

ORIGINAL ARTICLE

Management of Coeliac and Hepatic Artery Aneurysms: An Experience of 84 Cases

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WHAT THIS PAPER ADDS

Due to the rarity of hepatic and coeliac artery aneurysms, current guidelines are based on a low level of evidence from small retrospective observational studies. To date, this study includes the largest single centre experience of this condition. The results show that the complication rate was higher with open surgical repair, however, long term patency and survival was similar between open surgery and endovascular treatment. Although endovascular treatment is the preferred approach, open repair is a viable option when endovascular treatment is not feasible.

Objective: To report outcomes following open or endovascular treatment of true hepatic and coeliac artery aneurysms at a single referral centre.

Methods: This was a retrospective cohort study of consecutive patients treated for true hepatic and coeliac artery aneurysms between May 2002 and December 2021. Outcome measures included complications, graft patency, and survival rate.

Results: Overall, 84 patients were included with a median age of 63 years (interquartile range 55, 79). The majority (76%) of the patients were men. Frequent comorbidities included a history of tobacco (69%), hypertension (65%), hyperlipidaemia (32%), and diabetes (15%). Multiple synchronous aneurysms were detected in 22 patients (26%). There were 33 (39%) symptomatic aneurysms (abdominal pain without rupture [$n = 18$], rupture [$n = 10$], and sepsis [$n = 5$]). Seventeen patients (20%) had mycotic aetiology. Fifty patients (60%) underwent endovascular treatment with either covered stent placement ($n = 29$) or coil embolisation ($n = 21$), and 34 patients (40%) were treated with open surgery using allogenic iliac artery ($n = 15$), autologous saphenous vein ($n = 15$), GoreTex graft ($n = 2$), or ligation ($n = 2$). The complication rate was 32% in the open group and 18% in the endovascular group ($p = .048$). The overall 90 day post-operative mortality rate was 1.2%, five year primary patency was 90.0%, five year survival rate was 81.2%, and mean follow up was 6.9 ± 4.2 years.

Conclusion: Endovascular treatment is the preferred approach whenever technically possible. Despite higher post-operative morbidity, an open approach with vascular reconstruction using autologous or allogenic vascular grafts yields acceptable long term results.

Keywords: Coeliac artery aneurysm, Hepatic artery aneurysm, Mesenteric artery aneurysm, Visceral artery aneurysm

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INTRODUCTION

Hepatic artery aneurysms (HAA) and coeliac artery aneurysms (CAA) represent 20% and 4% of all visceral aneurysms, respectively.^{1,2} Death following spontaneous rupture has been reported to be as high as 40%.^{1–5} Treatment is

challenging due to the importance of maintaining arterial circulation to the liver and avoiding ischaemic injury to the biliary tree. True HAA and CAA are associated with atherosclerosis, connective tissue diseases, vasculitis, and infectious conditions.^{6–9} Based on the current literature the relationship between size and risk of rupture is uncertain.^{10,11} HAA and CAA repair is advocated for all symptomatic aneurysms. Due to the high morbidity and mortality rates associated with rupture, American guidelines recommend repair of asymptomatic HAA and CAA at a diameter > 20 mm,¹¹ while European guidelines recommend

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treatment at a diameter > 25 mm.¹² The traditional HAA and CAA treatment option has been open surgery with resection of the aneurysm and vascular reconstruction with bypass using autologous vein, synthetic graft, or allogenic graft. After the introduction of endovascular techniques, this has gained wide acceptance as a safe and minimally invasive alternative with encouraging results.^{1,6,11–14} The endovascular approach includes either coil embolisation or placement of a stent graft to preserve arterial flow when this is considered mandatory.

Due to their rarity there are only few cases reported, and current guidelines are based on a low level of evidence from case reports and small retrospective observational studies with limited follow up.^{5,11,12,15} This study presents a series of 84 cases of HAA and CAA and evaluates the different treatment strategies within a high volume tertiary referral centre over a 19 year period.

