Case Report: Superior mesenteric artery syndrome-induced pancreatitis

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Abstract

Superior mesenteric artery syndrome-induced pancreatitis is rarely reported .We report a case which can be explained by an occlusive post-papillary syndrome, which produces retrograde reflux of bile into the pancreatic duct, activating inflammation responsible for pancreatitis.

Keywords: Weight loss, superior mesenteric artery syndrome, intestinal obstruction, pancreatitis

Introduction

Superior mesenteric artery syndrome (SMAS) is a rare condition resulting from vascular compression, in which, the third part of the duodenum is compressed between the aorta and the superior mesenteric artery [1]. In fact, many predisposing factors for SAMS, with a potential impact on the aortomesenteric angle, have been identified. SMAS can present acute manifestations like proximal small intestinal obstruction, or more commonly chronic manifestations, such as weight loss, vomiting, decreased appetite, and postprandial abdominal pain [2]. Conservative management, which consists in parenteral nutrition and nasogastric

tube decompression, could be effective. Nevertheless, SAMS can result in several serious complications due to the close anatomical relationships between D3, the main bile duct, and the Wirsung canal. In this report, we present a case of SMAS-induced pancreatitis.

Case report

In this report, a 20-year-old Caucasian female student was admitted to the hospital for abdominal pain and vomiting. The patient had a medical history of acute severe colitis that failed to respond to intensive first and second-line medical therapy. She underwent a surgical procedure consisting of subtotal colectomy with double stomia. Postoperative follow-up was complicated by an infectious pneumonia and diarrhea with a good response to antibiotics prescription. The patient was hospitalized one month after a surgery for epigastric pain associated with bilious vomiting along with 15 kg weight loss during the following three months. On examination, the patient was weakened with performance status (PS) =2 and body mass index (BMI) =15 kg/m^2. The abdomen showed tenderness on the epigastric and right upper quadrant area with no signs of peritoneal irritation. Stoma openings were functional. Initial blood investigations revealed a normal white count of 5,270 white blood cells (WBCs) per microliter. The hemoglobin level was 8.38 g/dl. The albumin level was slightly low at 34 g/L. The liver enzyme panel showed cytolysis predominant to aspartate aminotransferase (ASAT) (three times the normal range) and cholestasis with elevated gamma-glutamyl transferase (GGT) and peripheral blood lymphocytes (PAL) (13 and four times the normal range, respectively). C-reactive protein (CRP) levels were elevated at 60 mg/L. However, the rest of the biological examination workup, including lipasemia, was unremarkable. Subsequently, an abdominal computerized tomography (CT) scan showed an acute angle of 16° between the SMA and the aorta (normal range between 25 and 65°) with compression upon the anterior third part of the duodenum. Dilatation of proximal duodenum was noted with an SMA-aorta distance of 3.7 mm (Figure 1). Consistent with the diagnosis of SMAS, an upper endoscopy showed biliary gastric stasis.

The diagnosis of the SAMS was confirmed on the basis of the clinical presentation (significant weight loss preceding symptoms of upper bowel obstruction) and imaging. Nevertheless, the diagnosis of acute pancreatitis was also confirmed by the presence of acute epigastric pain and the CT scan indicated Balthazar D grade acute pancreatitis (Figure 2). Nevertheless, an etiological assessment of acute pancreatitis was negative: absence of alcohol abuse and drug imputability, while blood investigations didn’t show hypercalcemia or

hypertriglyceridemia. Ultrasonography and Bili magnetic resonance angiography (MRA) showed neither a biliary stone nor a pancreatic mass or malformations.

The patient was initially maintained on total parenteral nutrition and a continuous nasogastric aspiration tube was inserted for decompression. Then, the patient started a nutritional assistance program via a nasojujenal tube for enteral alimentation (Nutrison®, 1.0 Kcal/ml) starting with an intake of 10 kcal/kg/d, rehydration with crystalloid fluids (1000 ml of 0.9% saline) and an antiemetic medication (metoclopramide, 10 mg three times a day for five days). Then, 10 days later, the nasogastric tube was removed and the oral hypercaloric diet was started with good tolerance.
After 15 days of admission, the patient was asymptomatic with good tolerance of oral nutrition, gaining weight. Since medical treatment had good results, surgical intervention was not required, and the patient was discharged home. Three weeks later, the patient had gained three kg-weight. During two years of follow-up, she was asymptomatic with no recurrence of acute pancreatitis or upper bowel obstruction.

Discussion

SMAS was first described by Von Rokitansky in 1861 [3], as clinical symptoms resulting from vascular compression of the third part of the duodenum in the angle between the aorta and the superior mesenteric artery [4]. The eponym Wilkie's (Wilke's) syndrome or duodenal ileus arterio-mesenteric ileus was first used by Grauer in 1948 to honor Wilkie's accomplishment in being the first to provide a comprehensive description of this disease in 75 patients in 1927 [5, 6], while SMAS is a rare entity. Today, a PubMed (PubMed, RRID:SCR_004846) search (1950 to December 2021) using the medical subject headings (MeSH) term "superior mesenteric artery syndrome" showed that more than 710 articles had been published, including case reports, original articles, and reviews. In fact, many predisposing factors for SAMS with potential impact on the aortomesenteric angle have been identified and can be summarized into these categories:

- Severe weight loss leading to depletion of mesenteric and retroperitoneal fat and subsequent shortening of the aortomesenteric distance.

