Rapid Formation and Hybrid Treatment of a Large Superior Mesenteric Artery Aneurysm

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Patient: Male, 41-year-old
Final Diagnosis: Superior mesenteric artery aneurysm • visceral aneurysm
Symptoms: No symptoms • asymptomatic
Clinical Procedure: —
Specialty: Surgery

Objective: Unusual clinical course
Background: Superior mesenteric artery (SMA) aneurysms account for about 5.5% of all visceral aneurysms, and are most commonly secondary to infectious causes or dissection. They tend to expand and rupture. Here, we present our successful diagnosis and treatment of a 41-year-old man with asymptomatic coeliac trunk stenosis, in whom the large aneurysm of the branch of the SMA developed in a very short time after conservative treatment of plastron appendicitis.

Case Report: A 41-year-old man was diagnosed with plastron appendicitis during abdomen ultrasound (US) examination. Following 2 weeks of conservative treatment with intravenous antibiotic therapy, complete resolution of symptoms was obtained and confirmed in the computed tomography (CT) scan, and no other pathologies were diagnosed. Three weeks later, during the US examination, a 33-mm aneurysm of the branch of the SMA was diagnosed. The patient was admitted to the Vascular Surgery Department, where a critical stenosis of the coeliac trunk secondary to the compression by median arcuate ligament and a 33-mm true visceral aneurysm of one of the branches of the SMA were diagnosed. Successful treatment of the aneurysm was performed. Surgical decompression of the coeliac trunk and subsequent elective endovascular embolization of the SMA aneurysm with angioplasty of the coeliac trunk were performed. The postoperative period was uneventful and the patient was released from the hospital and remains asymptomatic.

Conclusions: Visceral artery aneurysm can form very quickly. In some of the aneurysms, a combination of open surgical and endovascular methods should be performed.

Keywords: Aneurysm • Embolization, Therapeutic • Endovascular Procedures • Median Arcuate Ligament Syndrome

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**Background**

Visceral artery aneurysms are rare, with a prevalence of 0.01-0.2% in the general population [1,2]. Visceral artery aneurysms most commonly affect the splenic artery (60%), followed by the hepatic artery (20%), the celiac artery (4%), pancreatic branches (2%), gastroduodenal artery (1.5%), and inferior mesenteric artery (1%) [3,4].

Most (almost 95%) of the aneurysms are asymptomatic [5]. Risk factors for aneurysm formation include connective tissue diseases and inflammatory and genetic conditions. The role of atherosclerosis in visceral aneurysm formation seems to be marginal [6-8].

SMA aneurysms are most commonly secondary to an infectious cause or dissection. Coeliac trunk stenosis and occlusion (eg, Dunbar syndrome), which is associated with compensatory increased blood flow and enlargement of interconnected collaterals causing vessel wall weakening, may play a role [9,10]. They tend to expand and rupture (about 38-50% of patients present with ruptured aneurysm, with a mortality rate of 30-90%). The mortality rate in elective SMA aneurysm repair is <15%, with better results obtained via endovascular methods. The Society for Vascular Surgery guidelines therefore recommend treatment of all SMA aneurysms regardless of other risk factors [11-13].

Here, we present our successful diagnosis and treatment of a 41-year-old man with asymptomatic celiac trunk stenosis, in whom a large aneurysm of a branch of the SMA developed very quickly following conservative treatment of plastron appendicitis.

**Case Report**

A 41-year-old man was referred to the Department of General, Endocrine and Vascular Surgery of the Medical University of Warsaw due to rapid formation of an aneurysm of the artery originating from the SMA. The patient did not have any concomitant disorders and did not take any medications.

Two months earlier, he went to an outpatient clinic due to pain localized in the lower right abdomen, which began around navel and was aggravated by walking and coughing. He had mild fever (37.8°C) and nausea. In the abdominal US, the plastron appendicitis was diagnosed, and the patient was referred to a hospital (Figure 1). The US showed no other pathologies in the abdominopelvic cavity.

The patient was admitted to the General Surgery Department of one of the city hospitals, where a CT scan confirmed the plastron appendicitis. No other abnormalities were diagnosed. Conservative treatment with intravenous antibiotic therapy (cefuroxime, metronidazole, and gentamicin) was implemented, and after 2 weeks, complete resolution of symptoms was obtained. The good result of treatment was confirmed by the control abdominal CT scan. After 14 days of hospitalization, he was discharged from the hospital. In the abdominal CT scans and in the US examination, there were no pathologies of other organs of the abdominopelvic cavity diagnosed.

