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Hepatic artery aneurysm causing gastrointestinal haemorrhage – Case report and literature review

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ABSTRACT

INTRODUCTION: True hepatic artery aneurysms (HAAs) are rare, and when complicated by gastrointestinal haemorrhage, it becomes an even rarer disease entity. The mortality is high and imaging may fail to provide the diagnosis. We present a case of a true hepatic artery aneurysm complicated by a fistula to the duodenum which was first recognised during surgery.
PRESENTATION OF CASE: A 77-year-old man presented with upper gastrointestinal haemorrhage. Upper endoscopy revealed an ulceration in the duodenal bulb, which was refractory to endoscopic treatment. Computed tomography and angiography did not reveal the source of haemorrhage and as such, the diagnosis was delayed, until laparotomy was performed. Resection of the HAA and graft placement resulted in complete haemostasis.
DISCUSSION: True hepatic aneurysms communicating with the gastrointestinal tract have only been presented in case reports and short case series. Arteriosclerosis is a relatively common risk factor, but the underlying pathology is unknown. Meanwhile, gastrointestinal haemorrhage is a symptom of other, more common diseases in the gastrointestinal tract, and these factors, complicate the diagnostic workup.
CONCLUSION: In the case of treatment refractory duodenal haemorrhage, a visceral aneurysm should be considered. Even though angiography is performed, a HAA may remain undetected due to bleeding cessation. Improved computed tomography modalities could aid in the detection of gastrointestinal haemorrhage from HAAs, and ensure timely treatment by endovascular methods or surgery if the diagnosis is kept in mind in the initial evaluation.

1. Introduction

Hepatic artery aneurysms (HAAs) are rare, with an estimated incidence of 0.002-0.4% [1,2]. HAAs are thought to account for 20% of visceral aneurysms, making it the second most common after splenic artery aneurysms [2]. However, increased use of abdominal CT scans and therapeutic biliary procedures have increased the detection of at least pseudo HAAs [3]. Symptoms of a HAA range from abdominal pain (55%) to total lack of symptoms (2%). The number of asymptomatic cases may however be underestimated [3].

True HAA's communicating with the GI tract are only described in few case-reports. The Medline database was searched for potential articles using the MESH terms "hepatic artery", "aneurysm", and "gastrointestinal haemorrhage". A total of 187 articles were published from 1964 until 2014. Eighty-eight articles described pseudo-aneurysms or aneurysms arising from other arteries in the upper abdomen. The articles [4–34] on HAAs communicating with the GI-tract or the biliary system are presented in Table 1.

Fistulas from HAAs usually communicate with the biliary system. According to our literature search, a HAA with a fistula to the duodenum, has only been presented in seven previous studies/case reports. HAA present a diagnostic challenge due to the rarity of the disease with a variety of treatment options.

We present a case of a HAA complicated by a duodenal fistula. The case-presentation is made in full accordance with the current guidelines for surgical case reports (SCARE) [35].

2. Case presentation

A 77-year-old male with known hypertension (treated with bendroflumethiazid, amlodipine, and losartan), moderate daily alcohol consumption, and massive tobacco consumption, was admitted with hematemesis. Initial upper endoscopy did not reveal the bleeding source, and he was therefore managed conservatively. On the third day of admission, he developed a circulatory collapse.

Abbreviations: HAA, hepatic artery aneurysm; GI, gastrointestinal; CT, computed tomography.

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Table 1
True aneurysms of the hepatic artery and gastrointestinal haemorrhage.

