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**Columns:** **CASE REPORT**

**Rare cause of abdominal incidentaloma: Hepatoduodenal ligament teratoma**

Jeismann VB *et al.* Hepatoduodenal ligament teratoma as abdominal incidentaloma

Vagner Birk Jeismann, Rodrigo Blanco Dumarco, Celso di Loreto, Ricardo Correa Barbuti, José Jukemura

**Vagner Birk Jeismann, Rodrigo Blanco Dumarco, Ricardo Correa Barbuti, José Jukemura,** Department of Gastroenterology, School of Medicine, University of São Paulo, São Paulo, SP 05403-900, Brazil

**Celso di Loreto,** CICAP Pathology Laboratory, Oswaldo Cruz German Hospital, Sao Paulo, SP 01323-903, Brazil

**Celso di Loreto,** The Adolfo Lutz Institute, Sao Paulo, SP 01246-902, Brazil

**Author contributions:** Jukemura J, Dumarco RB and Jeismann VB performed the procedure; Dumarco RB and Jeismann VB reviewed the literature; Barbuti RC, di Loreto C and Jukemura J analyzed and reviewed the paper; Jeismann VB and Jukemura J wrote the paper.

**Correspondence to: Vagner Birk Jeismann, MD,** Department of Gastroenterology, School of Medicine, University of São Paulo, Avenida Doutor Enéas de Carvalho Aguiar, 255 Central Institute, Room 9074, São Paulo, SP 05403-900, Brazil. [vjeismann@gmail.com](mailto:vjeismann@gmail.com)

**Telephone:** +55-11-26617560 **Fax:** +55-11-26617560

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**Abstract**

The occurrence of a hepatoduodenal ligament teratoma is extremely rare, with only a few cases reported in the literature. This case report describes the discovery of a hepatoduodenal ligament lesion revealed during abdominal ultrasonography for cholelithiasis-related abdominal pain in a 27-year-old female. Cross-sectional imaging identified a 5 cm × 4 cm heterogeneous mass of fat tissue with irregular calcification located in the posterior-superior aspect of the head of the pancreas. An encapsulated lesion showing no invasion to the common bile duct or adjacent organs and vessels was exposed during laparotomy and resected. Intraoperative cholangiography during the cholecystectomy showed no abnormalities. The postoperative course was uneventful. Pathological analysis of the resected mass indicated hepatoduodenal ligament teratoma. This case report demonstrates that cross-sectional imaging, such as computed tomography, can reveal suspected incidences of this rare type of teratoma, which can then be confirmed after pathologic analysis of the specimen. The prognosis after complete surgical resection of lesions presenting with benign pathological features is excellent.

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**Key words:** Abdominal incidentaloma; Teratoma; Hepatoduodenal ligament; Surgery; Hepatobiliary surgery

**Core tip:** The 10th reported case of hepatoduodenal ligament teratoma is presented in a patient who underwent cross-sectional imaging for the evaluation of an abdominal mass. As incidences of hepatoduodenal ligament teratoma are extremely rare, this report may help physicians to suspect this disorder in an emergent group of patients with abdominal incidentaloma.

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**INTRODUCTION**

Teratomas are neoplasms comprised of mixed dermal elements derived from the three germ cell layers. Although the majority of teratomas are congenitally present in the gonads of men and women, they have been identified in extra-gonadal sites, such as the anterior mediastinum, retroperitoneum and sacrococcygeal regions[1]. Teratomas in the hepatoduodenal ligament are extremely rare, with only nine cases described in the literature[2-10] (Table 1). We report a case of hepatoduodenal ligament teratoma in an adult female patient examined for cholelithiasis-related abdominal pain.

**CASE REPORT**

A 27-year-old female presented with cholelithiasis-related pain. There was no history of jaundice, and the past medical history was unremarkable. Physical examination failed to detect the presence of an abdominal mass, and routine laboratory tests were normal. Abdominal ultrasonography revealed cholelithiasis and a mass adjacent to the hepatic hilum. Computed tomography (CT) and magnetic resonance imaging revealed a heterogeneous mass of 5 cm × 4 cm comprised of fat tissue and irregular calcifications located in the hepatoduodenal ligament at the posterior-superior aspect of the head of the pancreas (Figure 1).

Following patient consent, a laparotomy was performed. A Kocher maneuver with extensive mobilization of the duodenum exposed an encapsulated lesion. It was dissected and resected, and the multiple small vessels from the hepatic pedicle to the lesion were divided. There was no invasion of adjacent organs, vessels or the common bile duct (Figures 2 and 3). A cholecistectomy was performed and the intraoperative cholangiogram did not show abnormalities. The postoperative course was uneventful, and the patient was discharged after four days. The patient remains asymptomatic after six months. Histopathology confirmed that the mass was a mature teratoma. Microscopic examination revealed the presence of a cystic wall with cutaneous annexes and a mature neural area with glial fibrillary acidic protein (GFAP) immunoreactivity (Figure 4).

