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CASE REPORT

Rare cause of abdominal incidentaloma: Hepatoduodenal ligament teratoma

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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.

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

Table 1 Reported cases of hepatoduodenal ligament teratoma (adapted and expanded with permission from Ukiyama *et al*[8])

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	NA	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.

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 cholecystectomy 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 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

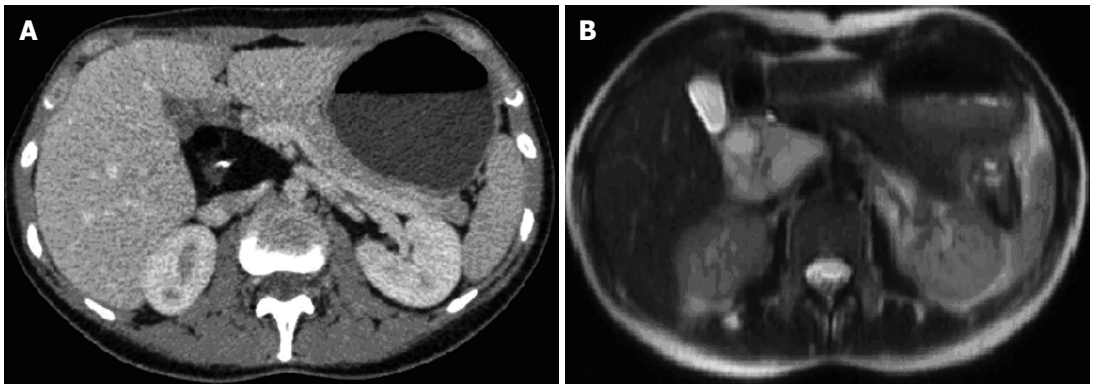

Figure 1 Cross-sectional imaging. A hepatoduodenal heterogeneous mass was revealed by A: Computed tomography; B: Magnetic resonance imaging.

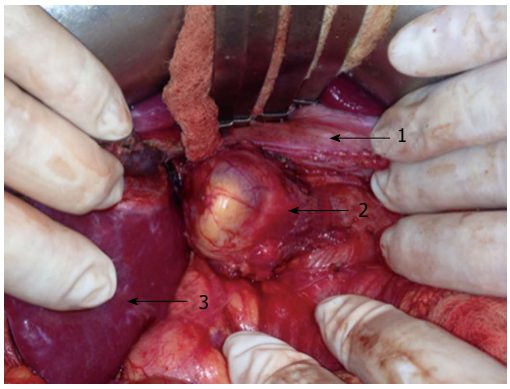

Figure 2 Operative finding. A laparotomy revealed an encapsulated lesion without invasion to adjacent organs or vessels (1: Common bile duct; 2: Teratoma; 3: Right lobe of the liver).

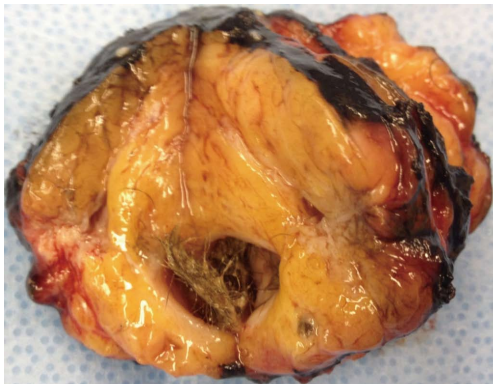

Figure 3 Tumor appearance. The resected heterogeneous lesion was composed of fat tissue, calcifications and hair.

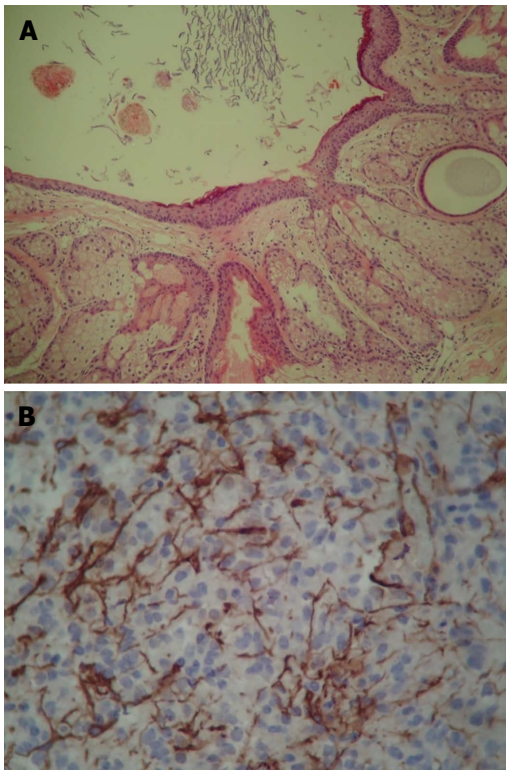

Figure 4 Histopathology of the tumor. Microscopic examination of the specimen revealed A: A cystic wall with cutaneous annexes; B: Glial fibrillary acidic protein immunoreactivity.

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.

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 hepatoduodenal teratoma is a rare occurrence and, to the best of the authors' 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 underdiagnosed due to a low index of suspicion. The pathological, radiological and surgical findings are well documented.

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