

## ANEURISMA INFECIOSO DO TRONCO CELÍACO – UMA ENTIDADE CLÍNICA RARA

### *INFECTIOUS CELIAC ARTERY ANEURYSM – A RARE CLINICAL ENTITY*

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### RESUMO

**Introdução:** O aneurisma infeccioso do tronco celíaco é um diagnóstico extremamente raro, com apenas alguns casos reportados até à data. Pretendeu-se assim realizar uma revisão da literatura relativa a esta entidade clínica, com ênfase nas estratégias de diagnósticas e terapêuticas.

**Métodos:** Uma revisão de literatura foi realizada utilizando a base de dados da MEDLINE de acordo com as recomendações PRISMA.

**Resultados:** No total foram identificados 11 casos de aneurisma infeccioso do tronco celíaco. As opções de tratamento foram extremamente variáveis englobando cirurgia aberta e endovascular. A primeira incluiu laqueação do aneurisma ou aneurismectomia com ou sem revascularização com *bypass*. As opções endovasculares incluíram embolização do aneurisma e colaterais, exclusão com recurso a *stentgraft* do aneurisma e num caso a técnica de Chimney foi usada para excluir o aneurisma mantendo a permeabilidade do tronco celíaco. Antibioterapia foi consistentemente uma parte essencial da estratégia terapêutica.

**Conclusões:** Dada a raridade do aneurisma infeccioso do tronco celíaco, a sua história natural não é conhecida. Não obstante, a decisão de tratamento cirúrgico, independentemente do tamanho do aneurisma, é unânime. Os resultados a curto prazo do tratamento endovascular são encorajadores, mas a implantação endovascular de material protésico num ambiente infetado torna a antibioterapia de duração indeterminada aconselhável.

### Palavras-chave

Aneurisma infeccioso; Aneurisma micótico; Tronco Celíaco;

### ABSTRACT

**Introduction:** Infectious celiac artery aneurysm (ICAA) is an extremely rare diagnosis, and only a few cases have been reported in the literature to date. We aimed to review this rare clinical entity, focusing on diagnosis and treatment strategies.

**Methods:** A systematic literature review was performed using MEDLINE database according to the PRISMA guidelines.

**Results:** A total of 11 cases of ICAA were identified in the literature to date. Treatment options were extremely variable and included both open and endovascular surgery. Open surgery included aneurysm ligation or aneurysmectomy with or without revascularization with *bypass*. Endovascular options are increasingly used and include embolization of the aneurysm and collaterals, *stentgraft* exclusion of the ICAA and in one case report, Chimney technique was used to exclude the aneurysm maintaining celiac trunk patency. Unsurprisingly, antibiotherapy was consistently an essential part of the treatment strategy.

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**Conclusions:** Due to the rarity of ICAA, natural history is unclear. Still, surgical treatment is unanimous regardless of aneurysm size. Short term results of endovascular treatment are encouraging, but endovascular implantation of prosthetic material in an infected environment is a concern, so lifelong antibiotherapy and close monitoring are advisable.

## Keywords

Aneurysm, infected (MeSH); Celiac artery (MeSH);

## INTRODUCTION

The name mycotic aneurysm was originally coined by Osler to describe aneurysms associated with bacterial endocarditis.<sup>(1)</sup> Mycotic aneurysms were defined as aneurysms occurring in a normal or atherosclerotic artery and usually resulting from septic emboli of endocardial origin, even though they could occur following other sources of haematogenous infection as pneumonia and osteomyelitis.<sup>(2)</sup>

Recently, the term mycotic has been widely replaced by the term "infectious aneurysm" which may result from primary infection of the arterial wall, due to hematogenous seeding or due to the extension of an adjacent infectious process that then envelops and primarily infects the arterial tissue externally.<sup>(3)</sup> Most primary infected arterial aneurysms (infectious aneurysms) have an eccentric saccular shape as opposed to secondary infection of a preexisting aneurysm (infected aneurysm).<sup>(3)</sup>

Mycotic/Infectious celiac artery aneurysm aetiology are extremely rare. Globally, aneurysms of the celiac artery represent less than 4% of the aneurysms of visceral vessels, and most are atherosclerotic in aetiology.<sup>(4,5)</sup>

Considering the paucity of infectious celiac artery aneurysms (ICAA), the natural history as well as treatment options and prognosis remain to be defined.<sup>(6)</sup>

The purpose of this paper is to report on the clinical presentation, diagnosis, treatment and prognosis of ICAA. Accordingly, a systematic literature review was performed in order to identify all reported cases of ICAA.