Reference	Article type	No. of cases	Patient age	Location of fistula	Diagnostics	Treatment	Cause
Mortimer and Gresham [6]	Case report	1	77	NA	Autopsy	Laparotomy	NA
Graham et al. [5]	Case report	1	61	Portal vein	Angiography	Surgical resection	Heriditary telangiectasia
Macdonald et al. [7]	Case report	1	52	Gallbladder	Laparotomy	Surgical resection	Cholecystitis
Gryboski and Clemett [8]	Case report	1	18 weeks	NA	Autopsy	NA	Congenital
Sandblom [9]	Case report	1	73	Pancreatic duct	Angiography	NA	NA
Gupta and Cope [10]	Case report[α]	1	30	Common bile duct	Angiography	Surgical ligation	Endocarditis
Santiago-Delpin et al. [11]	Case report	1	48	Common bile duct	Laparotomy	Surgical resection partial	Marfan syndrome
Croom et al. [12]	Case report	1	73	Common bile duct	Agiography	Surgical resection	NA
Balthazar [13]	Case report[α]	1	74	Common bile duct	Angiography	Surgical resection	NA
Harlaftis and Akin [14]	Case report and literature review	48	62[a]	Common bile duct[a] Gallbladder Cystic duct Peritoneal cavity Hepatic ducts Duodenum Unknown	Angiography[a]	Surgical ligation[a]	NA
Cranston and Smith [15]	Case report	1	83	Intestinal tract not otherwise specified	Angiography	Surgical resection	NA
Hügel et al. [16]	Case report	1	56	Duodenal bulb	Angiography	Surgical resection	NA
Stierli et al. [17]	Case report	1	51	Pancreatic duct	NA	Surgical resection	Giant cavernous hemangioma
Psathakis et al. [18]	Case report[αβ] and literature review	2	64–70	Abdominal cavity Gallbladder and cholecystic fistula	Laparotomy	Surgical resection	NA
Werner and Bonnevie [4]	Case report	1	73	Pancreatic duct	Angiography	Surgical resection, bypass grafting	NA
Hubloue et al. [19]	Case report and literature review	1	74	Duodenal bulb	Angiography	Surgical resection	Acromegaly
Sarkar et al. [20]	Case report[α]	1	65	Common bile duct	Angiography	Embolisation	NA
Pross et al. [22]	Case report	1	56	Duodenal bulb	Angiography	Surgical ligation	Intrahepatic artery chemotherapy
O'Driscoll et al. [21]	Literature review	1	35	Common bile duct	Angiography	Embolization metal coils	NA
Cho et al. [23]	Case report	1	49	Duodenal bulb	NA	Surgical resection	NA
Maralcan et al. [24]	Case report [α,β,δ]	1	65	Bile system not otherwise specified	Angiography	Surgical ligation	NA
Shuster et al. [25]	Case report	1	21	Duodenal bulb	Angiography	Surgical resection, and venous grafting	Polyarteriitis nodosa
Narula et al. [26]	Case report	1	85	Unknown	Angiography	No treatment	NA
Traversa et al. [27]	Case report	1	49	Common bile duct	Angiography	Embolisation metal coils	NA
Morisawa et al. [28]	Case report[α]	1	83	Common bile duct	Angiography	Embolisation metal coils and gelatine sponge	NA
Soon et al. [29]	Case report	1	43	Gall bladder	CT	Embolisation metal coils	Endocarditis
Papafragkou et al. [30]	Case report	1	74	Stomach	CT	Surgical resection	NA
Wu et al. [31]	Case report	1	50	Common bileduct	CT	Embolisation metal coils and *N*-butyl cyanoacrylate	Fibromuscular dysplasia
Huisman et al. [32]	Case report	1	48	Duodenum	CT	Surgical suture laparotomy + Stent placement	NA
Kobayashi et al. [33]	Case report	1	77	Duodenum	CT	Stent placement	NA
Komatsu et al. [34]	Case report[α]	1	53	No direct fistula could be found	CT	Surgical resection	Marfan syndrome

Note: Unless otherwise specified, the aneurysm was from the common hepatic artery. NA = Not available. CT = Computed Tomography.
[a] Data on the one patient from the case report. Ages from the literature review is not specified.
[α] Right hepatic artery.
[β] Left hepatic artery.
[δ] Abberant right heptic artery.

Acute upper endoscopy, was conducted, revealing an arterial bleeding in the second part of the duodenum. Endoscopic intervention failed to induce bleeding cessation, and subsequent angiography did not reveal a bleeding source (Fig. 1).

Despite subsequent embolization of the gastro-duodenal artery, the patient had another circulatory collapse, prompting acute laparotomy. During surgery, an aneurysm of the common hepatic artery with a fistula to the duodenal lumen was discovered. The aneurysm was resected, and a vascular prosthesis from the celiac trunk to the common hepatic artery was attached (Figs. 2–3). The treatment resulted in complete haemostasis. The post-operative

