

Calcified pancreatoduodenal artery aneurysm with filiform celiac stenosis: Follow up for ~6 years (70 months) with no intervention

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Abstract

A 72-year-old male patient was investigated with abdominal ultrasound (US) imaging before the coronary artery bypass graft surgery (CABG). The incidental finding on ultrasound was an aneurysm of the superior mesenteric artery branch. Consequent CT angiography confirmed aneurysm of the inferior pancreaticoduodenal artery (PDAA, size = 25,5x31,0x28,5mm) with filiform celiac artery stenosis. According to the European Guideline asymptomatic >25mm sized PDAA are treated with an urgent repair. However, the discrepancy between PDAA size and risk of rupture and individual parameters of the patient led the multifunctional team to assign regular follow-ups with US imaging. This case report describes rare pathology, which has been followed up for the last ~6 years (70 months) annually with no intervention, and discusses complexities in making decisions in the management of PDAA with celiac artery stenosis.

Key words: pancreatoduodenal artery aneurysm, celiac artery stenosis, visceral artery aneurysm, rare cases

Introduction

Pancreaticoduodenal artery aneurysms (PDAA) with celiac artery (CA) stenosis are rare cases with approximately 120 cases described in the literature between 1970 and 2022. Because of the risk of PDAA rupture and the associated high mortality rate with it (50%), the common interventions in the management include endovascular repair with either coil embolization and/or stent graft placement, and open surgery, which in most cases reserved for hemodynamically unstable patients in case of rupture [1,2]. As the PDAA rupture is not related to the size of the aneurysm, it makes challenging when to plan embolization/surgery and when to follow up without intervention in asymptomatic patients. The first guideline about PDAA management, Clinical Practice Guidelines of the European Society of Vascular Surgery (ESVS, published in 2017 - a year after the current case), suggests urgent repair of the

symptomatic aneurysms irrespective of the size and location, while asymptomatic aneurysms with the size of >25mm are suggested to be repaired [3]. ESVS also suggests asymptomatic patients with <25mm aneurysms be followed up every 2-3 years with CT or US. In this case report, we demonstrate an asymptomatic >25mm aneurysm (which according to ESVS should have been repaired) where intervention was considered to be unnecessary by the multidisciplinary team, with annual follow-up imaging for the last ~6 years (70 months). This case is the largest PDAA and the longest PDAA follow-up reported in the literature with no intervention so far [3].

Case presentation

The 72-year-old male was admitted to the ER department with chest pain. Based on the clinical symptoms and ECG results the patient was diagnosed

with ST-elevation myocardial infarction (STEMI) and received full anti-MI treatment with fibrinolytic in the hospital because of the unavailability of coronary angiography. In the next eight hours, the patient was taken to angiography and diagnosed with multivessel coronary artery disease with up to 80% occlusion of the circumflex artery and up to 60% occlusion of other main coronary arteries. The patient has been suggested CABG, and explained the risks and benefits of the procedure, following which informed consent was obtained. During the abdominal US examination, as part of the pre-operative assessment, an aneurysm of the superior mesenteric artery branch was revealed (Figure 1). Consequent CT with contrast revealed inferior pancreaticoduodenal artery aneurysm (fusiform shaped, size: 25,5 x 31,0 x 28,5 mm) and filiform stenosis of the celiac artery (D=2-3mm). The walls of the aneurysm were calcified (Figure 2). Considering the asymptomatic flow of the current pathology, potential ischemic injury to corresponding organs (liver, spleen, and stomach) in case of intervention in addition to celiac artery stenosis, technical difficulties associated with the fusiform shape

of the aneurysm and its anatomic localization multidisciplinary team decided to follow up the aneurysm and proceed to CABG. After successful cardiac surgery (April 2016), the patient was discharged home on the 17th day of hospitalization. Following CABG the patient's follow-up has been planned every 6 months for the first year and annually thereafter.

