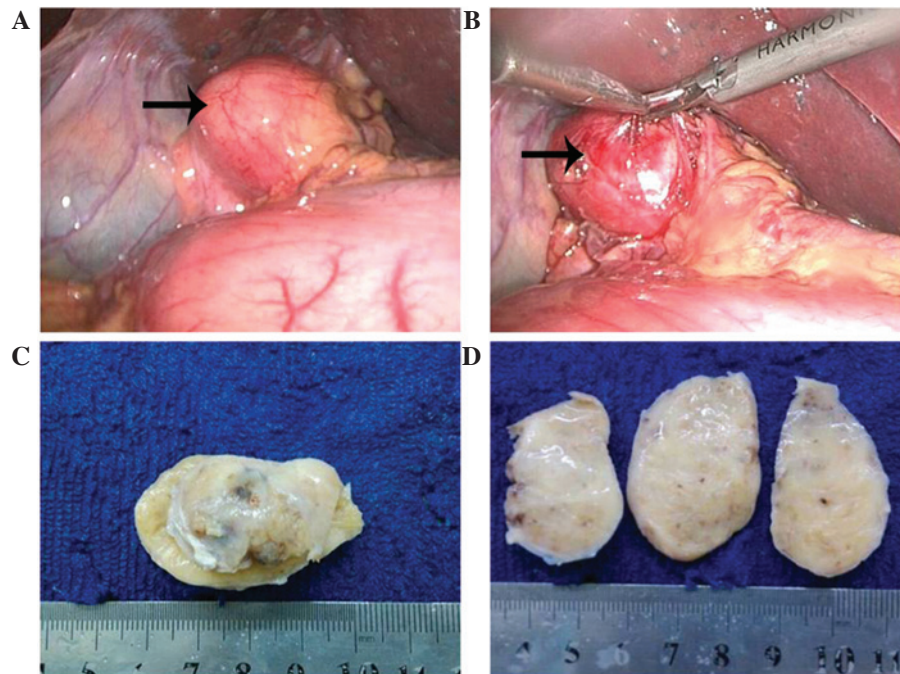


Figure 3. Intraoperative and macroscopic findings. Intraoperatively, (A) the tumor was found to be located in the hepatoduodenal ligament, (B) with no invasion of the surrounding tissue (arrows). (C) Macroscopic findings showed a capsulated yellowish-white mass. (D) The capsule was then removed and the gross specimen was described as a 4.5x2.5x2.5-cm localized mass.

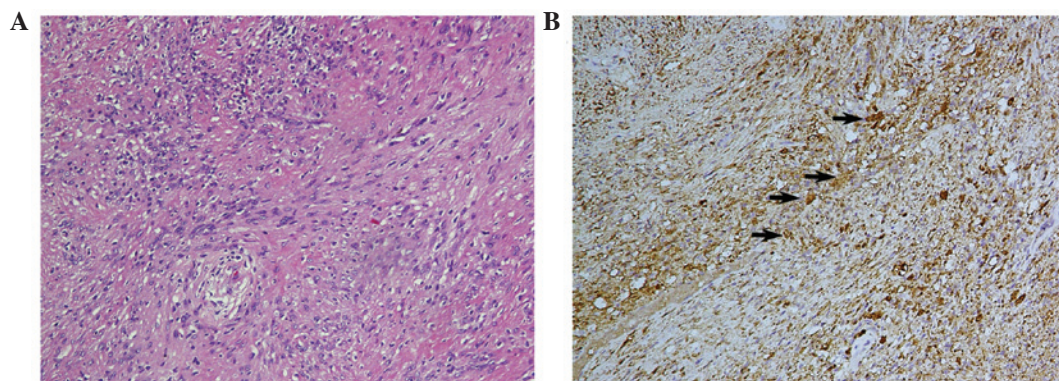


Figure 4. Pathological examination. (A) Microscopic examination showed that the tumor was mainly composed of spindle-shaped cells (stain, hematoxylin and eosin). (B) Immunohistochemical staining showed a strong tumor positivity for S-100 protein (arrows) (magnification, x10; lens diameter, 20 mm).

analysis showed that the tumor cells were vimentin- and S-100 protein-positive (Fig. 4B), and epithelial membrane antigen- and cluster of differentiation 34-negative. The final diagnosis of the tumor was schwannoma. The patient was followed-up for 7 months and, at the time of writing, was healthy and without any complications.

