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Research article

The 93 years Old Patient with Jaundice Caused by a Hepatic Artery Aneurysm. Case Report with Literature Review

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Abstract

The hepatic artery aneurysm (HAA) is a rare cause of cholestasis. The course of HAA, even in 75% of cases is asymptomatic. 14% of cases are complicated by rupture and hemorrhage. Before the development of imaging techniques such as computed tomography or ultrasound, the diagnosis of HAA was stated on the ground of the autopsy or during the laparotomy. Today a detection of an aneurysm is more often earlier, which enables successful treatment. 92.4% of visceral artery aneurysms are diagnosed accidentally during imaging in asymptomatic patients. The patient with an aneurysm of the hepatic artery causing mechanical jaundice, described by us is, to our knowledge, the oldest case report published. The literature review was performed using the database "PubMed" using the keywords "jaundice" and "aneurysm,". To identify additional potentially relevant data sources, we hand-searched the reference lists of the retrieved studies. Reports from 1834 to 2021 concerning mechanical jaundice caused by an aneurysm of visceral arteries were selected. The analysis included studies in which it was possible to determine the procedure, treatment results, and the artery with which the aneurysm was involved. In the reviewed works from 1834 to 2021, 77 cases were described. The oldest case so far was an 89-year-old woman, while the oldest men with this condition were 85 years old. Mechanical jaundice caused by the compression of HAA may occur in

patients in their 90-ties and needs endovascular or surgical intervention.

Keywords: Jaundice; Aneurysm; Hepatic artery aneurysm; Visceral artery aneurysm

Abbreviations: VAA Visceral artery aneurysm; HAA Hepatic artery aneurysm; CT Computed Tomography; ERCP Endoscopic Retrograde Cholangiopancreatography; EUS Endoscopic Ultrasound

1. Introduction

The hepatic artery aneurysm (HAA) is a rare cause of cholestasis. There are also publications describing mechanical jaundice caused by the compression of aneurysms originating from other arteries. It is the right and left branch of the hepatic artery, gastroduodenal artery, pancreatoduodenal arteries, celiac trunk, and arc of Buhler as in Table 1. The course of HAA, even in 75% of cases is asymptomatic. 14% of cases are complicated by rupture and hemorrhage [1]. Before the development of imaging techniques such as computed tomography or ultrasound, the diagnosis of VAA was stated on the ground of the autopsy or during the laparotomy [2-4]. Today a detection of an aneurysm is more often earlier, which enables successful treatment [5,6]. 92.4% of VAA are diagnosed accidentally during imaging in asymptomatic patients [7]. The first case of HAA that caused mechanical jaundice was described by Stokes in 1834 [2]. The diagnosis was made based on an autopsy. The first successful treatment was carried out by Kher in 1903 by ligating the HAA [4]. It is assumed that the first preoperative diagnosis through angiography was described by Doppman in 1963 [8]. In contrast, the first description of the embolization of a visceral aneurysm causing mechanical jaundice was published in 1994 [9], although an embolization of aneurysms has been performed since the 1970s. Endovascular, surgical, and alternative methods are used to treat VAA. In endovascular methods, thrombin, vascular adhesives, onyx, or spirals are administered to the aneurysm causing the aneurysm to clot or the stents can be inserted into the artery to cut off the blood supply to the aneurysm and preserve blood flow. Surgical methods are based on: ligation of the vessel with an aneurysm, aneurysmorrhaphy, excision of an aneurysm with anastomosis of the ends of the artery directly or with the use of a prosthesis as well as vein graft. Sometimes it is necessary to ligate vessels with an aneurysm with the formation of a bypass or to remove an aneurysm with a fragment of an organ. [6,10-13] Alternative methods to the above-mentioned are: percutaneous embolization of the aneurysm under ultrasound or CT control and embolization with the usage of EUS [13,14]. Due to the rarity of VAAs, reports on them are limited to case studies and case series [11]. This makes it difficult to determine the best therapeutic path for the choice of technique and timing of intervention [10]. There is no consensus on the optimal treatment of VAA [12]. Endovascular management is the treatment of choice, however, often due to unfavorable morphological conditions of the aneurysm, surgery remains the best or only solution [14]. According to the European Society of Vascular Surgery guidelines from 2017, all symptomatic VAA require repair regardless of their size and location. Asymptomatic patients with VAA <25 mm may be followed up with imaging control every 2-3 years. Invasive treatment should be considered for asymptomatic aneurysms> = 25 mm [15]. The endovascular procedure should be planned first. If it is technically possible and the patient does

