



Clinical Insights into Superior Mesenteric Artery Syndrome with Multiple Diseases: A Case Report

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Introduction

Superior mesenteric artery (SMA) syndrome is a rare disease caused by compression of the horizontal portion of the duodenum between the aorta and SMA. Clinical manifestations include recurrent episodes of postprandial nausea, vomiting, abdominal pain, and abdominal distention. Nutcracker syndrome (NCS) is a clinical syndrome caused by compression of the renal vein in the left branch of the inferior vena cava; it can be divided into pre-aortic compression and post-aortic compression based on the location of the compression. This symptom is primarily accompanied with hematuria, low back pain, pelvic pain, and gonadal varicose veins (varicocele or ovarian varices). Pancreatitis and gallbladder distention are common, including multi-related biliary stones and biliary tract abnormalities. Here, we describe a rare case of a combination of these multiple diseases.

Case Presentation

A 19-year-old female student, who was a member of the campus track and field team, suffered from nausea and vomiting after a month of an increased training load. The vomit was composed of stomach contents and bile, and the symptoms worsened after eating. The patient went to the local

hospital for treatment. Serological examination showed that her blood amylase was 238 U/L and urine amylase was 650 U/L. Abdominal CT did not show changes in the pancreas or pancreatic inflammation. (The patient was only able to provide a diagnostic report, not an image.) The first physician combined the clinical symptoms of the patient at the time; however, the diagnosis was unclear, taking into consideration that the pancreas might have enlarged. Fasting, acid suppression, and enzyme treatments were used, but the symptoms persisted. The patient experienced poor eating because of the vomiting and was only able to drink a small amount of liquid diet every day. In the following month, her weight dropped from 60 kg to 45.5 kg. The patient was unmarried, had no abdominal pain, and had no history of sexual activity. She had regular menstrual cycles; however, she reached a menopausal stage as the illness progressed; the last menstrual period she had was June 5, 2018.

Post-hospital examination showed that the patient was slender and tall (165 cm in height, 45.5 kg in weight, BMI = 16.71 kg/m²) with a mild depression in the abdomen; no abdominal tenderness or rebound tenderness was observed. Serological examination revealed the following: white blood cells: $5.15 \times 10^9/L$ (3.5–9.5); hemoglobin: 155 g/L (115–150); K⁺: 3.24 mmol/L (3.5–5.3); Na⁺: 135.2 mmol/L (137–147); Cl⁻: 96.8 mmol/L (99–110); amylase: 113 U/L (35–135); lipase: 128.6 U/L, (0–60); C-reactive protein (CRP) < 3 mg/L (0–3); hypersensitive CRP < 0.162 mg/L (0–3); urinary amylase: 934 (0–450); and three times urinary red blood cells: 42.2, 37.5, 38.3/uL (0–30.7). However, after 4 days, reexamination of urinary red blood cells showed negative results.

Based on the clinical manifestations and serological findings, we also suspected pancreatitis. Hence, the patient was asked to undergo Doppler ultrasound and spiral computed tomography. Abdominal Doppler ultrasound results revealed the following: pancreatic echo changes, gallbladder

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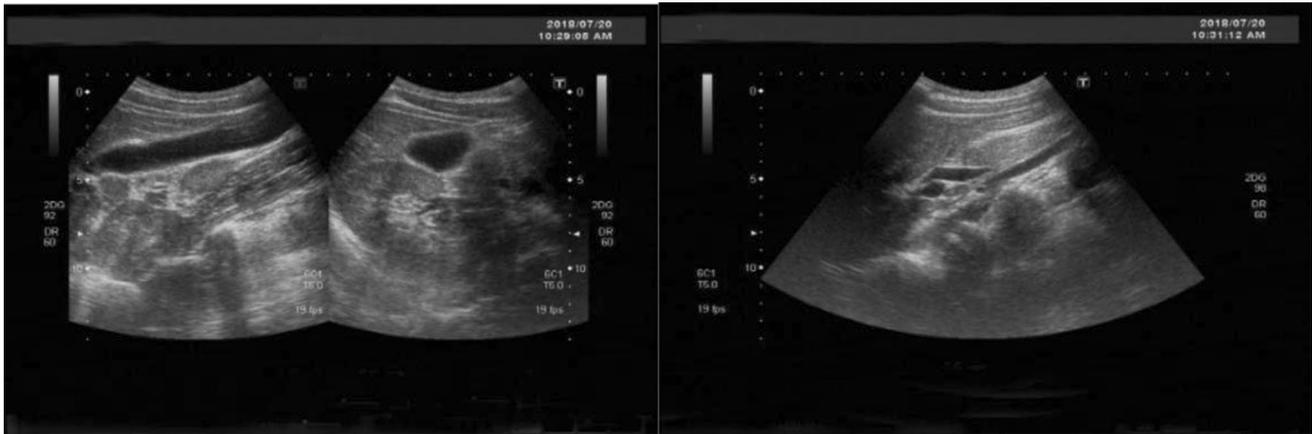


