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Superior Mesenteric Artery Syndrome, a Rare Complication of Severe Malnutrition: Two Cases from a Subsaharian Endocrinology Department

Nafy Ndiaye, Ngone Diaba Diack, Yakham Mohamed Leye, Sokhna Awa Balla Sall, Moussa Gueye, Mouhamed Fawaz, Mouhamadou Moustapha Ndong, Abdoulaye Leye

Department of Internal Medecine and Endocrinology, Teaching Hospital of Pikine, Dakar, Senegal
Email: fyfynami@yahoo.fr

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Abstract

Introduction: Superior mesenteric artery syndrome (SMAS), a rare diagnosis due to compression of the third duodenum between the superior mesenteric artery (SMA) and the aorta resulting in bowel obstruction, may lead to severe malnutrition. We report two cases of patients hospitalised in the Internal Medicine, Endocrinology, Diabetology, and Nutrition Department of the National Hospital Center (NHC) of Pikine. **Observations: Patient 1:** A 35-year-old female was referred for an aetiological diagnosis due to a rapid weight loss of 15 kilograms in one month, accompanied by persistent vomiting, following an appendectomy performed a month before admission. Upon clinical examination, she presented severe malnutrition (Buzby index of 76%), early post-prandial chronic vomiting, and a poor general condition. An abdominal CT scan revealed aortomesenteric clamp syndrome (AMCS) with an angulation between the aorta and the SMA of 13˚. The underlying cause in this patient was severe malnutrition. Fortunately, her condition improved with medical treatment. **Patient 2:** We report the case of a 30-year-old female hospitalized due to unusual weight-bearing post-prandial epigastric pain and intermittent vomiting over the past six months. Upon physical examination at admission, she exhibited severe malnutrition with a body mass index (BMI) of 14 kg/m^2, a Buzby index of 71%, trophic disorders, and a stage IV general condition assessment according to the World Health Organization (WHO). An abdominal CT scan revealed AMCS with an angle between the aorta and the SMA of 22˚ and an aortomesenteric space of 4 mm. The outcome was poor with medical treatment failure and, unfortunately, the patient died before surgery. **Conclusion:** SMAS is rarely evoked in clinical practice despite

the presence of contributing factors and suggestive clinical signs. The prognosis depends on management time.

Keywords

Superior Mesenteric Artery Syndrome, Malnutrition, Pikine

1. Introduction

Superior mesenteric artery syndrome (SMAS), also known as Wilkie's syndrome, results from intermittent extrinsic compression of the third part of the duodenum between the superior mesenteric artery (SMA) and the aorta [1]. It is a rare condition, with an incidence ranging from 0.1% to 0.3% and a mortality rate of 33% [1]. While it can occur at any age, it predominantly affects women and adolescents (aged 18 to 35 years) [2]. Clinical signs are often nonspecific, including sensations of gastric fullness and intermittent vomiting, leading to diagnostic delays [3]. The definitive diagnosis relies on abdominal CT scans, which measure the aortomesenteric distance (usually less than 8 mm) and angle (typically 22˚) with a specificity of 100% [4]. This syndrome has been described in patients falling into two broad categories; those who develop SMAS following surgery or due to compression and those associated with severe weight loss. [3]. Initial treatment is medical, involving gastric aspiration, correction of hydroelectrolytic disorders, and nutritional support; surgery is considered a second-line option [5]. This clinical entity is rarely reported in Senegal. Three previously reported cases were described, all in surgical settings [6] [7] [8]. Here, we present two cases of patients hospitalised in the Internal Medicine, Endocrinology, Diabetology, and Nutrition Department of the NHC of Pikine with SMAS secondary to severe malnutrition.

All patients treated in our university hospital centre were informed that their medical records may be used for scientific publications while respecting their anonymity. As this was an observational study, there were no ethical concerns.