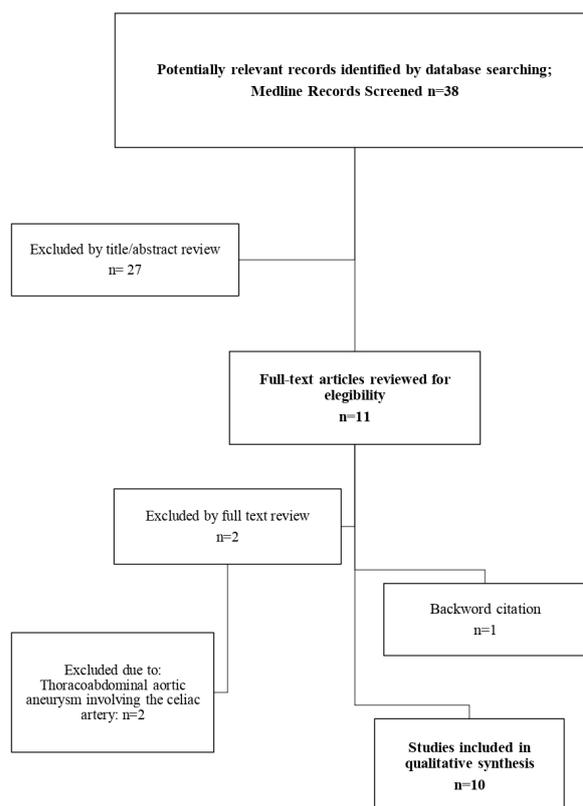
## METHODS

A systematic review was conducted according to the recommendations of the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) statement.<sup>(7)</sup>

MEDLINE database was searched with the following query: ((“coeliac artery”[All Fields] OR “celiac artery”[MeSH Terms] OR (“celiac”[All Fields] AND “artery”[All Fields]) OR “celiac artery”[All Fields]) OR (“celiac artery”[MeSH Terms] OR (“celiac”[All Fields] AND “artery”[All Fields]) OR “celiac artery”[All Fields] OR (“celiac”[All Fields] AND “trunk”[All Fields]) OR “celiac trunk”[All Fields])) AND ((“aneurysm”[MeSH Terms] OR “aneurysm”[All Fields] OR (“aneurysm, false”[MeSH Terms] OR “aneurysm”[All Fields] AND “false”[All Fields]) OR

“false aneurysm”[All Fields] OR “pseudoaneurysm”[All Fields])) AND (Mycotic[All Fields] OR infectious[All Fields])

The eligibility criteria included any publication reporting ICAA cases. Exclusion criteria included articles published in a language other than portuguese, english or Spanish. No time frame was instituted as an exclusion criterion in this systematic revision, considering the rarity of the clinical condition. Two reviewers initially screened the identified studies. Further assessment of articles that potentially met inclusion criteria was performed using the full text. (Figure 1) Data collected and analysed included clinical presentation, diagnosis, treatment, follow-up and prognosis.



**Figure 1** PRISMA diagram summarizing literature screening process. PRISMA – Preferred Reporting Items for Systematic reviews and Meta-Analyses