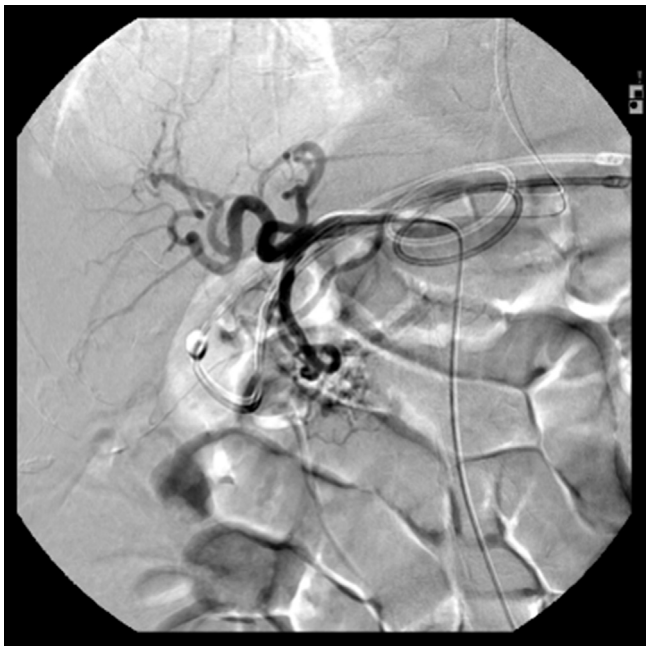

Fig. 1. Preoperative angiography. The catheter is placed in the common hepatic artery. The aneurysm of the common hepatic artery could not be visualised.

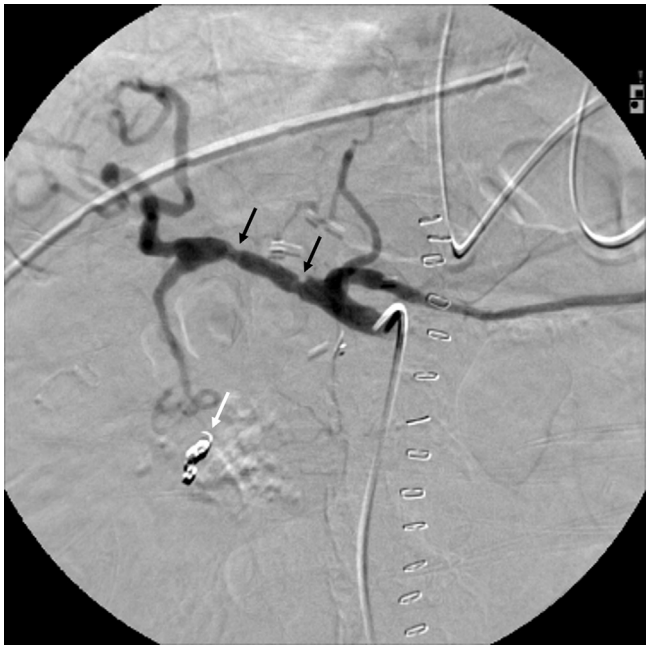

Fig. 2. Postoperative CT-angiography. Coils in the gastroduodenal artery (white arrow) and PTFE (polytetrafluoroethylene) prosthesis in the hepatic artery (black arrows).

course was complicated by colon pseudo-obstruction, for which surgical resection of the right colon was performed.

The patient was discharged five weeks after admission. He died from a cerebral infarction five years later.

3. Discussion

3.1. Diagnostics

Reported symptoms associated with HAAs are: upper or lower GI bleeding, abdominal pain, jaundice, anaemia or haemobilia [3].

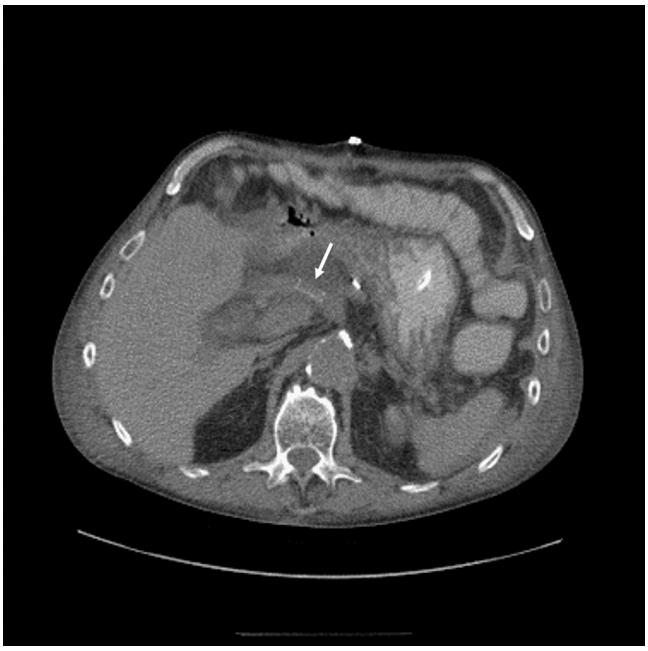

Fig. 3. Post-operative CT-scan. PTFE (polytetrafluoroethylene) prosthesis in the hepatic artery (white arrow).