2. Cases

2.1. Patient 1

A 35-year-old female was referred for an etiological diagnosis of a rapid weight loss of 15 kilograms in one month, associated with persistent vomiting, following an appendectomy performed one month before her admission. Upon clinical examination, she had severe malnutrition with a body mass index (BMI) of 14 kg/m^2 and a nutritional risk index (NRI) of 76%. She also experienced early post-prandial chronic vomiting and was classified as stage II according to the world health organization (WHO) general condition assessment. The diagnostic hypotheses considered were stenosing gastric ulcer, stenosing gastric neoplasia, and flange occlusion. The laboratory findings revealed microcytic hypochromic

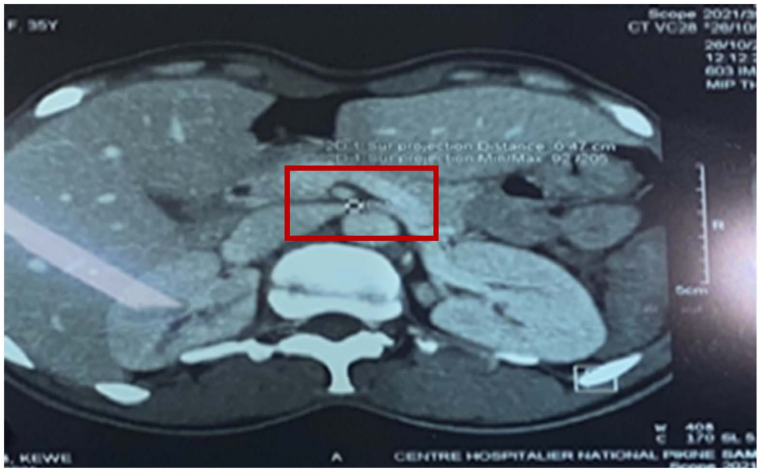

Figure 1. Abdominal CT: Cross-section showing reduction of the aorto-mesenteric distance in patient 1.

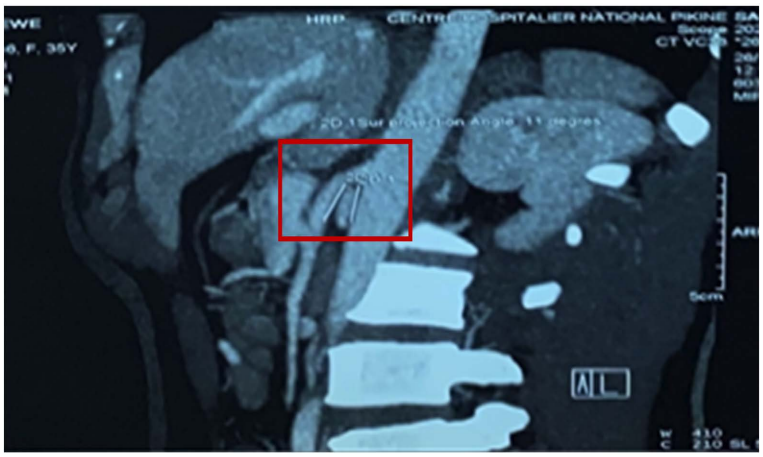

Figure 2. Abdominal CT: Sagittal section showing reduction of the aorto-mesenteric angle in patient 1.

anaemia, acute renal insufficiency with an estimated glomerular filtration rate of 34.95 ml/min, and hyponatraemia.

The abdominal CT scan revealed an SMAS with an angle of 13˚ between the aorta and the SMA and an aortomesenteric space of 4.7 mm. The underlying cause in our patient was severe malnutrition. Medical treatment was initiated, including correction of hydroelectrolytic disorders, a liquid diet with meal fractionation, and appropriate nutritional management. The patient's condition improved markedly, with cessation of vomiting, overall improvement, and normalisation of creatinine and sodium levels within one week.

Figure 1 and Figure 2 show the cross-sectional and sagittal views, respectively, of the aortomesenteric clamp in Patient 1.

2.2. Patient 2

A 30-year-old female was hospitalised for weight-bearing post-prandial epigastric pain, relieved by late post-prandial vomiting. There was no diarrhoea, con-

stipation, cessation of bowel movements, or fever. This presentation occurred against a backdrop of a significant weight loss of 25 kilograms in six months, accompanied by non-selective anorexia and loss of autonomy. Additionally, she was being monitored in the Neurology Department for axon-sensitive polyneuropathy.