not present a high perioperative risk, the procedure should be aimed at the reconstruction of the vessel and restoration of blood flow through the vessel. Patients in poor general condition and at high perioperative risk with aneurysms between 2 cm and 5 cm may be observed, but each case should be assessed on an individual basis. Aneurysms larger than 5 cm should be surgically treated [1]. Regardless of the size of the aneurysm, the intervention should be offered to women of childbearing age, planned liver transplant recipients, patients with pancreaticoduodenal, gastroduodenal, or intrahepatic aneurysms. In the case of unstable or uncontrolled hypertension, an aneurysm enlargement, or an arteriovenous fistula, the patient should be qualified for the intervention [1,15]. Visceral Artery Pseudoaneurysms, due to the high risk of perforation, should be provided urgently [14,15]. As reported by Pitton and ca. in a 10-year retrospective single-center analysis on 233 patients - no significant difference in diameter was observed between ruptured and unruptured visceral aneurysms, therefore the author suggests that the qualification should not be based solely on the size [7].

2. Case Presentation

A 93-year-old woman with painless jaundice was admitted to the General Surgery Department. Except for the compensated arterial hypertension, the patient suffered no other significant comorbidities.

The physical examination of the bedridden elderly patient stated no deviation except jaundice.

The laboratory blood tests revealed: elevated total bilirubin level 258,4 umol/L (n. 5.0-21.0 umol/L), higher activity of aminotransferases AST - 318 U/L (n. 0-35 U/L), ALT - 555 U/L (n. 0-35 U/L) and GGTP - 370 U/L (n. 0-38 U/L) with INR 1,20 (n. 0.80 - 1.20). On the ground of ultrasonographic doppler of the abdomen, cholestasis caused by hepatic artery aneurysm was stated.

Figure 1: Ultrasonographic finding in the hilum of the liver.

The diagnosis was confirmed by CT angiography. The fusiform aneurysm 85 mm x 40 mm was located under the liver and in the hilum. The aneurysm compressing gallbladder and common bile duct consisted of proper hepatic artery aneurysm (24.2 mm diameter and 38.6 mm length) and right hepatic artery aneurysm (36.3 mm diameter and 51.3 mm length). The celiac trunk was absent, splenic artery and left gastric artery arose directly from the abdominal aorta. The common hepatic artery branched from the superior mesenteric

artery. Enlargement of the abdominal aorta to 34 mm with an intraluminal thrombus 7 mm thick on a length of 50 mm was also described.