Three weeks after hospitalization, the patient, who was asymptomatic at that time, arranged an abdominal US examination in the outpatient clinic, in which a large 3.14×3.24×3.75 cm visceral aneurysm of the SMA was diagnosed (Figure 2).

The patient was referred to the Department of General, Endocrine and Vascular Surgery of the Medical University of Warsaw, where an angio-CT scan was performed, and he was found to have significant stenosis of the celiac trunk secondary to compression by the median arcuate ligament, as well as a 33-mm true visceral aneurysm of one of the branches of the SMA. The gastroduodenal arterial was tortuous, with small

![Figure 1. Plastron appendicitis. A thickened hypoechoic appendix measuring 1.16×1.59 cm, surrounded by a 3.68×3.92×5.7 cm plastron. (A) Transverse plane. (B) Longitudinal plane.](image-url)
aneurysms up to 13.2 mm in size. The reconstruction of the visceral circulation is presented on the Figure 3. Despite the presence of compression of the celiac trunk, the patient had never presented symptoms of Dunbar syndrome. The patient was qualified for early elective endovascular intervention. In the arteriography, a 33-mm true aneurysm was shown. It was localized on the branch of the SMA joining with gastroduodenal artery, which, due to the severe stenosis of the celiac trunk, was one of the major arterial blood supply pathways to the liver. Due to the high risk of liver malperfusion in case of potential complications, endovascular treatment was postponed, and the patient was qualified for the surgical decompression of the celiac trunk as the initial step of treatment.

Surgical decompression of the celiac trunk was performed. The peritoneal cavity was accessed with a subcostal bilateral incision. The celiac trunk was carefully dissected and its critical stenosis secondary to the compression by the median arcuate ligament was diagnosed. After cutting the median arcuate...
ligament, the celiac trunk was successfully decompressed with very good hemodynamic effect (Figure 4).

Four days after successful surgical decompression of celiac trunk, elective endovascular embolization of the SMA aneurysm and angioplasty of the celiac trunk were performed. From the right radial approach, an EverFlex 8×60 mm self-expandable peripheral stent (Medtronic, Minnesota, USA) was implanted to the celiac trunk. A hemodynamically significant deformation in the proximal part of the stent was visualized and the decision was made to implant a second stent. Subsequently, due to inability to introduce a guide wire antegrade via celiac trunk and the previously implanted stent, the decision was made to try a retrograde approach. From the retrograde approach, using left femoral artery access, a guide wire was introduced to the celiac trunk via the SMA and gastroduodenal artery and intercepted in the abdominal aorta. Postdilatation of the previously implanted stent was performed, then another stent, the ZILVER 518 8×60 mm self-expandable stent (Cook Medical, Indiana, USA), was implanted to the celiac trunk into a previously implanted stent (stent-in-stent), with good hemodynamic effect. In the next step of the procedure, the visceral aneurysm and its outflow branch were successfully embolized.

Figure 4. Decompressed celiac trunk (on the white vascular loop) and 2 large arteries branching from the celiac trunk: the common hepatic artery and the splenic artery. Aneurysm is not exposed.

Figure 5. (A) Arteriography made from SMA showing aneurysm and implanted stent in the celiac trunk: good patency and restored blood flow through celiac trunk with collateral artery connecting the 2 vessels. (B) Fully embolized aneurysm with patent visceral arterial bed.
with Ruby and POD Packing Coil systems (Penumbra, California, USA). Control arteriography showed complete exclusion of the aneurysm (Figure 5). The femoral access was closed using the FemoSeal vascular closure system (Terumo, Shibuya City, Tokyo, Japan) and the radial approach was closed with compression dressing.

A control angio-CT scan confirmed the technical success of the operation and complete exclusion of the aneurysm. The 96×62 mm postoperative hematoma surrounding the celiac trunk and reaching a lesser curvature of the stomach was diagnosed. The abdominal US showed significant decrease in size of the hematoma, which finally resolved spontaneously. The postoperative period was uneventful and the patient was released from the hospital on the 9th day after surgery. Following treatment, the patient remains under regular supervision of our department and, throughout a 1-year follow-up period is asymptomatic with good results of control US examinations.