Sixth months after CABG (January 2017), on regular follow-up CT angiography with a 3D reconstruction of PDAA was made (Figure 2b). The size of the aneurysm was consistent with prior CT measurements with no change in diameter. As the patient was asymptomatic, he has been taken to further regular follow-up. During the last 70 months we are observing the absence of clinical symptoms, the lack of PDAA increase in size, and the presence of a calcified wall of the aneurysm with up to 2.43mm in thickness (January 2022) (Figure 3). Listed clinical and investigational findings show us no hemodynamic changes and absence of an increase in the size of the aneurysm. Currently, the patient is on continual US follow-up.

Figure 1 - US findings of the superior mesenteric artery branch aneurysm in B- (a) and Color Doppler (b) mode with up to 30mm in diameter.

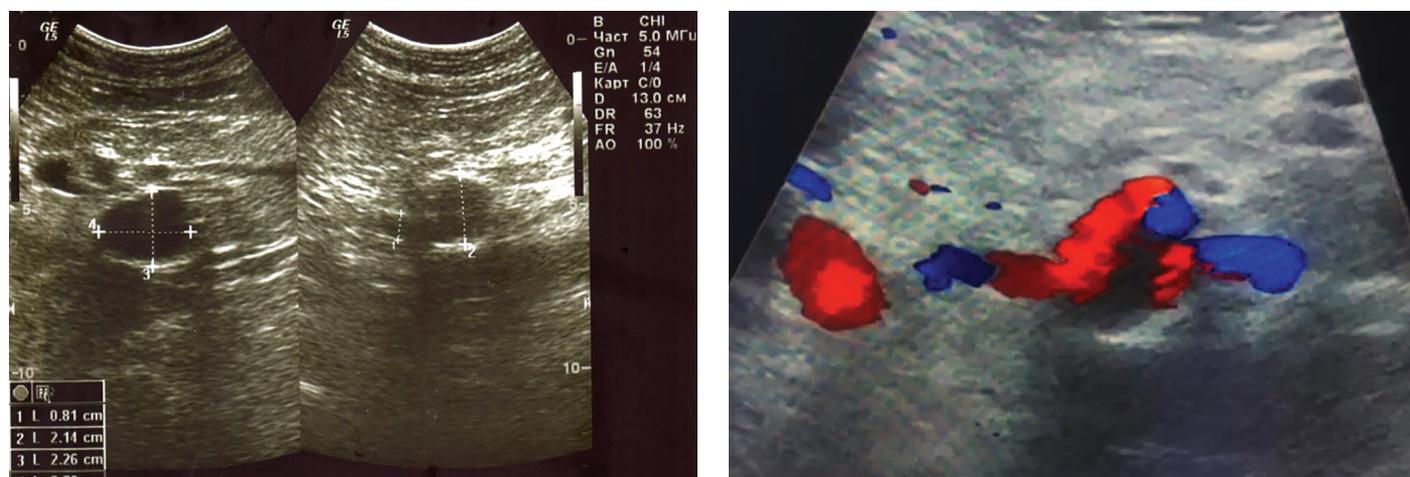


Figure 2 - Coronary angiography has shown the filiform stenosis of the celiac artery up to 1.5mm in diameter (a, b – vertical yellow arrow) and pancreaticoduodenal artery aneurysm with the size of 25,5 x 31,0 x 28,5 mm (horizontal blue arrow) with calcifications of its wall (blue arrowhead).

