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Cast Syndrome: Breaking the Self-Perpetuating Cycle through Increased Medical Awareness

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ABSTRACT

Introduction: This review study intends to highlight that symptoms as common as nausea, vomiting, early satiety, and postprandial pain, could be caused by a potentially life-threatening easy to diagnose gastrointestinal disorder that could be overlooked if not known.

Materials and methods: A review of chosen literature in the PubMed, Google Scholar, Web of Science database was conducted, using the following keywords: "Cast syndrome", "Superior Mesenteric Artery Syndrome", "SMA Syndrome", "Wilkie's syndrome"

Summary: Cast syndrome or Superior Mesenteric Artery (SMA) Syndrome is a condition caused by the compression of the third portion of the duodenum between the superior mesenteric artery and the abdominal aorta. The syndrome is often associated with significant weight loss, anatomical predisposition, and certain medical conditions that reduce mesenteric fat.

Conclusions: Superior Mesenteric Aortic syndrome contributes to a vicious cycle that significantly affects patients suffering from it. Increasing medical awareness is essential to reduce the likelihood of diagnostic errors. Patients who are malnourished and frequently experience nausea and upper abdominal pain should undergo thorough diagnostic evaluation to prevent misdiagnosis and incorrect assumptions. Effective management of SMAS relies on physician engagement and proper education, with treatment often leading to a simple surgical intervention that boasts a high success rate.
Keywords: Cast syndrome, Superior Mesenteric Artery Syndrome, SMA Syndrome, Wilkie's syndrome

Introduction

Cast syndrome is an uncommon disorder characterized by a blockage of the third section of the duodenum due to the narrow angle formed between the superior mesenteric artery (SMA) and the abdominal aorta (AA); Von Rokitansky described this condition firstly in the middle of the 19th century. Subsequently in 1927, Sir David Percival Dalbreck Wilkie documented a series of 75 cases of this syndrome, providing a comprehensive analysis of its pathophysiology and diagnostic characteristics, thus SMA syndrome is also known as Wilkie's syndrome [1,4,13]. The syndrome is mainly linked to a reduced angle between aforementioned vessels, which can result from various congenital or acquired conditions. It is more commonly observed in females (ratio of females to males, 3:2) and young adults aged 18–35, although it can develop at any age. [14,18,39]. It can be inferred that this is a relatively uncommon digestive disorder among many internists, given its incidence of approximately 0.1–0.3% and a prevalence of less than 0.5% in the general population [1,2,25].

Pathophysiology

Superior mesenteric artery (SMA) syndrome develops when the angle between the SMA and the abdominal aorta (AA) becomes excessively narrow. The mesenteric adipose layer placed between the aorta and the superior mesenteric artery serves as a support for the SMA keeping it above the vertebral column thereby decreasing the duodenal compression. Under normal

conditions, the aortomesenteric angle varies between 38 and 65 degrees. However, when it decreases to less than 25 degrees and the distance reduces to below 10 mm, intestinal compression occurs[5,6,9]. The major cause of SMAS is body weight loss leading to diminution of aforementioned mesenteric fat tissue [4,14]. However the reduction in the distance between the vessels can also be secondary to medical procedures, including scoliosis treatment—whether surgical or with the use of plaster—hence the origin of another name for the syndrome- Cast syndrome [1,7].

It is important to highlight that a small SMA angle alone is not enough for diagnosis. There have been cases reporting of narrow SMA angles in individuals with low BMI, especially in children who show no symptoms of SMA syndrome. [10].

Burns, fasting, rapid adolescent growth spurts, eating disorders, impaired gastrointestinal motility, retroperitoneal tumors, stomach trauma or history of brain injury are examples of conditions which can increase susceptibility of Wilkie's syndrome.

Apart from acquired causes, we can identify congenital factors, including a short or high insertion of the Treitz ligament, spinal abnormalities (such as scoliosis or Marfan syndrome), hereditary predisposition, a low origin of the SMA, and malrotation of the SMA and superior mesenteric vein (SMV) [12,13,14].

SMAS may be associated with left vein stenosis, and when symptomatic (microhematuria, varicocele, left flank pain, and vascular thrombosis), the phenomenon is called the Nutcracker syndrome. The association of both syndromes is a common occurrence and should be remembered when the diagnosis is made to control renal functions [11,25,40].