Fig. 1 Abdominal Doppler ultrasonography showing gallbladder distention and changes in the pancreas

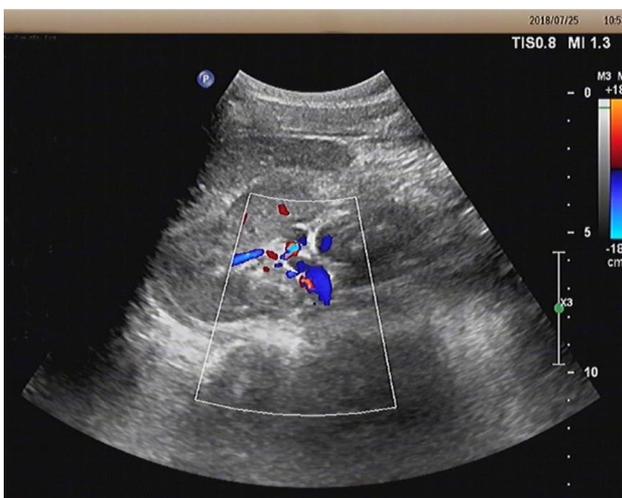


Fig. 2 Renal vein Doppler ultrasonography showing left renal vein stenosis and increased blood flow

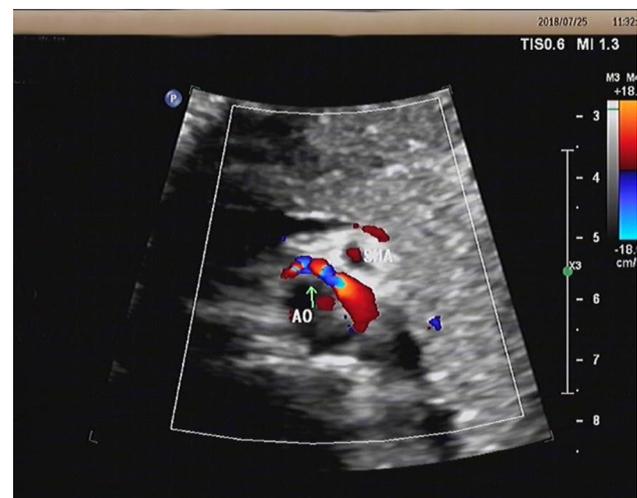


Fig. 3 Doppler ultrasonography showing a decreased angle between the aorta and the SMA

distention (approximate size $102 \times 22 \times 30$ mm), and the upper diameter of the common bile duct of 8 mm (Fig. 1). Renal vein Doppler ultrasound results revealed a diameter of 5.3 mm in the renal hilum of the left renal vein. The blood flow rate was 20.6 cm/s, the diameter of the renal vein at the angle of the SMA was 2.3 mm, and the mean blood flow rate was 76.7 cm/s. The internal diameter of the vein was 6.3 mm, and the average blood flow rate was 17 cm/s (Fig. 2). The SMA and the abdominal aorta were at an angle of approximately 17° (Fig. 3). Barium meal examination showed that the stomach was drooping and the food in the duodenum was stagnant. Spiral computed tomography showed that the pancreas was slightly swollen, had uniform density, and had no surrounding exudate; a small angle existed between the SMA and the abdominal aorta; and the left renal vein and duodenum were compressed (Fig. 4).

We diagnosed SMA syndrome, NCS, pancreatitis, and gallbladder distention and asked the urologist to discuss the case. Taking into account the patient's age and condition, we decided to intervene with nasal jejunal tube placement and enteral nutrition. Duodenal food accumulation was observed during catheterization. After the nasal jejunal tube was injected, the patient no longer vomited and was discharged from the hospital 3 days later. Follow-up revealed that she did not experience vomiting after her discharge. After 2 weeks, the patient's weight increased to 49.5 kg and BMI to 18.1 kg/m^2 . The patient pulled out the nasointestinal tube and exhibited no obvious discomfort. On August 28, 2018, her menstruation resumed. By October 10, 2018, the patient's weight had increased to 63 kg, but she refused to go to the hospital for evaluation.

