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Case Report

Pancreatic neuroendocrine tumor with aneurysms of the gastroduodenal artery: a case report☆,☆☆,★,★★

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ABSTRACT

Gastroduodenal artery (GDA) aneurysm is a very rare condition. It is divided into false aneurysms (pseudoaneurysms) associated with pancreatitis and true aneurysms secondary to celiac trunk stenosis. We report a 24-year-old patient who was diagnosed with pancreatic head neuroendocrine tumor and was incidentally found to have multiple GDA aneurysms in the absence of celiac artery stenosis. The aneurysms were embolized because of the presumed high risk of bleeding. The procedure was successful with no recurrence on follow-up computed tomography scan.

1. Introduction

The gastroduodenal artery (GDA) typically arises from the common hepatic artery of the celiac trunk and supplies blood to the proximal part of the duodenum, pylorus, and pancreatic head. It first branches into the supraduodenal artery, followed by the posterior superior pancreaticoduodenal artery and terminates as the right gastroepiploic artery and the anterior superior pancreaticoduodenal artery. These branches form anastomoses with the superior mesenteric artery (SMA) through the anterior and posterior pancreaticoduodenal arcades providing an important source of collateral circulation.

GDA aneurysm is uncommon [1] comprising only 1.5% of all reported visceral artery aneurysms (VAAs) [2]. The majority of those are pseudoaneurysms occurring in the setting of pancreatitis [3]. True aneurysms are most commonly associated with middle-aged men [4] and atheromatous celiac trunk stenosis [5]. We report a 24-year-old woman, presenting with a pancreatic head neuroendocrine tumor and multiple GDA aneurysms in the absence of celiac artery stenosis.

2. Case report

A 24-year-old female patient presented to our institution with severe epigastric pain. She was diagnosed with a pancreatic head neuroendocrine tumor.

Laboratory investigation revealed hypercalcemia with a calcium level of 20.7 mg/dl and aspartate aminotransferase (AST) and alanine aminotransferase (ALT) around 130–140 IU/L [normal ranges of ALT (7–55 IU/L), AST (8–48 IU/L)].

Computed tomography (CT) angiography of the abdomen showed a heterogeneously enhancing pancreatic head mass with areas of necrosis and calcification and multiple arterial aneurysmal dilatations in the pancreaticoduodenal arcade, the largest of which measured 1.5 cm arising from the anterior superior pancreaticoduodenal artery with peripheral calcification. Multiple metastases to liver and bone were also identified. There were no CT findings suggestive of pancreatitis or celiac artery stenosis (Fig. 1).

Digital subtraction arteriogram after embolization of the right hepatic artery for treatment of hepatic metastases showed multiple gastroduodenal arcade aneurysms (Fig. 2).

A decision to coil the aneurysms was taken in order to decrease the risk of rupture and bleeding. The pancreaticoduodenal artery was catheterised, and angiography confirmed multiple aneurysms in the pancreaticoduodenal arcade. These aneurysms were excluded from the circulation using multiple microcoils and microparticles.

Final angiogram postembolization did not show any further filling of the aneurysms (Fig. 3).

☆ Ethical Approval: For this type of study, formal consent is not required.
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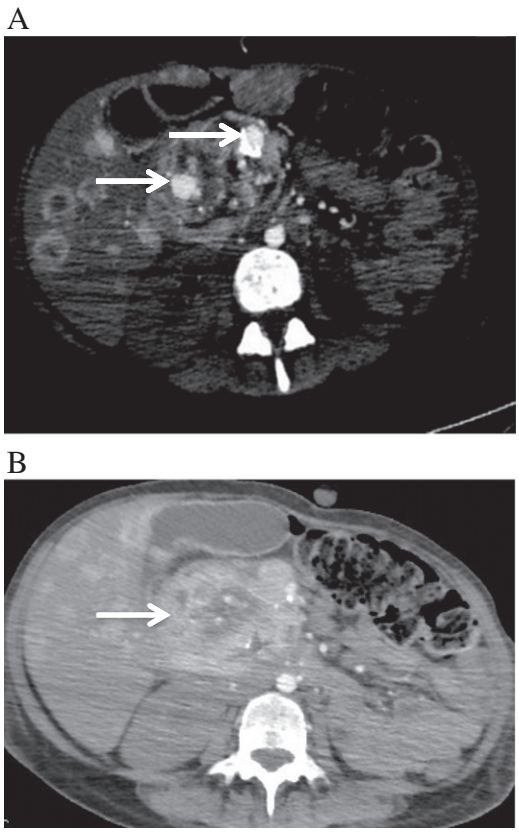