Symptoms

Main symptoms of patients with SMA syndrome can mimic more common gastrointestinal diseases. Symptoms vary from nausea, vomiting, bloating, postprandial discomfort, epigastric pain to early satiety and weight loss [4,13]. There is tendency for pain and discomfort to worsen when lying on the back however in certain positions such as drawing the knees to the chest, tension on the small bowel mesentery decreases and provides relief [18]. Some patients refrain from eating to avoid discomfort and pain, unconsciously worsening their condition by further reducing fat tissue [31,33]. Despite the initial reason, reduction of fat index creates a vicious circle in which the symptoms intensify the cause, contributing to the exacerbating the condition [16-18]. Because of its similarity to other conditions, numerous case reports have documented instances of misdiagnosis—some patients were treated for different diseases for

years, undergoing multiple hospitalizations and, in some cases, developing life-threatening complications [1,17,18,25,32].

A delayed diagnosis may cause severe duodenal obstruction, resulting in intense symptoms and potentially life-threatening gastric dilation or perforation [10,16-18,44]. Recurrent vomiting might lead to electrolyte abnormalities, dehydration, aspiration pneumonia or respiratory depression via metabolic alkalosis which is why it should be diagnosed promptly before life threatening symptoms occur [8,10,19,20,43].

Diagnosis

The diagnosis of SMA syndrome has always been a challenge because it is made by a process of exclusion and there is no single specific symptom score or test available [4,8]. The differential diagnosis covers a wide line of conditions starting from anorexia nervosa, bulimia, diabetes mellitus, motility disorders, reflux disease, peptic ulcer disease, biliary colic, pancreatitis, mega duodenum mesenteric ischemia or obstructing tumor [21-23].

It is advised to suspect SMAS when a patient (especially woman) with a low body mass index presents chronic, refractory symptoms of the upper digestive system [14]. Verification of medical assessment requires a thorough patient examination combined with imaging studies confirming both duodenal obturation and a reduction in the SMA-AA angle and distance [32]. CT and/or MR angiography represent the gold standard in which diagnosis can be made by visualising simultaneously duodenal obturation and disturbed configuration between vessels [8,27]. The angle between the abdominal aorta (AA) and the superior mesenteric artery (SMA) typically ranges from 38° to 56°. However, in cases of SMA syndrome, this angle is reduced to approximately 6°–25°. Similarly, the normal aortomesenteric distance, which should be between 10–20 mm, decreases to around 2–8 mm. [28]. Gastroendoscopy, abdominal Doppler US, psychological assessment, laboratory tests can also be useful in differential diagnosis and patient condition evaluation [28,30,39].

Treatment

Treatment methods could be divided on conservative and surgical. Management in the acute setting includes fluid resuscitation, electrolyte correction, total parenteral nutrition, and nasogastric tube insertion for gastric decompression [8]. After establishing a patient condition usually as a first line treatment it is suggested to start the management with focus on weight regain - feeding with small portions of high caloric fluids by a nasojejunal or jejunostomy feeding tube is advised [4,8,13,23,31]. Some postural change methods such as prone position

and left lateral decubitus position, and maneuvers such as the knee-chest maneuver, and the Hayes maneuver (compression infraumbilically posteriorly and cephalad) may potentially alleviate epigastric pain [13,42]. Parenteral nutrition seem to be reasonable solution if aforementioned methods fail. If all conservative methods are ineffective, the patient then warrants surgical intervention.

Laparoscopic duodenojejunostomy is considered by many as the procedure of choice for causing minimal enteric bypass; and a number of case series reported high success rates (80%-100%) [35-38]. Since gastrojejunostomy can lead to greater nutritional loss, it appears reasonable to consider it a second-line treatment. Regarding Strong's procedure, which involves dividing the ligament of Treitz and rotating the duodenum to the right, some advocate for its use. However, it has been associated with a high failure rate, possibly due to persistent duodenal entrapment between the pancreaticoduodenal arteries [4,39].

Conclusions

Although numerous publications, case reports, and single-center studies exist on this syndrome, awareness among clinicians remains low. SMAS is a rare condition that can impact individuals of all ages and both genders. The majority of patients with this syndrome initially present to the primary care provider. Because the symptoms are nonspecific and there is no sensitive test, the diagnosis in most cases is delayed. Heightened knowledge among professionals is crucial to avert mistakes in diagnosis and treatment. Most patients need dietary consultation and education on lifestyle changes to prevent significant malnutrition. Unfortunately, conservative measures might not be enough, and some patients eventually require surgery. Further research is needed to improve early detection strategies and optimize therapeutic approaches to enhance patient outcomes.

Disclosure

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