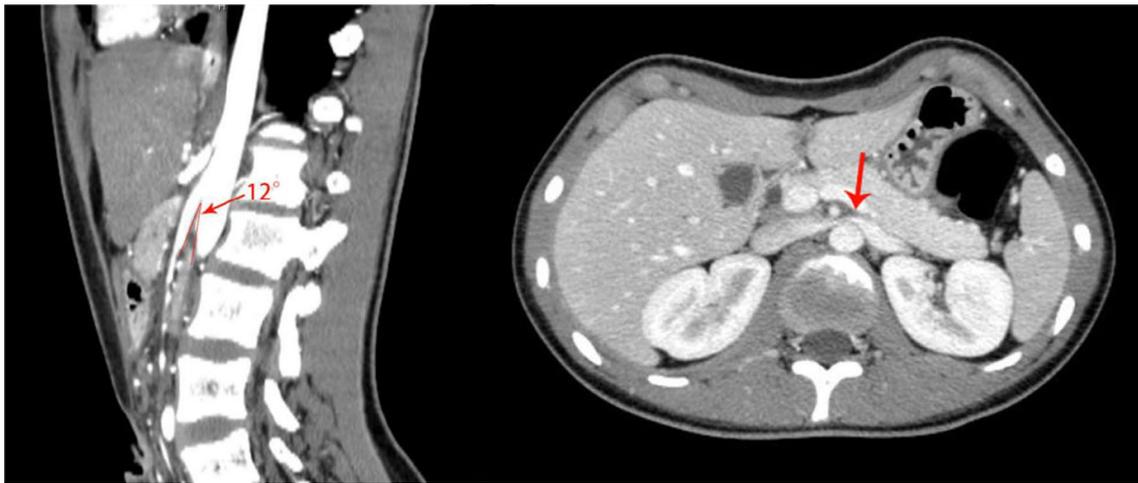


Fig. 4 Spiral computed tomography suggesting that the angle between the SMA and the abdominal aorta was reduced and the left renal vein and third duodenum were compressed

Discussion

This case is particularly unusual because the patient's clinical symptoms were not evident and were accompanied by SMA syndrome, NCS, pancreatitis, and gallbladder distention. Among these symptoms, SMA syndrome and NCS commonly cause a decreased angle between the SMA and the aorta. This case has pushed us to consider the following two problems.

What Is the Relationship Between the Several Diseases in This Case? Is It an Accidental Emergence or Is There a Causal Relationship?

The causes of SMA syndrome are diverse. The known causes are anatomical variation, mesenteric fat reduction, anatomical deformation caused by surgery or trauma, and consumptive disease or trauma, and the key point in its pathogenesis is the small angle between the SMA and the aorta [1]. In this case, the patient's mesenteric fat was reduced due to exercise load, thus causing SMA syndrome and vomiting. Given the poor treatment, the symptoms of vomiting persisted, and her poor nutritional status led to a continuous decline in weight, which, in turn, increased the vomiting. Given its low incidence, we reviewed recent case reports of SMA syndrome combined with NCS and SMA syndrome combined with pancreatitis; however, no report of SMA syndrome with gallbladder distention was found [2–4]. Given the horizontal pressure on the duodenum, a large amount of bile and digestive juices accumulate in the duodenum and stomach. Increased pressure in the digestive tract may cause bile reflux into the pancreatic duct and biliary tract; this phenomenon, in turn, may cause pancreatic inflammatory changes and gallbladder distention, thereby

providing a new possibility for the pathogenesis of some cases of noncalculous pancreatitis [5]. The left renal vein and duodenum are located between the SMA and the aorta. The combination of these two conditions has been rarely reported because the same cause leading to one of these two diseases requires further research [6]. In summary, we believe that the combination of the abovementioned diseases is not an accidental phenomenon, and gallbladder distention, NCS, and pancreatic inflammation may be important complications caused by SMA syndrome.

Does an Increase in Intestinal Pressure Cause Bile Reflux, and Is the Latter Associated with Pancreatitis and Gallbladder Distention?

SMA syndrome is caused by compression of a duodenal horizontal segment. The increased intestinal pressure after obstruction may affect duodenal papillary dysfunction, leading to bile reflux into the common bile duct and pancreatic duct. Whether or not bile reflux can cause pancreatitis and gallbladder distention (or cholecystitis) remains a controversial issue. In 1901, Opie [7] detailed the communication between the bile duct and the pancreatic duct at the duodenal papilla. This condition is a triggering event; therefore, bile can flow from the biliary tract into the pancreas and cause disease. This hypothesis is called the "common channel" theory. Armstrong and Taylor [8] reported that cholangiography in patients with a history of preoperative acute gallstone pancreatitis showed a high incidence of biliary and pancreatic reflux, whereas patients without a history of acute gallstone pancreatitis had a low incidence of biliary regurgitation. Howel and Bergh [9] found a positive correlation between biliary and pancreatic reflux and an elevated serum amylase after performing cholecystectomy

and cholangiography. They noted that infusion of bile into the pancreatic duct resulted in significant hyperamylasemia. Hyperamylasemia may be an important cause of the development of pancreatitis. Anderson et al. [10] found that the reflux of trypsin may initiate chronic inflammation of the gallbladder, which, in turn, may play a role in the formation of gallstones and, in some cases, the pathogenesis of acute cholecystitis. Although these studies are not absolute proof, we speculate that bile reflux plays a role in the pathogenesis of acute cholecystitis, pancreatitis, and other diseases.

Conclusion

Although SMA syndrome is a rare disease, it can be associated with other variable symptoms in addition to vomiting. These symptoms may be related to a series of other complications, especially when combined with NCS. We should be aware of these potentially serious diseases to ensure a timely diagnosis and optimal treatment timing.

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Compliance with ethical standards

Conflict of interest The authors have no conflicts of interest to declare, and they did not use any outside assistance in preparing the manuscript.

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