MATERIAL AND METHODS

Patient selection

This was a retrospective study of all patients undergoing treatment for true HAA and CAA at Rikshospitalet, Oslo University Hospital from May 2002 to December 2021. Oslo University Hospital is the only national centre for abdominal transplantation surgery in Norway (5.4 million inhabitants). The indication for treatment was either symptomatic aneurysm regardless of size or asymptomatic aneurysm with a diameter > 20 mm. All medical records were reviewed to ensure inclusion of true aneurysms only. Pseudoaneurysms were excluded from this study because of their completely different pathophysiology. In the present authors' institution, pseudoaneurysms were most often seen as a complication of pancreatic cancer surgery,^{6–8} with associated short life expectancy. Data collection included patient demographics, medical history, clinical presentation, aneurysm characteristics evaluated by radiologists, treatment, and complications. The severity of complications was classified according to the Comprehensive Complication Index (CCI) grading system.¹⁶ Complications that required surgical or radiological interventions were categorised as major, hence a cutoff for CCI was set at 25 to define a major complication. Subgroup analyses regarding treatment method, anatomic location, rupture status, and type of bypass used for vascular reconstruction were performed. The primary outcome was procedure related morbidity and mortality. Secondary outcomes included patency, need for re-intervention, and long term survival assessed by the Norwegian Population Register. The study was approved by the institutional review board according to the general guidelines provided by the regional ethics committee (20/02489). The manuscript was completed in accordance with the STROBE statement¹⁷ and SQUIRE guidelines.¹⁸

Medical examination and treatment

Triple phase computed tomography (CT) of the abdomen was performed in all patients. Aneurysm size, anatomical

features, and localisation were evaluated. Mycotic aneurysm was diagnosed if the patient fulfilled at least two of the three criteria:¹⁹ (1) clinical presentation with fever, concomitant infection, or relevant pain; (2) blood test confirming systemic infectious response such as leucocytosis, elevated C-reactive protein level, or positive blood culture; (3) radiological findings showing uptake on PET-CT, rapid expansion of aneurysm sac, saccular or multilobular aneurysm, peri-arterial gas, or soft tissue mass.

Routinely, all cases were discussed and evaluated by an experienced abdominal transplant surgeon and vascular interventional radiologist. In accordance with current guidelines,^{11,12,15} an endovascular approach was chosen for all patients when technically possible. Criteria for judgement of technical feasibility for endovascular treatment were mainly accessibility to the diseased vessel and ability to maintain end organ perfusion by either covered stents or pre-existing collaterals. In cases of intrahepatic aneurysms, occlusion of end arteries was accepted as a treatment alternative if small vessel size excluded use of covered stents. The use of covered stents was planned in the presence of adequate proximal and distal landing zones in the affected artery without the need for sacrifice of notable hepatic artery branches. Endovascular treatment was performed through a transfemoral approach in all cases. Due to the long inclusion period of this study, a large variety of devices, including covered balloon expandable and covered self expanding stents as well as micro- and macrocoils and vascular plugs were used. Technical success was defined as complete exclusion of the aneurysm from the arterial circulation as assessed by angiography immediately after the procedure.

Open surgery was performed only in cases where an endovascular approach was considered technically unsuitable. Surgical access was established through a midline laparotomy with right subcostal extension. Samples for microbiological culture as well as histology of the aneurysm wall were taken whenever possible. To mitigate the potential risk of post-operative graft infection, biological bypass grafts (autologous saphenous vein or allogenic iliac artery) were preferred whenever possible. Vascular allografts were routinely stored at temperature of 4°C in preservation solution (IGL-1 or UW), for a maximum of 14 days. Blood type compatible grafts were used in all cases. No immunosuppressive treatment was given to any of the patients receiving an allograft in this study. Post-operative anticoagulation to patients who underwent open surgery was prophylactic low molecular weight heparin (LMWH) until full mobilisation and then lifelong aspirin. The patients who underwent endovascular stent placement received dual antiplatelet therapy for three months and then lifelong aspirin. After one year of routine follow up by CT angiography, subsequent radiology was obtained based on various clinical indications. All relevant radiological examinations of patients included in this study were collected from local hospitals for assessment of long term patency. Patency was defined as a patent stent or bypass on CT angiography. Follow up of coil embolisation was assessed by the absence of circulation in the aneurysm on CT angiography.