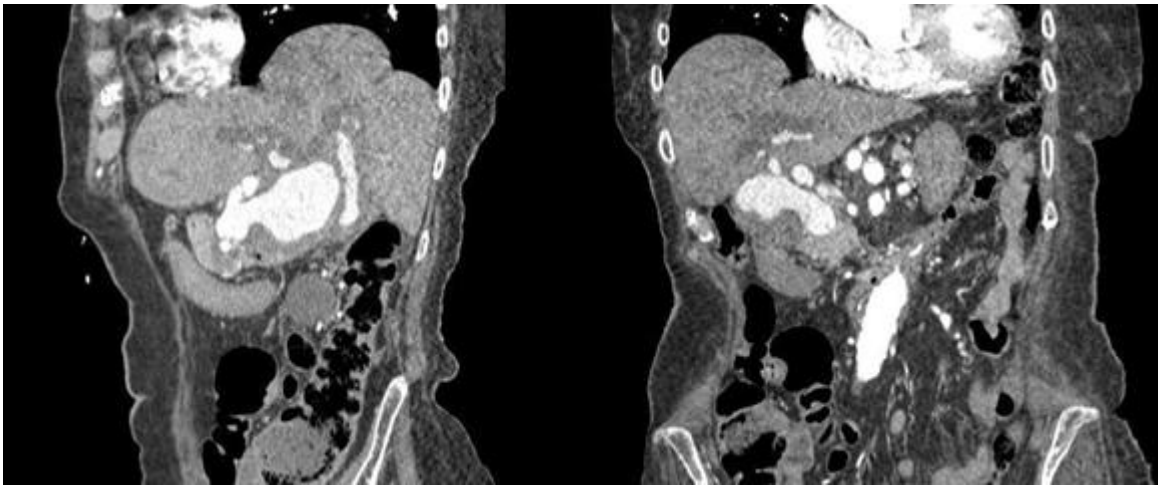

Figure 2: Proper hepatic artery aneurysm and right hepatic artery aneurysm in CT angiography of the splanchnic vessels.

To restore bile flow ERCP with papillotomy and stent (8.5 F 10 cm) placement inside the common bile duct was performed. The procedure was successful, and reduction of the total bilirubin level to 60.3 umol/l [n. 5.0 - 21.0] was noticed.

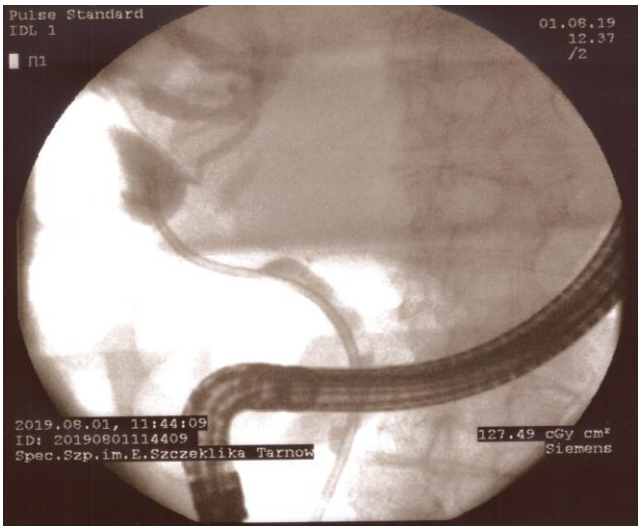

Figure 3: Stent placement during ERCP.

The postoperative course was complicated with upper gastrointestinal bleeding. An Esophagogastroduodenoscopy was performed and a source of the bleeding was found on the posterior wall of the stomach. The hemobilia was excluded and the hemorrhage was stopped with hemostatic clips. There were no other symptoms of gastrointestinal bleeding during hospitalization. Because of the aneurysm's size

and anatomy, the patient was disqualified from the endovascular procedure by two vascular surgeons. Open surgery was proposed but the patient didn't agree to the suggested treatment. She was discharged home with recommended ambulant control and USG of the abdomen in 2 months. The patient died at home a couple of weeks after discharge