Fig. 1. (A) CT angiography showing two aneurysms (straight arrows) within the pancreatic head, one of which is partially calcified; (B) Axial CT in the arterial phase showing a heterogeneously enhancing mass (arrow) in the head of the pancreas compatible with neuroendocrine tumor.

Follow-up CT angiography 5 days postprocedure confirmed absence of flow in the aneurysms.

3. Discussion

GDA aneurysms are divided into true and false aneurysm, the latter being more common and frequently seen in the setting of acute and chronic pancreatitis. Periarterial inflammation caused by pancreatic proteolytic enzymes secondary to pancreatitis is an important cause of GDA pseudoaneurysms [6]. Atherosclerosis, congenital absence of celiac axis, and autoimmune diseases like systemic lupus erythematosus, Wegener's granulomatosis, and polyarteritis nodosa are possible etiologies of true GDA aneurysms [7].

The pancreaticoduodenal arcade is the main collateral pathway between the celiac axis and the SMA. Increased blood flow in this arcade, as compensation for celiac artery stenosis, is associated with pancreaticoduodenal artery aneurysm [8].

The same theory suggests that occlusion or stenosis of the SMA or celiac axis could predispose to the formation of a GDA aneurysm [9]. After researching online databases, including PUBMED and Medline, we did not find any reports of pancreatic head neuroendocrine tumor associated with GDA aneurysms. It is possible that in this case, the presence of the hypervascular neuroendocrine tumor induced an increased flow through the GDA arcade, leading to aneurysms formation.

Another possible etiology is the tumor induced neovascularization. Pancreatic neuroendocrine tumors exhibit a high degree of vascularization [10] and are also known to secrete many proangiogenic molecules, in particular, vascular endothelial growth factor [11]. This could lead to the formation of numerous friable vessels prone to aneurysmal dilatation secondary to increased blood flow. A third possible etiology is that the tumor may have an aneurysmal component secondary to its histological origin. This is similar to an angiomyolipoma which derives from perivascular epithelioid cells. It contains fat, smooth muscles, and vessels, the latter of which develop aneurysms. Even though there is a possibility of a potential common pathogenesis of neuroendocrine tumors and aneurysms, to our knowledge, we found no evidence linking the two pathologies in the literature.

The gold standard diagnostic test is visceral angiography, serving both diagnostic and therapeutic purposes by delineating the arterial anatomy and allowing therapeutic intervention [12]. It has the highest sensitivity (100%) followed by CT (67%) [12].

Ruptured GDA aneurysm may lead to death in up to 40% of cases [13]. Open surgical and endovascular techniques are available to treat GDA aneurysms. Endovascular techniques are more often used to treat VAAs with very high success rate and low recurrence, morbidity, and mortality [1]. Endovascular treatment aims at excluding aneurysms from the parent circulation. Treatment options include coiling (such as in our patient), stent graft placement or balloon occlusion, and embolization with liquid agents such as glue, onyx, or thrombin [14]. We found no evidence that favors one method over the other. However, the use of stents and balloon occlusion in a tortuous and tapered artery is challenging [15]. In addition, when injecting liquid agents, there is possibility of reflux into the patent parent arteries causing thrombosis [15]. For these reasons, and given the complexity of this case and the size of the tumor, decision favored endovascular coiling. The procedure was successful with complete occlusion of the aneurysms.

4. Conclusion

GDA aneurysms are uncommon, and there are no known associations between GDA aneurysms and pancreatic neuroendocrine tumors. We describe a patient with pancreatic neuroendocrine tumor with multiple aneurysms successfully treated with endovascular therapy and present possible flow and tumor-related etiologies for this unusual presentation.

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