**DISCUSSION**

Teratomas are composed of structures derived from the three germ layers, namely the ectoderm, mesoderm and endoderm. Most mature teratomas are benign, but can undergo a malignant change in one of their elements[6]. Although plain abdominal radiographs show calcification in most (60%) extra-gonadal teratomas, either in the wall of the cyst or in structures such as teeth or bones, CT is generally the most helpful imaging modality for diagnosis[7].

An extensive review of the literature identified nine reported cases of hepatoduodenal teratoma[2-10]. Six of the described cases were in children[2-5,8,10], and the oldest patient identified was 38 years old at the time of diagnosis[7]. A small gender difference is evident, as the lesions were more often described in women[5,6,9,10]. Clinical manifestations were variable, including jaundice[2,4,10], portal hypertension[3,6] and a palpable abdominal mass[7,8,10]. Some patients demonstrated elevated levels of serum alpha-fetoprotein, carcinoembryonic antigen and carbohydrate antigen 19-9, but these do not appear to be clinically useful[7]. The majority of cases reported tumors with benign pathology features, except for one case with an endodermal sinus tumor[4], and two cases that did not provide described pathology report details[2,3]. All patients underwent surgical resection, and two patients received chemotherapy[2,4], with only one incidence of recurrence[2]. Since a definitive diagnosis is only achieved following histologic examination of the cyst, surgical resection remains the primary treatment with an excellent prognosis[7]. In conclusion, this is the first reported asymptomatic case, to our knowledge, of hepatoduodenal ligament teratoma, indicating that teratomas should not be ruled out in cases of abdominal incidentaloma.

**COMMENTS**

***Case characteristics***

The patient was asymptomatic.

***Clinical diagnosis***

The patient was diagnosed with abdominal incidentaloma uncovered during investigation of cholelithiasis-related abdominal pain.

***Differential diagnosis***

Benign and malignant abdominal tumors were alternative diagnoses.

***Laboratory diagnosis***

No abnormalities were found in the laboratory tests.

***Imaging diagnosis***

A heterogeneous mass of 5 cm × 4 cm with fat tissue and irregular calcifications was located in the posterior-superior aspect of the head of the pancreas, into the hepatoduodenal ligament.

***Pathological diagnosis***

Analysis by microscopy revealed a mature teratoma cystic wall with cutaneous annexes and glial fibrillary acidic protein staining of a mature neural area, findings that are consistent with a teratoma.

***Treatment***

The patient was treated by surgical resection of the tumor.

***Related reports***

A hepaduodenal teratoma is a rare occurrence and, to the best of our knowledge, this is the 10th reported case and the 1st asymptomatic reported case.

***Term explanation***

Hepatoduodenal ligament teratoma refers to a neoplasm that is comprised of mixed dermal elements derived from the three germ cell layers and located at the portion of the lesser omentum extending between the porta hepatis of the liver and superior part of the duodenum. Computer tomography is a technology that uses computer-processed X-rays to produce cross-sectional imaging of the human body. Abdominal incidentaloma has been defined as an intraabdominal tumor found in a patient without symptoms, usually during evaluation of unrelated diseases or screening programs. Cholelithiasis is defined by the presence/formation of stones within the biliary tract, most commonly the gallbladder.

***Experiences and lessons***

Teratomas must be included on differential diagnosis of all abdominal incidentalomas.

***Peer review***

This case report provides a description of a rare disease that may be under-diagnosed due to a low index of suspicion. The pathological, radiological and surgical findings are well documented.

**REFERENCES**

1 **Engel RM**, Elkins RC, Fletcher BD. Retroperitoneal teratoma. Review of the literature and presentation of an unusual case. *Cancer* 1968; **22**: 1068-1073 [PMID: 5686638 DOI: 10.1002/1097-0142(196811)22: 5<1068:: AID-CNCR2820220525>3.0.CO; 2-3]

2 **Frexes M**, Neblett WW, Holcomb GW. Spectrum of biliary disease in childhood. *South Med J* 1986; **79**: 1342-1349 [PMID: 3775460 DOI: 10.1097/00007611-198611000-00007]

3 **Akimov OV**. [Hepatoduodenal ligament teratoma followed by hypertensive syndrome of the portal vein]. *Arkh Patol* 1989; **51**: 60-62 [PMID: 2719564]

4 **Kim WS**, Choi BI, Lee YS, Chi JG, Park HR, Kim I, Yeon KM, Han MC. Endodermal sinus tumour associated with benign teratoma of the common bile duct. *Pediatr Radiol* 1993; **23**: 59-60 [PMID: 8469596 DOI: 10.1007/BF02020227]

5 **Demircan M**, Uguralp S, Mutus M, Kutlu R, Mizrak B. Teratoma arising from anomalous common bile ducts: a case report. *J Pediatr Surg* 2004; **39**: e1-e2 [PMID: 15065072 DOI: 10.1016/j.jpedsurg.2003.12.036]