3. Discussion

Along with the aging of the population and associated increasing numbers of patients aged over 90, surgeons face the problem of acute intervention in geriatric patients. The data show high perioperative mortality in this age group of 11% in the case of emergency surgery [16]. In line with the above, the treatment of the patient we described was planned in two stages. In the first stage, endoscopic biliary drainage was done. In the second stage, treatment of the aneurysm was planned. After biliary drainage was successful, the vascular procedure could be performed electively. Morphology of aneurysms - size, and vascular anomaly - arising of the hepatic artery from the superior mesenteric artery, high risk of aneurysm dissection, and gastrointestinal bleeding during the procedure were the reasons for the patient's disqualification from endovascular treatment. The patient did not consent to the open surgery, which could cure the underlying disease, however, taking into account the cardiovascular capacity and sickness burden, it could probably accelerate the death. The patient survived hospitalization but died a couple of weeks after discharge. The autopsy was abandoned, therefore the cause of death cannot be determined. Most authors do not follow up, therefore the long-term effects of therapy or the fate of patients after hospitalization are often impossible to determine. If the time of discharge is taken as the cut-off point, as in most publications, the patient survived. Three similar cases have been reported in the literature. Due to the lack of consent to the vascular procedure, the intervention in the patient described by Julianow was also limited to ERCP with a biliary stent. After 12 months, a follow-up was performed, the patient did not present any complaints. The originally described HAA was 64 mm in diameter [17]. The patient presented by Rathiel was disqualified from the vascular procedure due to cardiovascular insufficiency, while endovascular embolization was abandoned due to concerns about extensive ischemia of the treated area [18]. There is no information regarding the patient's fate after the discharge. The patient presented by Peter did not make it to the procedure scheduled for the next day. Death was due to massive hemorrhage [19]. The literature review was performed using the database "Pub Med" using the keywords "jaundice" and "aneurysm,". To identify additional potentially relevant data sources, we hand-searched the reference lists of the retrieved studies. Reports from 1834 to 2021 concerning mechanical jaundice caused by an aneurysm of visceral arteries were selected. The analysis included studies in which it was possible to determine the procedure, treatment results, and the artery with which the aneurysm was involved. In the reviewed works from 1834 to 2021, 77 cases were described. The oldest case so far was an 89-year-old woman, while the oldest men with this condition were 85 years old. The literature review is summarized in Table 1 and Table 2. The conservative treatment or surgery without vascular procedure in the case of VAA complicated by jaundice almost always resulted in the patient's death. In 28 cases only 1 survival was recorded. The only case with a patient's survival was described by McEwan-Alvarado. There is no information about follow-up [8]. ERCP without other

procedures was performed in 4 patients, 3 of them were discharged, and one died during hospitalization. The endovascular treatment was used in 28 cases, in 10 cases it was preceded by ERCP. There was no mortality after endovascular treatment. Ten patients required vascular surgery after an intravascular treatment. In the case of the surgical vascular procedure 5 patients out of 28 died.

	Author	Year	Age	Gender	Artery Involved	Aneurism Size (cm)	Managment	Outcome
1	Stokes [2]	1834	35	M	HA	"large orange"	conservative	Death
2	Jackson [20]	1834	22	M	HA	"Pullets egg"	conservative	Death
3	Lebert [21]	1855	30	F	HA	"pigeons egg"	conservative	Death
4	Wallman [3]	1858	36	F	HA	"childs head"	conservative	Death
5	Quincke [22]	1871	25	M	RHA	"chestnut"	conservative	Death
6	Standhartner [21]	1875	23	M	RHA LHA	Walnut and small nut	conservative	Death
7	Borchers [21]	1878	17	M	Intrahepatic	2,2 cm and 2,5 cm	conservative	Death
8	Caton [21]	1886	40	M	HA	inch in diameter	conservative	Death
9	White [23]	1892	18	M	RHA LHA	Tangerine orange	conservative	Death
10	Sanerteig [21]	1893	31	M	RHA LHA	Apple and cherry	conservative	Death
11	Schmidt [21]	1894	40	F	RHA	-	conservative	Death
12	Mester [21]	1895	42	M	RHA	5cm x 3,5 cm	conservative	Death
13	Sainton [21]	1896	46	M	HA	"Large orange"	conservative	Death
14	Sommer [21]	1902	28	M	HA	-	conservative	Death
15	Kehr [4]	1903	29	M	RHA	Hens egg	ligation	survival
16	Alessandri [21]	1906	22	M	RHA	Hens egg	conservative	Death
17	Campbell [24]	1911	53	M	HA	"pigeon's egg"	open surgery	Death
18	Dean [25]	1912	22	M	HA	1,5 ins. X 1,25 ins.	conservative	Death
19	Taylor [26]	1933	22	M	HA	3 cm x 2 cm	open surgery	Death
20	Malloy [27]	1942	52	F	HA	2cm	conservative	Death
21	Gordon-Taylor [28]	1943	61	F	HA	-	aneurysmoraphy	Death
22	McNamara [29]	1946	44	M	HA	-	laparotomy	Death
23	Sampsel [30]	1952	68	M	PCDA	-	open surgery	Death
24	Quattlebaumn [31]	1954	54	F	HA	2,5 cm	ligation	survival
25	Quattlebaumn [31]	1954	78	M	HA	-	4x laparotomy	Death
26	Tetreault [32]	1965	76	F	HA	2.5 cm	2x laparotomy	Death
27	Collier [33]	1966	63	M	HA	12 x 9 x 7 cm	open surgery	Death
28	McEwan-Alvarado [8]	1967	57	M	HA	-	laparotomy, conservative	survival
29	Mehnert [34]	1971	75	M	HA	-	ligation	survival
30	Hasselgren [35]	1976	62	F	PCDA	-	Aneurysmectomy	survival
31	Scheflan [36]	1977	56	M	PCDA	-	Aneurysmectomy	survival
32	Harlaflis [37]	1977	62	F	HA	1.6 by 1.2 cm	ligation	survival
33	Mojab [38]	1977	17	M	RHA LHA	-	ligation	survival
34	Kadir [39]	1978	72	M	PCDA	-	conservative	Death
35	Lewis [40]	1982	35	M	RHA	14x 8,5 cm	ERCP, PTC, surgery	survival
36	Lewis [40]	1982	62	M	GDA	-	Discovered post mortem	Death