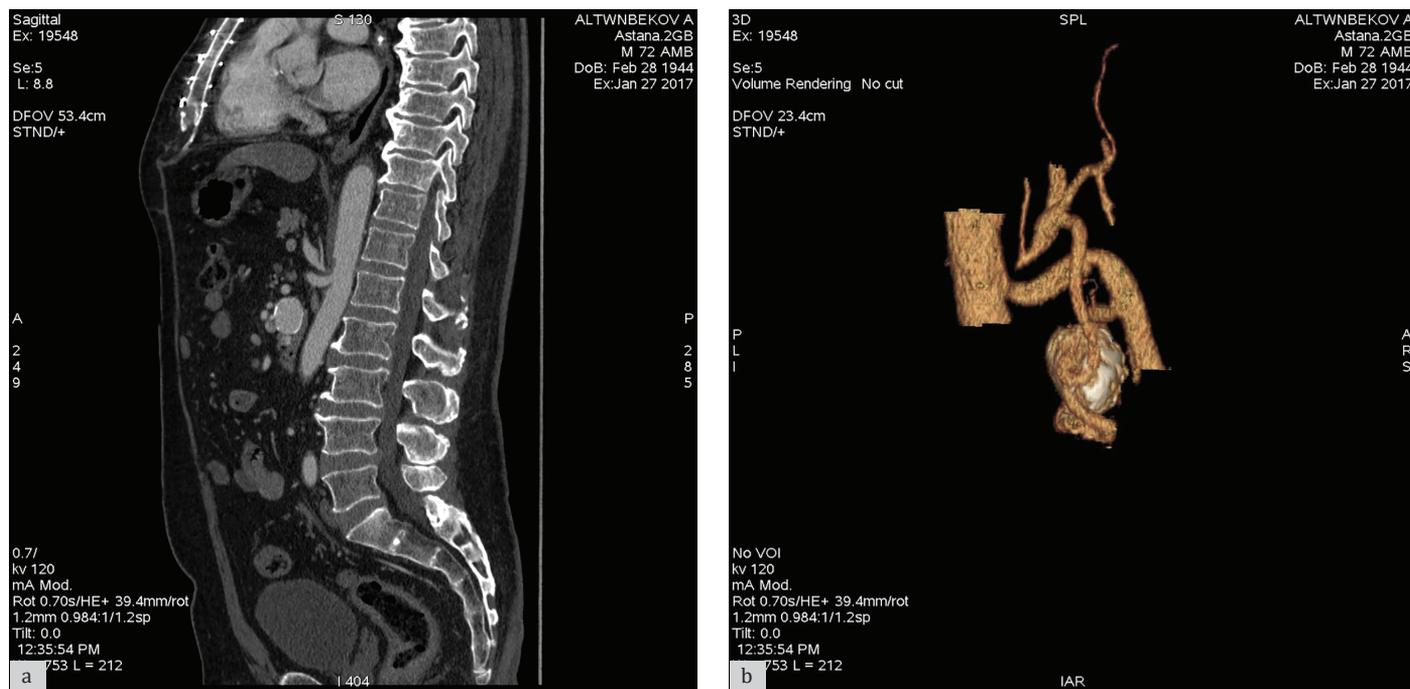
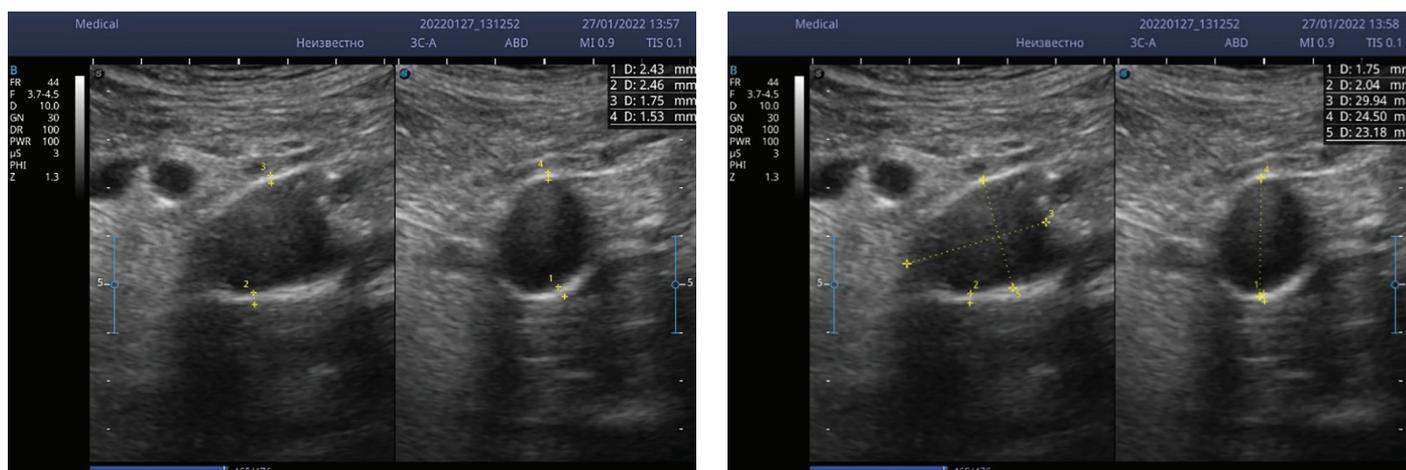