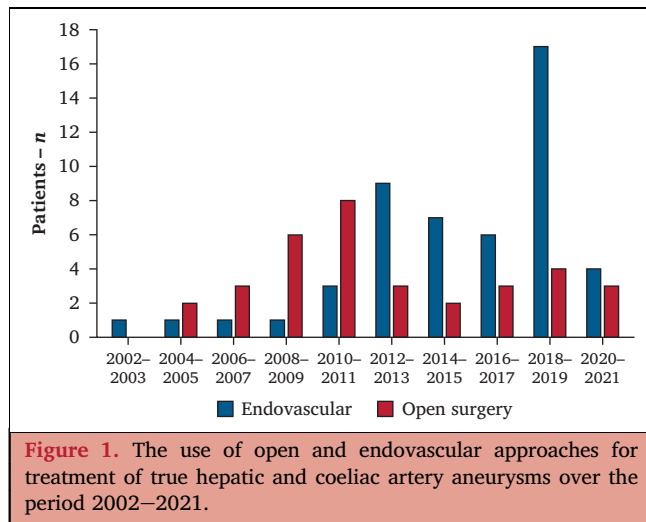


Figure 1. The use of open and endovascular approaches for treatment of true hepatic and coeliac artery aneurysms over the period 2002–2021.

Statistical analysis

Data are presented as mean \pm standard deviation (SD), or median with interquartile range (IQR) for continuous data and counts with percentage for categorical data. Groups were compared using Student *t* test for parametric variables, Mann–Whitney *U* test for non-parametric variables, and chi square test for categorical variables. Survival was estimated from date of treatment to date of death or 25 June 2023. Patency was estimated from date of treatment to date of last follow up CT angiogram. Kaplan–Meier curves were made and patients were censored at the time of occlusion, death, or at the end of follow up. Logistic regression was used to calculate the significance of risk factors for developing post-operative complications. The unadjusted bivariable association between risk factors and post-operative complications was examined, and then clinically relevant variables were included in a multivariable model adjusted for confounding factors such as age and smoking. A *p* value $< .050$ was considered statistically significant. All calculations were made using Stata Statistical Software 17 (College Station, TX, USA).

RESULTS

Demographic data and clinical presentation

A total of 84 consecutive patients were included (Fig. 1). The median age was 63 years (IQR 55, 79), and 64 (76%) of the patients were men. Frequent comorbidities included a history of tobacco use (69%), hypertension (65%), hyperlipidaemia (32%), and diabetes (15%) (Table 1). The most common aetiology was atherosclerosis in 62 patients (74%). Seventeen patients (20%) had mycotic aneurysms, nine of them intrahepatic. The original focus of infection in the intrahepatic mycotic aneurysms was endocarditis ($n = 3$), cholangitis ($n = 3$), liver abscess ($n = 2$), and infected pancreatic cyst ($n = 1$). The extrahepatic mycotic aneurysms resulted from endocarditis ($n = 2$), sepsis ($n = 2$), and

infection of unknown origin ($n = 4$). Positive bacterial cultures from the aneurysm wall were identified in 30% in the open surgery group with no statistically significant correlation with mycotic aetiology. Five patients (6%) suffered from a genetic or autoimmune disease (fibromuscular dysplasia $n = 1$, polyarthritis nodosa $n = 2$, ANCA positive vasculitis, FOXE3 gene mutation). In 51 patients (61%) the aneurysm was asymptomatic, and the median aneurysm diameter was 27.5 mm (range 20 – 79 mm). The symptomatic aneurysms (39%) presented with abdominal pain without rupture (21%), rupture (12%), and sepsis (6%) (Table 1). Three out of 10 patients (30%) with ruptured HAA had an aneurysm diameter < 20 mm (12, 14, and 15 mm) on presentation. The remaining seven ruptured aneurysms had a mean diameter of 34 mm \pm 14 mm (range 21 – 64 mm) with one mycotic, three genetic or autoimmune disease, and six atherosclerotic aetiologies.