37	Green [41]	1984	45	F	GDA	7,5 cm	Surgery	? survival
38	Bret [42]	1987	61	M	CA/HA	-	ERCP without stent, PTC, surgery (open repair)	Death
39	Blomely [9]	1994	53	M	GDA (PSA)	8,5 x 6,0 x 5,5 cm	Coli embolization	Recovery
40	Gryffith [43]	1997	33	M	LHA (PSA)		ERCP, embolization	? survival
41	Widjaja [44]	1999	51	M	PCDA	-	Transcatheter embolization	survival
42	D Odriscoll [45]	1999	55	M	HA	-	ERCP, Coli embolization	? survival
43	Chandramohan [46]	2001	62	F	HA	3 cm	ERCP, Coli embolization	survival
44	Ryan [47]	2001	88	F	RHA	-	coli embolization	survival
45	Bramis [48]	2002	49	M	HA	12 cm x 10 cm	Coli embolization, operation.	? survival
46	Mazziotti [5]	2003	74	F	RHA	3 cm	aneurysmoraphy	survival
47	Akatsu [49]	2004	64	F	HA (PSA)	3 cm	selective TAE, biliary reconstruction.	survival
48	Donmez [50]	2005	20	M	GDA (PSA)	9 · 8 · 7cm	coli embolization	survival
49	Tsitouridis [51]	2006	48	M	HA	6,5 x 13	PTC and surgery	survival
50	Aoufi [52]	2007	47	M	GDA	7 cm	ligation, relaparotomy hemihepatektomy	Death
51	Parmar [53]	2009	59	M	HA/RHA	7 cm x 15 cm	resection and reconstruction	survival
52	Colak [54]	2009	57	M	PCDA	-	Untreated	Death
53	Witczak [55]	2009	78	F	RHA (PSA)	2 x 3 cm	ERCP, Coli embolization	survival
54	Bronstein [56]	2010	37	M	CA/HA/SMA	3.6x 3.4x 7.4 cm	Surgery: resection and reconstruction	survival
55	Bennett [57]	2010	85	M	RHA (PSA)	19×8×8mm	ERCP and stenting, embolisation	survival
56	Jayia [58]	2011	65	F	Arc of Buhler	5.8 x 6.6 cm	coli embolization	survival
57	Ramirez-Maldonad [59]	2011	70	F	RHA (PSA)	7 cm in diameter	coli embolization, right hepatectomy	survival
58	Bach [60]	2012	48	M	HA	4,5 cm	Coli embolization	survival
59	Noor [61]	2012	20	M	RHA	-	Coli embolization	survival
60	Shirashi [62]	2012	75	M	HA (PSA)	8 x 7,2 x 7 cm	Surgery	survival
61	Wattez [63]	2013	64	F	PCDA	-	Laparotomy, injection of polymeric synthetic	survival
62	Morris [64]	2013	14	M	HA	5,02 cm x 4,6 cm and 2 cm	Laparotomy, microcoli embolization	survival
63	Peter [19]	2014	73	F	HA 2x	5,7x5,4x5,0; 1,3x5,3 x1,3 cm	ERCP stent	Death
64	Julianov [17]	2014	76	M	HA	6.4 cm	ERCP stent	survival
65	Raithel [18]	2014	85	M	HA	4.3 cm	ERCP stent	survival