- Diseases of the spine, including orthopedic correction surgeries, use of corrective corsets ("cast syndrome"), congenital (short Treitz ligament) or acquired anatomical abnormalities (after surgical correction of aneurysms) [4, 7].

In this case report, the BMI of the patient was 15 kg/m^2 with 15 kg weight loss in three months in a postoperative and an acute severe colitis context.

The most frequent clinical findings include intermittent or postprandial abdominal pain (59–81%) whereas acute presentation is rarely described with a rapid evolving upper intestinal ileus followed by vomiting, nausea and anorexia resulting in an aggravation of weight loss [2]. In fact, a CT scan can calculate the angle between the AMS and the aorta, which is reduced between 7° to 22°, whereas it is normally between 38° and 65°. The aorto-mesenteric distance is also reduced and generally measures between 2–8 mm, while the normal distance is 10 to 28 mm [1]. A diagnosis of SAMS must be deduced with caution since it is generally confirmed after the presence of symptoms of SAMS associated with an aortomesenteric angle <22–25° and an aortomesenteric distance <8 mm [4] .

The treatment of SAMS in the last few years has led to a substantial shift towards medical treatment, thanks to advances in both enteral and parenteral nutrition. In fact, this treatment consists of setting up a nasogastric tube to cause decompression of the stomach and duodenum. It is advised to put the patient in the left lateral position, and above all compensate for hydro electrolyte disorders and establish an enteral hypercaloric diet by a nasojejunal tube. The success rate after medical treatment has been estimated to be around 72% but with a recurrence rate of 30% [8]. Then, surgical treatment, which consists mostly in the confection of a derivation by gastro-jejunostomy or duodeno-jejunostomy, is considered after medical treatment failure [4]. It is important to know that persisting obstruction in the third duodenum may lead to several complications due to the closeness between D3, the main bile duct, and the Wirsung canal: biliary reflux, gastritis and duodenal ulcers and pancreatitis.

In our case, the patient had acute pancreatitis with a negative etiological assessment. This complication of SAMS is rarely reported [7, 9, 10]. It is a mechanism that could be explained by bile-reflux into the pancreatic duct due to a secondary occlusive post-papillary syndrome. Refluxed bile may activate pancreatic enzymes, such as phospholipase A2, which may result in acute pancreatitis [7, 11, 12] (Figure 3).

On the other hand, our patient also presented with cholestasis, which could be explained within the framework of SAMS by two mechanisms. First of all, by a compression of the distal portion of the principal biliary duct, but imagery didn’t show dilatation of the latter. Secondly by a sphincter of Oddi dysfunction, the hypertension of this sphincter is responsible for its lack of relaxation, while bile outflow would lead to intrahepatic cholestasis. The second

hypothesis seems to be the reason for cholestasis in our case. The sequence of events in this case report are labeled in Figure 4.

Conclusions

SMAS is a multifaceted rare entity whose presentation can be varied due to the close anatomical relationships between D3, the main bile duct, and the Wirsung canal. Pancreatitis is a rarely reported complication of SAMS. It is mainly caused by occlusive postpapillary syndrome, causing retrograde reflux of bile into the pancreatic duct, which activates inflammation responsible for pancreatitis.

Data availability

Underlying data

All data underlying the results are available as part of the article and no additional source data are required.

Consent

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient/parent/guardian/relative of the patient.

Competing interests

No competing interests were disclosed.

Grant information

The authors declared that no grants were involved in supporting this work.

Figure legends

Figure 1: Abdominal CT images showing SAMS. Sagittal (A) and axial (B,C) abdominal CT scan in aortic phase show SMAS: Acute angulation of the superior mesenteric artery (12 °) (A), with reduced aortomesenteric distance (3.7 mm) (B) resulting in compression of the third portion of the duodenum, leading to dilatation of the proximal duodenum (arrow in C). CT, computerized tomography; SAMS, superior mesenteric artery syndrome.

- Figure 2: Abdominal CT images showing acute necrotizing pancreatitis. CT scan images in arterial phase showing the enhancement of the entire pancreatic parenchyma (white star) associated with heterogeneous peripancreatic collections (blue stars), fully encapsulated, containing area of fat (white arrow), fluid density, and areas with greater attenuation (blue arrow) compatible with walled-off necrosis. CT, computerized tomography.
- Figure 3: A labeled diagram illustrating the mechanism of SAMS-induced pancreatitis. SAMS, superior mesenteric artery syndrome.
- Figure 4: Schematic diagram of the sequence of events in this case report.

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