Discussion

The visceral aneurysm coexisting with celiac artery stenosis was first described in 1973 by Sutton and Lawton [14]. In the same paper, the authors suggested that narrowing or the stenosis of the celiac artery may be a causative or predisposing factor for aneurysm formation due to increased blood flow through the collaterals [14].

Narrowing of the celiac trunk due to compression by the median arcuate ligament is a common finding in radiological examinations, with incidence of up to 5.1%. Features of external celiac compression are observed in 20% of the healthy population [15-20]. However, visceral arteries aneurysms are rather rare findings and their coexistence with median arcuate ligament syndrome (MALS) or stenoses of other visceral arteries is seldom found, suggesting the role of other factors in aneurysm formation, such as connective tissue diseases and inflammatory and genetic conditions [21,22]. The role ofatherosclerosis in visceral aneurysm formation seems to be marginal [6-8]. Our patient was a young, healthy, 41-year-old man without history of connective tissue disorders or genetic burden.

It was suspected that the aneurysm formed “de novo” in our patient after the episode of plastron appendicitis, that was treated conservatively with antibiotics. While making the diagnosis of plastron appendicitis, the patient had US examination in an outpatient clinic followed by a CT scan in the emergency room. These examinations, as well as the control CT scan before discharge of the patient from the hospital, did not show any other abnormalities in the abdominopelvic cavity. A few weeks after the episode of acute appendicitis, during the US examination, the aneurysm was first diagnosed. This suggests that abdominal cavity inflammation together with celiac trunk compression facilitated aneurysm formation, as supported by the literature [6-8].

Typical diagnosis of MALS is based on clinical symptoms combined with diagnosis of celiac trunk compression in imaging examination. Wide use of CT showed that asymptomatic compression of the celiac trunk by median arcuate ligament is seen in 3% of patients [23,24]. Hao et al showed that 87% of patients with celiac trunk compression remain asymptomatic [25].

Every aneurysm in the visceral circulation has a potential risk of rupture; therefore, in case of a newly diagnosed SMA aneurysm, treatment is advised [23]. In our patient the aneurysm was localized on the branch of the SMA, which, because of substantial narrowing of the celiac trunk, was the main source of arterial blood to the liver.

Although endovascular intervention is considered the first-line treatment in such pathologies, the initial arteriography showed the risk of liver malperfusion was high [11,26]. Therefore, to minimize the risk of liver ischemia, our patient initially underwent open surgical decompression of the celiac trunk.

The treatment plan was based on the previous experience of our unit. In 2018, a workgroup from our hospital proposed an algorithm for treatment of both ruptured and unruptured visceral aneurysms. According to this algorithm, certain issues determine the choice of treatment: the presence or absence of aneurysm-free collaterals joining celiac trunk and SMA, the presence or absence of other comorbidities favoring open repair, and the anatomy (Figure 6). An endovascular approach remains the first-line
treatment in unruptured aneurysms. In patients without collagenals joining the celiac trunk with the SMA, that are free from aneurysms, combined surgical decompression of the celiac trunk with subsequent endovascular aneurysm embolization should be performed as staged procedures [27]. In our patient, the main collateral was arising from the aneurysm; therefore, the risk of liver malperfusion in case of sole embolization was high.

Open surgical division of the median arcuate ligament was performed to preserve blood flow through the liver during aneurysm embolization. Staging of the procedures during combined treatment should be based upon personal experience. In our patient, staging was possible as the treatment was elective, not emergent; therefore, the endovascular procedure could be postponed according to the experience reflected in the algorithm developed in our hospital by a multidisciplinary team. In subsequent arteriography, there was still narrowing of the celiac trunk, so it was decided that stent placement was needed as the first step of endovascular treatment. In the second step of the procedure, the aneurysm was successfully embolized.

The preferred approach for small-artery aneurysms is endovascular treatment, with a wide range of methods to choose from: endovascular coiling, stent-assisted coiling, or embolization. The open surgical approach can be used as a rescue option in case of endovascular treatment failure [23]. In our opinion, treatment of patients with visceral arterial bed aneurysms should be tailored to each patient to ensure its safety and efficacy.

Conclusions

Visceral artery aneurysms are rare and can form very quickly. The treatment decision should be tailored each patient and their comorbidities. Endovascular treatment is the preferred choice in the vast majority of cases. In all patients with no connection between the celiac trunk and the SMA, with other unfavorable conditions or anatomy, combined or open surgical treatment should be considered.

Declaration of figures’ Authenticity

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References: