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OPEN ACCESS CASE REPORT

Rare Diagnostic Case of Superior Mesenteric Artery Syndrome: Case Report

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ABSTRACT

Wilkie's syndrome, another name for superior mesenteric artery (SMA) syndrome, is an uncommon cause of proximal small intestinal blockage. It happens when the angle between the abdominal aorta and the SMA narrows, causing compression of the third section of the duodenum between them, near their origin.

Here, we describe radiological findings of such a rare case of proximal small bowel obstruction in a young female patient who presented long time of repeated chronic symptoms of recurring post-eating epigastric pain and vomiting, with no symptomatic relief on medications.

An abdominal CT scan revealed distension of the duodenum and stomach, with narrowing of the third portion of the duodenum as it was compressed between the SMA and aorta. The reduced angle between the SMA and the abdominal aorta was indicative of SMA syndrome. These findings were confirmed following a duodenojejunostomy procedure.

Accurate diagnosis and reporting of SMA syndrome are crucial, as the non-specific clinical presentation of small bowel obstruction can principally lead to delayed detection, resulting in chronic symptoms, complications, repeated hospitalizations, and electrolyte imbalances.

Keywords: Case report, Computed Tomography, Wilkie's syndrome, Superior Mesenteric Artery syndrome, Jejunal-duodenal Anastomosis.

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CASE PRESENTATION

A female patient, age 20, arrived from our clinic to undergo an abdominal computed tomography (CT) scan. She has repeated chronic abdominal complaints. She reported having non-radiating postprandial epigastric discomfort, stomach fullness, and vomiting episodes for the last three years. She also had intermittent constipation during this period, which was managed conservatively. No symptomatic relief while taking medicine.

Her complaints were progressive in severity, with projectile vomiting, significant reduction in her diet intake, and weight loss. A body mass index (BMI) score of 15.2 kg/m^2; she had a slim build (IBW: ideal body weight of 49 kg) upon hospital admission.

The initial examination and basic biochemical tests were unremarkable; the ultrasound imaging did not reveal any abnormalities. An upper gastrointestinal endoscopy revealed gastritis and tested positive for Helicobacter pylori. The patient was managed conservatively with parenteral nutrition, but her symptoms were not completely relieved.

She was discharged from the hospital with a planned outpatient follow-up appointment with the gastroenterology specialist. To rule out other potential causes, such as gastric outlet obstruction, the specialist requested an abdominal CT scan.

Notably, the patient had no significant past medical history, including diabetes, infectious diseases (e.g., tuberculosis, brucellosis), or previous abdominal surgeries or burns.

Radiological image A contrast-enhanced CT scan of the abdomen and pelvis revealed narrowing of the third part of the duodenum as it crossed the midline between the superior mesenteric artery (SMA) and the aorta. This resulted in dilatation of the stomach and first and second parts of the duodenum (Figure 1). Reconstructed sagittal CT scan section (Figure 2) enabled precise visualization of vascular compression of the duodenum and measurement of the aortomesenteric angle and distance, which were found to be significantly reduced (23° and 5 mm, respectively) compared to normal values (38°-56° and 10-28 mm.

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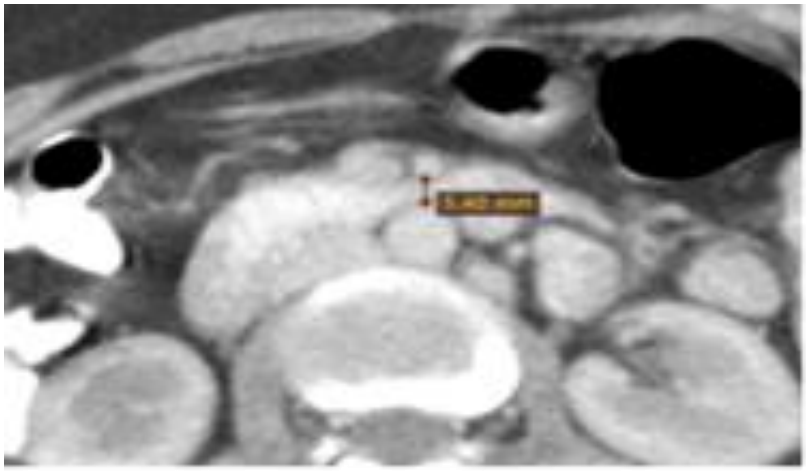

Figure (1): CT scan axial section of Superior Mesenteric Artery

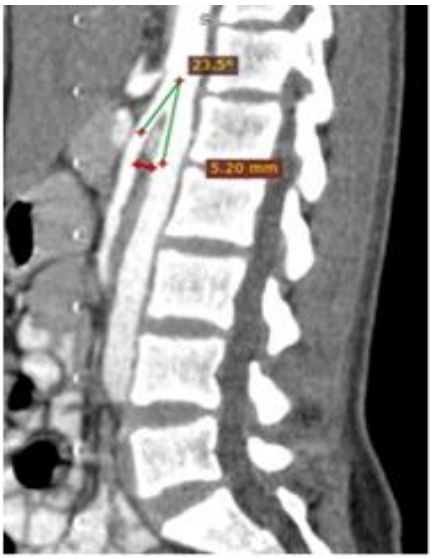

Figure (2): Sagittal reconstruction of Superior Mesenteric Artery

Ethical Approval

This case has been approved ethically by the Medical Ethics Committee at Faculty of Medicine and Health Sciences, University of Science and Technology, Aden, Yemen. Furthermore, informed consent was obtained from the patient.

DISCUSSION

SMA syndrome is a rare condition caused by blockade of the third portion of the duodenum owing to external squeezing between the aorta and SMA [1, 2]. Von Rokitansky [3] first demonstrated this entity in 1842. This condition was later extensively detailed in 1927 by Wilkie based on his study of 75 cases, earning it the alternative name "Wilkie's syndrome [4]. By 2022, over 730 articles had documented around 2,400 cases of SMA syndrome [5].

Timely recognition of SMA syndrome is crucial, as it can lead to metabolic imbalances and dehydration if left untreated [6], with a mortality rate reaching 33% [7]. According to radiographic studies, its prevalence in the general population ranges from 0.013% to 0.78% [8]. The condition is most commonly diagnosed at a median age of 17 years (typically between 13 and 18) [1]. SMA syndrome occurs more commonly in females, with an estimated female-to-male ratio of approximately 3:2 [5, 9].

Significant weight loss and the resulting reduction in mesenteric fat are key factors that contribute to SMA syndrome, as the condition is associated with a

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narrowing of the angle between the aorta and SMA [10].

Its causes might be acquired or congenital, including malabsorptive diseases, burns or medication-induced hypermetabolism, and cachexia associated with conditions like tuberculosis or cancer. Prolonged bed rest in a supine position also increases the risk of duodenal compression. Scoliosis and its treatments, such as surgery or casting, are well-recognized causes of SMA syndrome. Additionally, certain intestinal surgeries, including colectomy and ileal pouch-anal anastomosis, can exacerbate the condition by altering mesenteric tension and reducing the SMA-aortic angle (SMA-Ao). A congenital short or hypertrophic ligament of Treitz is a significant contributing factor to the condition in pediatric cases [5, 11].

Patients with SMA syndrome typically present with nonspecific symptoms, including biliousness, vomiting, weight loss, early repletion, upper abdominal pain, postprandial discomfort, and bloating. These symptoms can be similar to those of conditions like anorexia nervosa and functional dyspepsia, making diagnosis challenging [6, 11].

Severe duodenal obstruction during the acute period can cause severe symptoms and potentially fatal consequences, including stomach dilatation. Persistent nausea and vomiting during the chronic phase may cause inadequate food intake, which could result in significant weight loss and exacerbate the illness [6].

The diagnosis of SMA syndrome relies on clinical manifestations and a high level of suspicion, which is confirmed through radiological examinations [12]. Barium studies can exhibit a proximal obstruction, with or without gastric dilatation, and a delay in transit time that relieves when the patient is in a left lateral, prone, or knee-chest position [13]. While imaging modalities such as abdominal ultrasound, CT, magnetic resonance imaging (MRI), endoscopy, and endoscopic ultrasound can detect SMA compression, three-dimensional (3D) CT scanning is the gold standard because it provides accurate aortomesenteric angle and distance measurements and enables the detection of complications [5, 14]. Aortomesenteric distance and angle are typically between 10 and 28 mm and 38° and 56°, respectively [2]. By lowering the distance to less than 8 mm and narrowing the angle to less than 25°, the third part of the duodenum can be compressed. Additionally, pulsatile external duodenal compression may be seen during endoscopy [15].

Management

Initial conservative management includes stomach decompression, motility agents, parenteral nutrition, and lifestyle changes [16]. Surgical options for treatment include division of the ligament of Treitz, gastrojejunostomy, or duodenojejunostomy [16-18]. In the present case, duodenojejunostomy, considered the preferred surgical option with a 90% success rate [6], was carried out through an open laparotomy.

CONCLUSION

This case underscores the importance of prompt diagnosis and treatment of SMA syndrome, a rare but potentially life-threatening disorder. The patient presented with nonspecific symptoms, which made the diagnosis challenging. However, the use of advanced imaging modalities, including 3D-CT scans, enabled accurate diagnosis and guided surgical intervention.

This case's successful outcome emphasizes the need for a team effort that includes gastroenterologists, surgeons, and radiologists in the management of SMA syndrome. It also highlights the need for healthcare professionals to be more aware of SMA syndrome, especially in patients who have risk factors like scoliosis or significant weight loss.

Conflict of interest

The authors declare that no conflict of interest.

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