**Table 1** Summary of clinical prognosis, diagnosis, treatment and prognosis of patients with ICAA

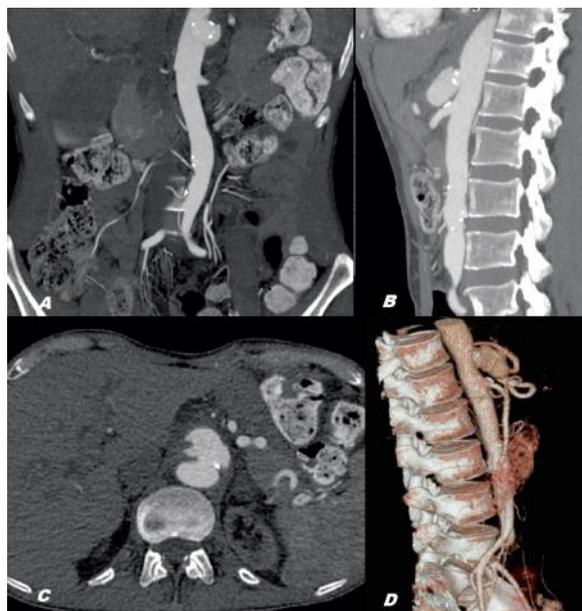
Article	Year	n	Age	Gender	Presumptive etiology	Microbiologic agent	Clinical Presentation	Imaging
Sweany, H.C. et al	1919	1		Male	Infective endocarditis	NE	Asymptomatic	Autopsy feature
Zeppa, R. et al	1966	1	19	Female	Septicemia	Staphylococcus Aureus	Pulsatile mass in the epigastrum	DSA: poorly defined vascular mass occupying most of the right upper quadrant
Werner, K. et al	1991	1	19	Female	Infective endocarditis	Culture negative	Fever + Epigastric pain	CTA: ICAA + SMA aneurysm
Carrel, D. et al	1992	2	61	Male	Septicemia	Staphylococcus Aureus	Cough + Hemoptisis + Abdominal pain	CT: Gastroesophageal mass; Autopsy: ruptured ICAA + hemothorax
			73	Male	Septic Arthritis	Staphylococcus Aureus	Hemoptisis + Abdominal pain	Autopsy: Ruptured ICAA + Hemothorax
Viglione, G. et al	1993	1	39	Male	Infective endocarditis	NE	Fever + epigastric pain	DSA: Thrombosed celiac trunk aneurysm
Serafino, G. et al	2005	1	54	Male	Unkown	Staphylococcus Aureus + Streptococcus Sanguis	FUO + Hypovolemic shock 2 weeks later	CTA: Ruptured ICAA
Aki, A. et al	2012	1	60	Male	Infective endocarditis	Streptococcus spp	Abdominal pain in a patient admitted for bacterial endocarditis	CTA: ICAA growth from 17 mm to 50 mm with pseudoaneurysm suspicion
Batagini, N.C. et al	2015	1	56	Male	Infective endocarditis	Staphylococcus Aureus (MRSA)	FUO	CTA: ICAA 40x32mm width; Absence of proximal normalizing neck
Tanaka, M. et al	2017	1	48	Male	Infective endocarditis	Staphylococcus Aureus (MSSA)	FUO	CTA: 12 mm width ICAA with progressive growth; 6mm CHA aneurysm;
Coelho, A. et al	2018	1	54	Male	Bacteremia due to AVF cannulation technique	Staphylococcus Aureus (MSSA)	FUO + Lumbar/epigastric pain	CTA: 32 mm ICAA with no normalizing proximal neck

CHA: Common hepatic artery; CTA: Computed Tomography Venography; FUO: Fever of unknown origin; ICAA: Infectious celiac artery aneurysm; MRA: Magnetic Resonance Angiography; NA: Not applicable; NE: Not specified;

	Treatment	Antibiotherapy	Outcome	Follow-up
	NA	NA	Death	NA
	Aneurysmectomy + celiac artery-distal splenic artery bypass with autologous conduit *celiac trunk anatomic variant with only splenic artery emerging	Penicillin course	Uneventful recovery; Normal liver function at 2 years;	2 years
	Celiac trunk and SMA aneurysms ligation	6-week antibiotic course not otherwise specified	Uneventful liver function recovery.	NE
	NA	NA	Death	NA
	NA	NA	Death	NA
	Aneurysmectomy + Hepatic artery end-to-side anastomosis to the superior mesenteric artery	NE	Chronic diarrhea possibly related to celiac plexus stimulation during dissection	NE
	Celiac artery and common hepatic artery ligation + splenectomy	Amoxicillin + clavulan acid course	Uneventful recovery	1.5 years
	Embolization of the aneurysm and collaterals	Ampicillin + gentamicin course	Uneventful recovery; Normal perfusion of liver and spleen	1 year
	Aortohepatic bypass with autologous conduit + Aneurysm ligation	Vancomycin course (6-weeks)	Uneventful recovery	NE
	Embolization using platinum coils of the proximal CHA, and main splenic artery as well as in the collaterals that supplied blood to the aneurysms	6-week antibiotic course not otherwise specified	Uneventful recovery	NE
	Chimney exclusion of the aneurysm	Lifelong cefazolin (iv) on haemodialysis sessions	Uneventful recovery	3 months