Anatomic location

Synchronous aneurysms were detected in 22 patients (26%), most commonly in the aorto-iliac region ($n = 10$), followed by the superior mesenteric artery ($n = 4$), renal artery ($n = 3$), and splenic artery ($n = 2$). The synchronous aneurysms were either observed or treated after coeliac and/or hepatic artery repair. Seventy five (89%) aneurysms were extrahepatic. The most common locations were the common hepatic artery (35%), followed by the coeliac trunk (25%), intrahepatic arteries (11%), and proper hepatic artery (9%) (Table 2).

Treatment

Fifty patients (60%) underwent endovascular treatment with either implantation of covered stents ($n = 29$) or coil embolisation ($n = 21$) (Table 2). All nine intrahepatic aneurysms were treated by coil embolisation. Thirty four patients (40%) were treated by open surgery (Table 2). No statistically significant differences regarding gender and comorbidities were demonstrated between the endovascular and open surgery groups (Table 1). Patients who underwent open surgery had a statistically significantly larger median aneurysm diameter compared with the endovascular group (32.5 mm vs. 24.0 mm, $p = .008$). Vascular reconstruction and bypass were performed using autologous saphenous vein ($n = 15$), allogenic iliac artery ($n = 15$), or synthetic graft ($n = 2$). Two patients underwent ligation of the aneurysm as the only treatment.

Technical failure, complications, and follow up

Major post-operative complications (CCI > 25) occurred in 11 patients (32%) in the open surgery group, and nine patients (18%) in the endovascular group (Table 3). The overall 90 day post-operative mortality rate was 1.2% ($n = 1$), with one post-operative death in the open surgery group. Eight patients (23%) in the open surgery group had complications requiring re-laparotomy (Table 3). There were four patients (8%) with primary technical failure due to failed attempt and three patients (6%) with endoleak in the endovascular

Table 1. Comorbidities, clinical presentation, and aetiology presented for the endovascular and open groups

Parameters	Total (n=84)	Endovascular (n=50)	Open surgery (n=34)	p value
Age – y	63 (55, 79)	66	62	.26
Men	64 (76)	37 (74)	27 (79)	.57
Median aneurysm diameter –mm	27.5	24.0	32.5	.009
<i>Comorbidities</i>				
History of smoking	58 (69)	36 (72)	22 (65)	.78
Hypertension	55 (65)	33 (66)	22 (65)	.90
Hyperlipidaemia	27 (32)	17 (34)	10 (29)	.66
Diabetes	13 (15)	5 (10)	8 (23)	.10
<i>Clinical presentation</i>				
Incidental	51 (61)	31 (62)	20 (59)	.77
Pain	18 (21)	9 (18)	9 (26)	.35
Rupture	10 (12)	6 (12)	4 (12)	.95
Sepsis	5 (6)	4 (8)	1 (3)	.34
<i>Aetiology</i>				
Atherosclerosis	62 (74)	34 (68)	28 (82)	.14
Mycotic	17 (20)	13 (26)	4 (12)	.11
Genetic	5 (6)	3 (6)	2 (6)	.98

Data are presented as n (%) or median (interquartile range).

group, which were successfully managed by endovascular re-intervention. One patient with intra-abdominal haemorrhage in the endovascular group was treated by endovascular re-intervention and the other patient underwent open surgery. The patient who developed liver necrosis in segment 2/3 after coil embolisation of an intrahepatic aneurysm underwent left hemi-hepatectomy. Aortic dissection and pseudoaneurysm in the common femoral artery were treated conservatively. The one patient who died was haemodynamically unstable at the time of admission due to intra-abdominal haemorrhage from a ruptured common hepatic artery. Emergency laparotomy and vascular reconstruction using an iliac artery allograft was performed. The patient developed post-operative bowel perforation and died due to multi-organ failure on the 13th post-operative day. Autopsy revealed fibromuscular dysplasia. There were no long term (> 90 days) procedure related complications in either group. Patients undergoing open surgery had a longer median stay in the intensive care unit (1 vs. 0 days, $p < .001$) and the surgical ward (8 vs. two days, $p < .001$). Multivariable logistic regression showed a statistically significantly lower rate of