These symptoms mimic those from other and more common GI diseases, providing little information as to the underlying pathology. The mortality rates are 50–100% in the case of rupture, making early diagnostics paramount [24].

Pseudo-HAAs causing gastrointestinal haemorrhage are reported multiple times, with different pathogenesis: abdominal trauma [36–38], surgery (liver transplantation, cholecystectomy etc.) [39,40], cholecystitis [41,42], cholecystolithiasis [43], or infectious liver disease [44]. However, in the case of HAAs, the underlying pathology is not so obvious. Like other aneurysms, the only predisposing factors are: atherosclerosis, connective tissue disorders (e.g. fibromuscular dysplasia, Marfan syndrome etc.), and vasculitis (e.g. polyarthritis nodosa) [45]. In HAAs presenting with GI haemorrhage, the blood loss can range from chronic haemorrhage to massive haemorrhage with the risk of circulatory collapse [3]. Patients are often asymptomatic in intervals, due to mural thrombus formation providing intermittent bleeding cessation [19]. Endoscopy may reveal a non-pulsatile mass compressing the duodenal bulb, but it is usually not sufficient to ensure the diagnosis [46].

In our case, the final diagnosis was delayed, as a preoperative CT-scan did not reveal the aneurysm. However, the device used was a 64-slice CT scanner. The implementation of the 128-slice CT modality in our department has increased the accuracy for haemorrhage detection (0.3–0.5 mL/min) [47], and it is possible that this could have ensured the diagnosis. Furthermore, the aneurysm could not be visualised with celiac trunk angiography (Fig. 1), which prompted prophylactic endovascular embolization of the gastroduodenal artery [48]. Consequently, even though imaging does not reveal an HAA, the possibility should still be considered, especially in treatment refractory cases.

3.2. Treatment

Treatment methods used in the case of true-HAA's range from reconstructive surgery with resection or ligation [24,25], to endovascular treatment strategies, using embolisation or stent placement [32,33].

Embolisation is an effective treatment modality in the case of saccular aneurysms, or fusiform aneurysms with good collateral blood supply. In case of insufficient collateral flow or concomitant liver disease, surgical procedures not impairing antegrade flow may be considered [49].

Covered metal stents have been considered contraindicated in the case of enteric fistula, due to the risk of contamination. However, the successful use of metal stents in the treatment of HAA has been described twice [32,33]. No infection occurred in any of the cases. Endovascular stent placement could therefore be a safe treatment of GI haemorrhage arising from an HAA.

Treatment using endovascular techniques requires, that the diagnosis can be made during angiography. In our case, the only possibility was surgical resection and grafting, as the aneurysm was not discovered, before open surgery was performed.

4. Conclusions

In this report, we present a rare case of an HAA presenting with GI haemorrhage. This is an uncommon cause of GI haemorrhage, which can be difficult to diagnose. The complete arterial supply from the celiac trunk should be visualised during angiography, as massive haemorrhage from a fistula to the GI tract could arise from other sites than the gastroduodenal artery. However, even though complete angiography is performed, the diagnosis may still be missed. As the survival of patients with HAA relies on early and efficient treatment, this cause should be kept in mind in the case of refractory GI haemorrhage.

Competing Interests

The authors have none to declare.

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Ethical approval

The Regional Ethics Committee where not prompted for approval under Danish Law.

Consent

"Written informed consent was not obtained from the patient for publication of this case report and any accompanying images. The next of kin could not be localized, and therefore a written consent is not available for review by the Editor of this journal.Ïhe use of this patient case was approved by the Chief of Surgery at Aalborg University Hospital. The study was registered at Research Registry on 6. June 2017 (UIN = researchregistry2621).

Author contributions

SP was the surgeon who performed the operation, made the clinical case description and drafted the manuscript.

SLR compiled the literature search and aided in drafting the manuscript.

Guarantor

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