## RESULTS

A total of 38 potentially relevant articles were initially selected, based on the initial query. After analysis of title and abstract, literature review retrieved a total of 9 articles reporting 10 cases of ICAA.<sup>(4, 6, 8-15)</sup> An additional case report was retrieved by backword citation. (Figure 1). Full text analysis of reported cases of ICAA revealed male predominance (n=9; 81.8%) and that the leading etiology was infective endocarditis (n=6; 54.5%). Two cases were due to unspecified septicemia and one case was presumed to be the consequence of knee septic arthritis. In another case bacteremia with a skin ubiquitous agent (*Staphylococcus Aureus*) that ultimately caused the infectious aneurysm was presumed to be a complication of hemodialysis arteriovenous fistulae puncture technique (Buttonhole technique). *Staphylococcus Aureus* was the leading microbiologic agent (n=6; 54.5%). (Table 1) Clinical presentation varied from asymptomatic (n=1) to symptoms as variable as pulsatile abdominal mass (n=1), fever of unknown origin with or without abdominal pain (n=8) and 2 reports of hemoptysis. (Table 1) Diagnosis was determined based of autopsy features in 2 cases. In the remaining cases, Digital Subtraction Angiography (DSA) or Computed Tomography Angiography (CTA) were crucial to reach the diagnosis of ICAA. Figure 2 demonstrates CTA images of an ICAA. In almost all cases the aneurysms were isolated, with the exception being one case of an ICAA with a synchronous common hepatic artery aneurysm.<sup>(14)</sup>



**Figure 2** Coronal (A), Sagittal (B), Axial (C) and 3-d (D) images of an ICAA CTA previously reported in the literature<sup>(15)</sup>; CTA: Computed Tomography Venography; ICAA: Infectious celiac artery aneurysm;



Microbiologic confirmation of diagnosis was reached in patients submitted to open surgery. In the remaining cases, presumptive infectious etiology was based on clinical and imagiologic features. (Table 1)

As for treatment options, they were variable and included both open and endovascular surgery options. In the first, treatment options included aneurysm ligation and aneurysmectomy with or without revascularization with bypass. Endovascular options included embolization of the aneurysm and collaterals. In one case report, Chimney technique was used to exclude the aneurysm maintaining celiac trunk patency. (Table 1)

In all cases antibiotherapy was an essential part of treatment, in most cases with a 6-week course. In the Chimney technique case, life-long antibiotherapy was maintained on hemodialysis sessions due to concerns with stentgraft infection. (Table 1)

These reports had a maximum follow-up of two years, with 3 early deaths to report (25%). One case of chronic diarrhea possibly related to celiac plexus stimulation during dissection was reported with uneventful recovery in the remaining cases. (Table 1)

## DISCUSSION

Infectious aneurysms are a rare but potentially life-threatening condition, due to the elevated rupture risk.<sup>(6)</sup>

In contrast to the preantibiotic era, when most infected aneurysms were associated with bacterial endocarditis, the majority of infected aneurysms now occur in intravenous drug abusers, in patients with depressed immunity such as those with diabetes mellitus, chronic illnesses, or malignancies and after invasive intravascular procedures. Accordingly, incidence of infected aneurysms does not seem to be decreasing.<sup>(16)</sup> In addition, the dissemination of imagiologic diagnostic tools certainly has contributed to an increasing diagnostic yield.