major complications (CCI > 25) in the endovascular group ($p = .048$). Other variables such as age, gender, BMI, smoking, aneurysm diameter, mycotic aetiology, or rupture had no statistically significant impact on the rate of major complications (Table 4). The mean follow up was 6.9 ± 4.2 years. Of the 84 patients included in the study, patency was calculated for stent ($n = 29$) or bypass ($n = 32$). Of these 61 patients, 50 patients (82%) have more than one year of radiological follow up, and 25 patients (41%) more than five years (Fig. 2A). The mean radiological follow up was 5.0 years \pm 4.1 years and log rank test ($p = .26$) showed no statistically significant difference in patency between the endovascular stent and open bypass groups. There were no cases of dilatation or stricture of the allogenic or autologous conduits in the follow up period. Five graft occlusions were identified during follow up (stent graft $n = 1$, autologous $n = 2$, allogenic $n = 2$). All five occlusions were asymptomatic and CT scan demonstrated intrahepatic arterial circulation via collaterals, and they were all treated conservatively. Overall cumulative one, three, and five year patencies for the whole group were 94.6%, 92.5%, and 90.0%, respectively. There were no long term complications

Table 2. Localisation and treatment of 84 hepatic and coeliac artery aneurysms

Localisation	Total (n=84)	Endovascular		Open surgery			
		Stent (n=29)	Coil (n=21)	Autologous (n=15)	Allogenic (n=15)	Synthetic (n=2)	Ligation (n=2)
Coeliac trunk	27 (32)	15	3	2	5	2	–
Common hepatic	31 (37)	8	5	9	9	–	–
Proper hepatic	8 (10)	4	–	2	1	–	1
Right hepatic	4 (5)	1	3	–	–	–	–
Left hepatic	4 (5)	1	1	2	–	–	–
Left gastric	1 (1)	–	–	–	–	–	1
Intrahepatic	9 (11)	–	9	–	–	–	–

Data are presented as n (%) or n.

Table 3. Complications in 84 patients undergoing endovascular ($n = 50$) or surgical ($n = 34$) treatment of hepatic and coeliac artery aneurysms. Five patients in the open group had multiple complications

Endovascular $n = 50$			Open surgery $n = 34$		
Complication	Intervention	Patients	Complication	Intervention	Patients
Endoleak	Re-intervention	3 (6)	Pleural effusion	Drainage	4 (12)
Stent migration	Re-intervention	1 (2)	Pneumonia	Intravenous antibiotics	3 (9)
Haemorrhage	Re-intervention	1 (2)	Pulmonary embolism	Anticoagulation	3 (9)
Haemorrhage	Operation	1 (2)	Haemorrhage	Re-operation	2 (6)
Liver necrosis	Operation	1 (2)	Hepatic artery thrombosis	Embolectomy	2 (6)
Aortic dissection	No treatment	1 (2)	Bile leak	Re-operation	2 (6)
Pseudoaneurysm femoral art.	Compression	1 (2)	Abdominal abscess	Drainage	1 (3)
			Multi-organ failure	Death	1 (3)
Total		9 (18)	Total		11 (32)

Data are presented as n (%).

in patients treated with coil embolisation and no long term re-interventions were needed in the entire study population. The five year overall survival rate (Fig. 2B) was 81.2%, with comparable survival rates in the open and endovascular groups ($p = .49$).

DISCUSSION

Due to the rarity of this condition, few centres have much experience in this field, and the literature is limited to case reports and small case series with limited follow up. Thus, comparable treatment outcomes of true HAA and CAA are difficult to find. Mortality for both elective and urgent patients treated by either an open or endovascular approach is reported to be 6 – 14%,⁶ and post-operative complications range from 24% to 43%.^{1,3,4,6,15,20} The five year patency rates have been reported to be between 79% and 86%.^{3,4} The outcomes in the current study are in line with previous series.

There was no statistically significant difference in pre-operative comorbidities between the endovascular and open surgery groups, and both displayed comparable risk profiles similar to that seen in patients with aortic abdominal aneurysms.²¹ Synchronous aneurysms were detected in 26% of the patients, emphasising the need for liberal use of complete radiological screening in these patients.