Figure 3 - Last US imaging of PDAA (January 2022): shows the calcified wall with up to ~2.46mm thickness and unchanged diameter of the aneurysm (D~29.94mm) compared to the size ~6 years ago (Figure 2). Note: Slight deviation in the size measurements on US and CT is acceptable, because of difficulty to get the same cut on the US as of CT.



Discussion

Pancreaticoduodenal artery aneurysms (PDAA) are amongst the rarest pathology of all visceral artery aneurysms (2%) along with the gastroduodenal artery (1.5%) and inferior mesenteric artery (rarest) [4]. Most PDAA are associated with celiac artery stenosis (68-74%) [5]. The development of PDAA is explained by the first development of celiac artery stenosis or occlusion (atherosclerotic lesions, etc.) which leads to an increased pressure in PDA, and in gastroduodenal arteries (GDA), leading to an increased artery wall tension, with subsequent arterial wall weakening and aneurysm development [5]. However, Hye Jeong Yoon et al. have shown that a reverse causal relationship may also be possible, theoretically, demonstrating it based on the simulation using an electric circuit, where the development of aneurysm may result in proximal artery stenosis or occlusion, may it be a superior mesenteric artery or celiac trunk [6].

In the majority of cases, patients with PDAA present with abdominal pain (54%) and in 26% of cases, the pathology is discovered incidentally [7]. A review of PDAA cases has shown that the size of the PDAA with rupture ranged between 0.6cm and 2.0cm, while the unruptured PDAA ranged between 0.7cm and 6.0cm [8]. As the mortality associated with the rupture of the PDAA is high (50%) and the rupture is not related to its size, it brings a dilemma in the management plan, leaving us a new question of whether to include the size of the aneurysm into the criteria in the management plan or not, if yes then what should be the threshold for non-surgical/surgical intervention. Takao et al. reviewed the largest series of unruptured PDAA with no intervention applied to 8 aneurysms in 5 patients. They have proposed that the "risk of rupture of true PDAA might be lower than expected from the data on ruptured aneurysms; however, careful follow-up of untreated aneurysms is necessary". Most notably, the decision to follow up the PDAA with celiac stenosis without intervention was made based on the potential risk of ischemic injury to celiac artery/SMA-supplied organs (liver, spleen, and stomach) and based on the unclear risk of complications after PDAA embolization, despite the absence of reported complications on literature directly associated with the procedure itself. However, in several cases, patients developed other complications like aneurysmal rupture or infection after the intervention. The message of the case series was to draw attention to "balance the risk of aneurysm rupture against the risk of complications from preventive treatment" [9].

In 2017 (a year after the current case), the ESVS was made the first guideline to clarify when to proceed to intervention and when to observe [10]. ESVS divided people with PDAA into two categorical groups, based on symptoms (symptomatic or not) and size (>25mm or <25mm). Symptomatic patients and/or >25 mm-sized aneurysms were absolute indications for urgent surgical repair. While the only criteria for follow-up were asymptomatic patients with D<25mm [3].

To sum up, the current case demonstrates the independence of PDAA aneurysm size with its rupture for the follow-up period between April 2016 and January 2022 (70 months). The decision not to intervene with the patient but to follow up was made based on the patient's parameters (age 73y/o, arterial hypertension, DM type2, MI), absence of symptoms, characteristics of the aneurysm (wall calcification, fusiform shape) and the presence of celiac stenosis, which might have increased complications in case of intervention.

However, current pathology needs further investigations and statistical analysis to find out the risks, which might be directly or indirectly associated with PDAA development and with its outcome in case of intervention or follow-up. We suggest including in the PDAA management criteria blood flow dynamics including Intra aneurysmal velocity (1), which has a direct effect on the wall shear stress, also the aneurysmal wall characteristics (2), such as the presence of calcifications and aneurysmal wall thickness.

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