66	Hsiao [65]	2015	40	M	RHA (PSA)	3,3 cm	Coli embolization	survival
67	Abdalla [66]	2015	40	M	RHA (PSA)	-	Embolization	survival
68	Yin [35]	2015	84	M	PCDA	2 cm	Laparotomy, drenage, embolizacion	Death
69	Yen [67]	2015	46	M	GDA	7 cm x 12 cm	ERCP, Coli embolization	survival
70	Luckhurst [68]	2016	65	F	RHA (PSA)	Multiple	Faliure of embolization and subsequent surgery	survival
71	Wyżgowski [69]	2016	73	F	RHA (PSA)	2x 4.4 cm	Coli embolization, open ligation	survival
72	Gallego-Otaegui [70]	2016	48	M	PDA (PSA)	35 mm	microcoil embolization	survival
73	Vultaggio [71]	2016	89	F	RHA branch	10 mm x 6 mm	microcoil embolization	survival
74	Linzay [72]	2017	56	M	HA	-	ERCP, Coli embolization	survival
75	Roberto [73]	2017	66	F	RHA LHA	Multiple	laparotomy LHA ligation	Death
76	DAS [74]	2019	72	F	RHA (PSA)	-	ERCP, coli embolization, partial cholecystektomy	survival
77	Chapman [75]	2021	74	M	GDA(PSA)	3.8 x 4.9 x 5.4 cm	ERCP aneurysmorraphy	survival
78	Our case	2021	93	F	RHA, HA	85 x 40 mm	ERCP with stent	survival

Table 1: Literature review

Abbreviations: HA Hepatic artery; RHA right hepatic artery; LHA left hepatic artery; GDA gastroduodenal artery; PCDA pancreatoduodenal artery; CA celiac axis; SMA superior mesenteric artery; PSA pseudoaneurysm; ERCP Endoscopic Retrograde Cholangiopancreatography; PTC Percutaneous transhepatic cholangiography; TAE transcatheter arterial embolization.

Treatment	Cases	Deaths
Conservative treatment, surgery without vascular procedure	28	27
Surgery with vascular procedure	17	3
ERCP	4	1
Endovascular procedure	11	0
ERCP + Endovascular procedure	7	0
ERCP + Surgery with vascular procedure	1	0
ERCP + Endovascular procedure + Surgery with vascular procedure	3	1
Endovascular procedure + Surgery with vascular procedure	7	1

Table 2: Treatment procedure

4. Conclusion

Jaundice due to HAA can be observed in patients over 90 years of age, so this possibility should be taken into account in the differential diagnosis. VAA with jaundice requires an endovascular or surgical intervention. In hemodynamically stable cases ERCP with biliary stenting can be performed before causative endovascular or surgical treatment, to prepare the patient for further treatment. ERCP with biliary stenting can also be performed in the absence of the patient's consent to the vascular procedure to protect the liver function.

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