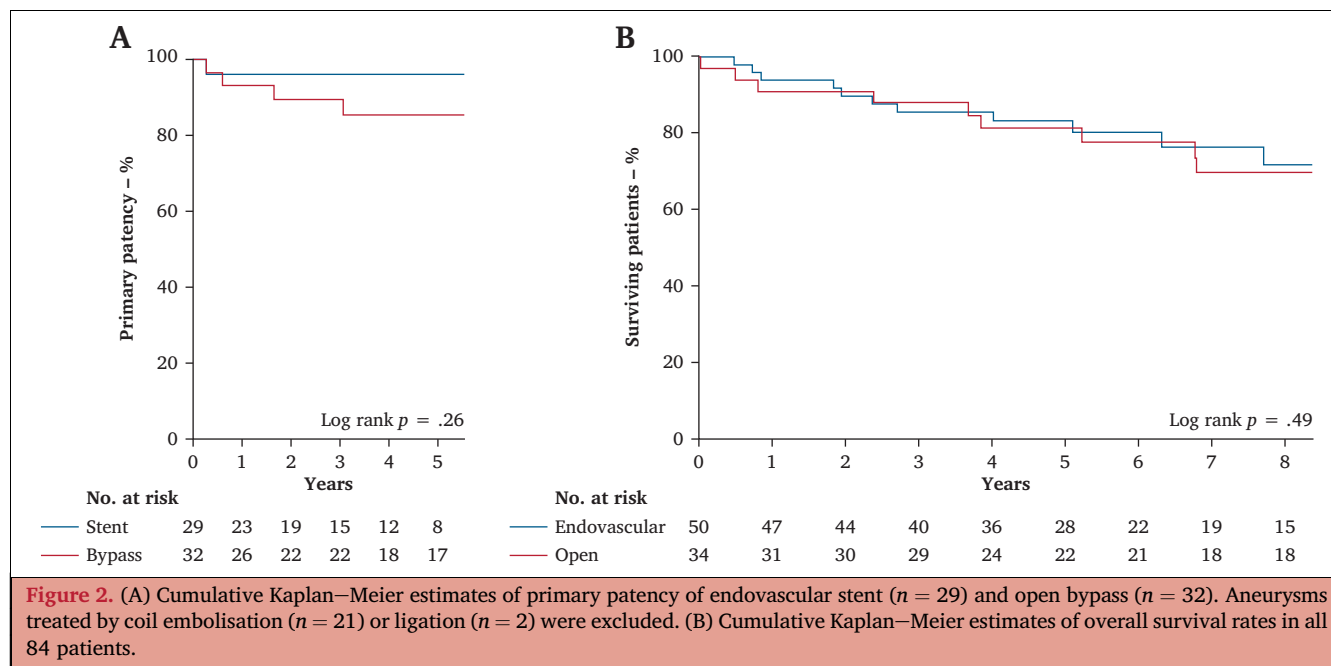
During the last decades, there have been notable changes in the reports of aetiology, diagnostic methodology, clinical presentation, and treatment alternatives for HAA and CAA. Older reports usually describe symptomatic presentation with fatal rupture, mycotic aetiology, and a diagnosis often made at postmortem examination.^{2,22,23} Contemporary data display a different and more diverse range of types and manifestations. As confirmed by the present findings, the most common aetiology today is atherosclerotic aneurysms. However, 20% of the patients had mycotic aneurysms, and this was especially the case in all instances of intrahepatic aneurysms. In comparison, the proportion of abdominal aortic aneurysms of mycotic aetiology is estimated to be between 0.6% and 2%.¹⁹ There were 30% positive bacterial cultures from the aneurysm wall in the open surgery group, which is comparable with bacterial growths in aortic aneurysms.^{24–26} Due to the present authors' experience over the years with a relatively high number of mycotic aneurysms, use of biological graft material has been preferred, whenever possible, to minimise the likelihood of graft infection. If the patient's own great saphenous vein was of unacceptable diameter or quality, allogenic iliac artery was used instead. Prosthetic grafts were used only when biological grafts were not available. This cohort included 15 patients with allogenic vascular reconstruction using blood group compatible donor iliac artery without post-operative immunosuppression. The allogenic and autologous conduits displayed similar long term patency and survival rates comparable with patients treated by endovascular techniques. This suggests that open surgery with a bypass graft is a viable option when endovascular treatment is not feasible.