6 **Wang H**, Dong J. Teratoma in the hepatoduodenal ligament followed by portal hypertension syndrome. *J Gastroenterol Hepatol* 2004; **19**: 477-479 [PMID: 15012796 DOI: 10.1111/j.1440-1746.2004.03366.x]

7 **Sasaki H**, Ajiki T, Takase S, Fujino Y, Suzuki Y, Tominaga M, Ku Y, Kuroda Y. Images of interest. Hepatobiliary and pancreatic: mature cystic teratoma in the hepatoduodenal ligament. *J Gastroenterol Hepatol* 2005; **20**: 317 [PMID: 15683440 DOI: 10.1111/j.1440-1746.2005.03784.x]

8 **Ukiyama E**, Endo M, Yoshida F. Hepatoduodenal ligament teratoma with hepatic artery running inside. *Pediatr Surg Int* 2008; **24**: 1239-1242 [PMID: 18807051 DOI: 10.1007/s00383-008-2205-x]

9 **Souftas V**, Polychronidis A, Giatromanolaki A, Perente S, Simopoulos C. Dermoid cyst in the hepatoduodenal ligament: report of a case. *Surg Today* 2008; **38**: 959-961 [PMID: 18820876 DOI: 10.1007/s00595-007-3744-9]

10 **Bagga D**, Jindal B, Naredi BK, Yadav DK, Acharya SK, Mahato R, Gupta K. Portal teratoma causing obstructive jaundice in children: a rarity. *J Pediatr Surg* 2012; **47**: 1449-1452 [PMID: 22813813 DOI: 10.1016/j.jpedsurg.2012.04.016]

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**Figure 1 Cross-sectional imaging.** A hepatoduodenal heterogeneous mass was revealed by A: Computed tomography; B: Magnetic resonance imaging.

**Figure 2 Operative finding.** A laparotomy revealed an encapsulated lesion without invasion to adjacent organs or vessels (1: common bile duct; 2: teratoma; 3: right lobe of the liver).

**Figure 3 Tumor appearance**. The resected heterogeneous lesion was composed of fat tissue, calcifications and hair.

**Figure 4 Histopathology of the tumor**. Microscopic examination of the specimen revealed A: A cystic wall with cutaneous annexes; B: Glial fibrillary acidic protein (GFAP) immunoreactivity.

**Table 1 Reported cases of hepatoduodenal ligament teratoma (adapted and expanded with permission from Ukiyama *et al*)**

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| Patient | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
| Year Reported | 1986 | 1989 | 1993 | 2004 | 2004 | 2005 | 2008 | 2008 | 2012 | 2013 |
| Ref. | Frexes  *et al*[2] | Akimov *et al*[3] | Kim *et al*[4] | Demircan *et al*[5] | Wang *et al*[6] | Sasaki *et al*[7] | Ukiyama *et al*[8] | Souftas *et al*[9] | Bagga *et al*[10] | Our case |
| Age | Neonate | 6 yr | 5 yr | 4 mo | 29 yr | 38 yr | 20 mo | 26 yr | 11 yr | 27 yr |
| Sex | n.a | NA | Male | Female | Female | Male | Male | Female | Female | Female |
| Origin | Extrahepatic bile duct | HL | CBD | Anomalous CBD | HL | HL | HL | HL | HL and fistulization with the CBD | HL |
| Signs and Symptoms | Jaundice | Portal hypertension | Jaundice | Jaundice, abdominal distension | Portal hypertension | Abdominal mass | Abdominal mass | Abdominal pain | Jaundice, abdominal mass | Asymptomatic |
| Size | Small mass | NA | NA | Cystic mass  15 cm | Solid mass  7 cm × 6 cm × 6 cm | Cystic mass  8 cm | Solid mass  9 cm × 6 cm × 6 cm | Cystic mass  11 cm | Cystic mass  9 cm × 9 cm | 5 cm × 4 cm |
| Pathology | Teratoma | NA | Endodermal sinus tumor associated with teratoma | Benign cystic teratoma | Benign teratoma | Benign cystic teratoma | Benign teratoma | Dermoid cyst | Benign cystic teratoma | Benign teratoma |
| Treatment | Local excision, recurrence, re-excision with chemotherapy | NA | Whipple’s operation with chemotherapy | Extirpation with CBD | Extirpation | Extirpation with CBD, Roux-en-Y, Choledocho-jejunostomy | Extirpation | Excision of the tumor | Extirpation leaving the outer cyst wall *in situ* (Lilly technique), hepatico-duodenostomy | Extirpation |
| Prognosis | Asymptomatic after 5 yr | Death | Death | Asymptomatic after 4 yr | Asymptomatic after 2 yr | NA | Asymptomatic after 5 yr | Asymptomatic after 33 mo | Asymptomatic after 2 yr | Asymptomatic after 6 mo |

CBD: Common bile duct; HL: Hepatoduodenal ligament; NA: Not available.