The present study was approved by the Ethics Committee of the Third People's Hospital of Haining (Jiaxing, Zhejiang, China).

Discussion

Schwannomas originate from the sheaths of the peripheral nerves and have been shown to occur in all ethnic groups worldwide (2). Secondary degenerative changes of schwannomas, including cyst formation, calcification, hemorrhage, and hyalinization, sometimes appear (8). Schwannomas occur in most parts of the body, but the head, neck and flexor surfaces of the extremities are the most common sites (1). They rarely occur in the retroperitoneum, accounting for 6% of all primary retroperitoneal tumors (9,10). The occurrence of schwannomas in the gallbladder (11), gastroduodenal ligament (12), pancreas (13), bowel mesentery (14) or colon (15) is extremely rare. To the best of our knowledge, only two cases of schwannoma involving the hepatoduodenal ligament have been reported (6,7). The clinical characteristics of the aforementioned cases are listed in Table I. These tumors are usually asymptomatic and often incidentally discovered. Due to their low incidence and, therefore, lack of adequate understanding of the mechanism of this type of tumor, forming a preoperative diagnosis is challenging.

Prior to treatment, comprehensive imaging modalities, including US, CT and magnetic resonance imaging (MRI), should be used to establish a probable diagnosis and determine the lesion limits; however, these imaging techniques rarely provide a definitive diagnosis, due to a lack of distinguishing features of these tumors on imaging scans. Generally, schwannomas manifest as well-defined, hypoattenuating masses on non-enhanced CT scans, and on contrast-enhanced CT scans show peripheral enhancement with an irregular pattern. Delayed peripheral enhancement until the late venous phase reflects a fibrous capsule and an internal fibrillary element on CT scan (11). MRI comprises another useful tool for determining the nature of the tumor. On MRI scans, schwannomas are usually presented as masses with a low signal intensity on T1-weighted images, and inhomogeneous high signal intensity on T2-weighted images (16).

Pathology is important for a definitive diagnosis. Histologically, schwannomas consist of compact cellular lesions. Microscopically, schwannomas are encapsulated tumors with a biphasic growth pattern consisting of Antoni A (interlacing and cellular fascicles) and Antoni B (less cellular and myxoid) regions. In addition, typical Verocay bodies in the Antoni A areas may be detected using hematoxylin and eosin staining. In immunohistochemistry, positive uniform S-100 staining may be detected, which is an indicator of schwannoma (17,18). In the present case, the histological findings were predominantly cellular Antoni A areas and immunohistochemistry showed a strong positive staining of the tumor cells for S-100 protein, which supported the benign nature of the schwannoma in the present case.

Table I. Clinical characteristics of 3 patients with benign schwannomas in the hepatoduodenal ligament.

Author, year	Gender	Age, years	Symptoms	Imaging technique	Location	Number	Size, cm	Preoperative diagnosis	Treatment	Follow-up, months	Status	Ref.
Nagafuchi <i>et al</i> , 1988	F	62	Asymptomatic	US, CT, CA MRI, ERC	Hepato-duodenal ligament	Solitary	9x5x4.5	NA	Laparotomy	26	Survived	(14)
Pinto <i>et al</i> , 2011	M	29	Asymptomatic	US, MRI endoscopy, biopsy	Hepato-duodenal ligament	Solitary	4.5x2.9	Spindle cell neoplasia or stromal tumor	Laparotomy	NA	NA	(15)
Present case	M	50	Right abdominal pain	US, CT, CECT	Hepato-duodenal ligament	Solitary	4.5x2.5x2.5	Stromal tumor	Laparoscopic surgery	7	Survived	

F, female; M, male; NA, not available; US, ultrasound; CT, computed tomography; CECT, contrast-enhanced computed tomography; MRI, magnetic resonance imaging; ERC, endoscopic retrograde cholangiogram; CA, celiac angiography.

In conclusion, surgical intervention may be the optimal treatment for schwannomas, as surgery may be used to determine the location and nature of the lesion and to successfully treat it. In the present case, a large mass had developed; however, no resultant symptoms were present prior to trauma. Definitively determining the nature of the tumor preoperatively was challenging, despite the use of imaging modalities. To the best of our knowledge, this was the first case of benign schwannoma of the hepatoduodenal ligament treated by laparoscopic surgery. Following complete tumor excision, patients with benign schwannomas generally have a good prognosis.

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