In the present cohort 39% had a symptomatic presentation. This is more frequent than has been reported in abdominal aortic aneurysms, where the symptomatic rate is 19%.²⁷ Due to the widespread use of cross sectional imaging and ultrasound to diagnose abdominal disease, it is reasonable to expect an increasing incidence of asymptomatic patients with visceral aneurysms.²³ Indication and timing of prophylactic intervention must rely on the risk of rupture balanced against the risk of procedure related morbidity and mortality. Unlike aortic aneurysms, the natural history of HAA and CAA is not well characterised. Using

Table 4. Multivariable logistic regression of risk factors for developing a major complication (CCI > 25) after treatment for hepatic and coeliac artery aneurysms

Variable	OR (95% CI)	p value
Age	1.0 (0.95 – 1.04)	.75
Men	0.7 (0.20 – 2.15)	.49
BMI	0.9 (0.81 – 1.04)	.19
Smoking	0.5 (0.16 – 1.44)	.19
Aneurysm diameter	1.0 (0.97 – 1.02)	.73
Mycotic	1.0 (0.22 – 4.75)	.97
Rupture	2.0 (0.4 – 8.7)	.38
Endovascular treatment	0.3 (0.09 – 0.98)	.048

OR = odds ratio; CI = confidence interval; BMI = body mass index.



the relative increase in aneurysm diameter compared with the normal artery diameter, Røkke *et al.*²⁸ estimated the risk of rupture in HAA and CAA indirectly based on data from aortic aneurysms and estimated the cutoff for treatment to be approximately 20 mm. Some reports indicate that asymptomatic aneurysms < 20 mm can be safely observed due to low risk of rupture,⁴ while others found no association between diameter and rupture.^{1,29} Guidelines¹² note a low level of evidence to guide clinical decision making and conclude that further large scale observation studies with long term follow up are needed. In the present report, three of 10 patients with a ruptured HAA had an aneurysm diameter < 20 mm on presentation. One of the patients suffered from polyarteritis nodosa while the other two had atherosclerosis. Aetiology may play a role, as inflammatory arterial disease may display a more rapid and unpredictable clinical course. This cohort unfortunately does not have sufficient sample size to provide concise guidance, but it is probably advisable to have a somewhat lower diameter threshold for intervention in selected cases of non-atherosclerotic HAA and CAA. Overall, the present results may support current guidelines stating that a HAA with diameter > 20 mm or 25 mm should be considered for repair.

This study has its obvious limitations. During the long study period of 19 years there has been a tremendous improvement in endovascular techniques and potential for less invasive treatment modalities. This could have affected the treatment strategy over time. As endovascular technique was the primary choice of treatment for all patients whenever this was considered technically possible, direct comparison between the two groups is associated with clear selection bias confirmed by larger aneurysms in the open surgery group. As hepatic and coeliac artery aneurysms are relatively rare, single centres will experience few cases over short time spans. To better reflect the current

therapeutic landscape of endovascular and open repair, a multicentre cohort study would be of great scientific value. Furthermore, this is a single centre experience and selection bias in referral practice and institutional treatment may have influenced the results. The retrospective nature of the study poses limitations to the general validity. This is particularly relevant for elucidating the choice of optimal conduit in open repair or factors predicting the risk of rupture. There are also low numbers of patients with radiological follow up at five years. Nevertheless, the authors believe that the present study data provide valuable information on the treatment approach for HAA and CAA due to the large number of patients included and relatively long follow up period.

Conclusion

Endovascular treatment is the preferred approach whenever technically possible. Despite higher post-operative morbidity, an open approach with vascular reconstruction using autologous or allogenic vascular grafts yields acceptable long term results.

CONFLICT OF INTEREST STATEMENT AND FUNDING

None.

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