Isolated infectious celiac artery aneurysm is an exceedingly rare diagnosis, with only 12 reported cases in the literature.<sup>(4,5,8,10-13)</sup>

Whereas the intact arterial wall is highly resistant to infection, congenital defects and intimal diseases (such as atherosclerotic involvement) can render the arterial wall susceptible. The most common sources of arterial infection are urinary tract infection, gastrointestinal infection, salmonellosis, respiratory infection, cellulitis, osteomyelitis, wound infection, dental extraction, and intravenous line sepsis.<sup>(17)</sup>

Presumptive infectious aetiology is usually based on CTA findings, including saccular anatomy, rapid growth, periarterial edema appearing as fat stranding or as hypoattenuating concentric rim and perianeurysmal gas.<sup>(18)</sup> Clinical and analytic findings including fever, back pain, elevated inflammatory markers and bacteraemia also point towards infectious etiology. Aneurysms of the celiac artery, together with those of the superior mesenteric artery, are more often symptomatic than other

visceral artery aneurysms.<sup>(19)</sup> In fact, infectious aneurysm must be ruled out in patients with fever, abdominal or back pain, and a pulsatile abdominal mass, as it is the most frequently described clinical presentations. Leucocytosis and an elevated erythrocyte sedimentation rate are often present, but are nonspecific findings, and blood cultures might be negative.<sup>(20)</sup>

When in rupture patients may present with hypovolemic shock or with a combination of haemoptysis and haemothorax. (Table 1) Due to the rarity of ICAA, natural history and prognosis are unclear. A unique case report documented CTA evolution from normal celiac artery with minimal haziness of the surrounding fat to celiac aneurysm rupture within 2 weeks, enhancing the rapid and unpredictable nature of ICAA.<sup>(13)</sup> Therefore, even though criteria for intervention has not been well established, consensus exists for repair regardless of size.<sup>(6)</sup>

Regarding open surgery, previous reports included aneurysm ligation with or without revascularization with autologous bypass. Celiac aneurysm ligation without reconstruction of the arterial blood flow has been reported in several cases. In this situation, collateral circulation to the liver depends on the gastroduodenal artery. Graham et al, described 14 cases of celiac artery ligation without reconstruction of blood flow, reporting one death due to hepatic necrosis related to interruption of collateral pathways during extensive surgical dissection and one death due to superior mesenteric artery occlusion with thrombus on which collateral circulation depended.<sup>(21)</sup>

In 2015, Batagini et al also described a case of a ICAA with a similar anatomy that was treated with a two-stage approach: First, transperitoneal aortohepatic bypass with autologous conduit with ligation of the celiac artery and second, retroperitoneal aneurysmectomy. (Table 1)<sup>(6)</sup> Although effective, this approach seems rather aggressive in patients who are frequently frail due to the infectious disease.

Accordingly, Saltzberg et al recommended endovascular repair as a first line of treatment for all visceral artery aneurysms in anatomically suitable cases, excluding those located in the distal splenic artery.<sup>(22)</sup>

Two previous reports of endovascular treatment of ICAA involved its embolization. (Table 1)<sup>(8,9)</sup> Aki et al described an endovascular isolation and ICAA packing using N-butyl cyanoacrylate with embolization coils, with no recurrent infection or aneurysm recanalization in a 12-month follow-up.<sup>(8)</sup>

Shu et al described a tuberculous pseudoaneurysm involving the aorta and celiac artery that was excluded by placing an aortic stent graft with celiac origin coverage.<sup>(23)</sup> Chimney technique was used to exclude a ICAA in one clinical case, given the unique anatomy of the aneurysm with involvement of the ostia of the celiac artery with no proximal normalizing neck.<sup>(15)</sup> These two approaches allowed the maintenance of celiac artery patency, thereby diminishing the risk of hepatic necrosis.

Even though these endovascular approaches offer a minimally invasive and effective solution to a problem with a technically complex major open surgery alternative, implantation of prosthetic material in an infected environment is a concern. Although a few case reports showed good outcomes with endovascular approach associated with antibiotic therapy, long-term follow-up periods are currently lacking. Lifelong antibiotherapy and close imagiologic and analytic monitoring may be advisable due to theoretical concerns for recurrent infection or progression.<sup>(20)</sup> However, given the lack of knowledge on natural history, no follow-up guidelines are available. We should emphasize the small number of included patients in this systematic review, related to the rarity of this diagnosis, limiting the ability to make general recommendations regarding diagnosis, treatment and follow-up. The main limitation of this study is, not surprisingly, publication bias as probably most ICAA diagnosed were not reported. In conclusion, ICAA is an incidental finding in CTA in patients being studied for fever of unknown origin with or without abdominal pain. Consensus exists on treatment regardless of size, and endovascular treatment seems to be an effective minimally invasive solution, with low early morbi-mortality. Still long term data is currently lacking.

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