On admission, the physical examination revealed:

- Severe malnutrition with a BMI of 14 kg/m^2 and an NRI of 71%.
- Trophic disorders: fine and brittle hairs, thin and fissured nails, angular cheilitis, and muscular atrophy.
- WHO stage IV general condition impairment.

Upon analysis, we observed normochromic normocytic anaemia with a haemoglobin level of 7.1 g/dL, without the involvement of other blood cell lines, a C-reactive protein (CRP) level of 2 mg/L, cholestatic syndrome with GGT within normal limits, hepatic cytolysis with ASAT elevated to three-fold the normal value, and normal renal function and blood ionogram. Total bilirubin was within the normal range and HIV and Hepatitis B and C virus serologies were negative. The fasting blood glucose level was 0.87 g/L.

Upper digestive endoscopy revealed congestive and candidal esophagitis, with no lesions observed in the gastric and duodenal mucosa.

The abdominal ultrasound outlined hepatic steatosis without signs of portal hypertension, while an abdominal CT scan highlighted SMAS, with an aortomesenteric angle of 22˚ and an aortomesenteric space of 4 mm. Additionally, there was homogeneous hepatomegaly in a steatotic liver with multiple uncomplicated gallstones.

Given this condition, a nasogastric tube was placed with meal fragmentation and an enriched diet for a week. In the absence of improvement, surgical management was decided; nevertheless, death occurred before the intervention. Table 1 summarises the epidemiological, diagnostic, therapeutic, and evolutionary data of our patients.

Table 1. Epidemiological, diagnostic, therapeutic, and evolutionary data of our patients

	Patient 1	Patient 2
Age (years)	35	30
Diagnostic timeframe (months)	1	6
Circumstances of discovery	Postprandial vomiting Severe malnutrition (NRI = 76%)	Weight-bearing epigastralgia Intermittent vomiting Severe malnutrition (NRI = 71%)
Diagnostic criteria	Aortomesenteric angle = 11° Aortomesenteric space = 4.7 mm	Aortomesenteric angle = 18˚ Aortomesenteric space = 4 mm
Evolution under treatment	Favourable under medical treatment	Medical treatment failure Death before surgery

3. Discussion

SMAS is a rare condition, with an incidence ranging between 0.1% and 0.3%. Racial or ethnic predispositions are yet to be identified. The two cases reported herein occurred in women in their thirties. The clinical manifestations of SMAS arise secondary to the duodenal obstruction, where the patient may present with early satiety and/or loss of appetite, epigastric pain, potentially severe post-prandial pain/fullness, nausea and vomiting that may be bilious, eructation, reflux, and "food fear" leading to malnutrition, weight loss, and poor weight gain [9]. Food intolerance promotes ongoing weight loss, which may further reduce the intra-abdominal adipose tissue and exacerbate the problem, resulting in a vicious cycle and deterioration of the clinical condition [3]. In its normal state, the duodenum is protected by fatty tissue at the level of the aortomesenteric space. The angle between the SMA and the aorta ranges from 25˚ to 60˚. A positive diagnosis of aorto mesenteric clamp syndrome (AMCS) relies on an abdominal CT scan, which measures the aortomesenteric distance and angle (which should be less than 8 mm and 22˚, respectively) with 100% specificity [3]. In patient 1, the aortomesenteric angle was 11˚ and the aortomesenteric space was 4.7 mm. In patient 2, the aortomesenteric angle was 18˚ and the aortomesenteric space 4 mm. Upper gastrointestinal endoscopy did not contribute to the diagnosis [10], however, endoscopic assessment can be useful to rule out further pathologies, such as duodenal tumours or peptic ulcer disease, which can also result in duodenal blockage [11].

Many acquired and congenital factors are involved in the aetiology of this syndrome. A low origin of the SMA or an abnormally high origin of the Treitz ligament is important congenital causes of SMAS. The loss of retroperitoneal fat due to, for example, polytrauma, burns, eating disorders, major surgical procedures, and cast immobilization, causing a reduction in the aortomesenteric angle, are the main acquired aetiological factors for SMA compression syndrome [12]. Severe malnutrition was present in both of our patients. The causal link between malnutrition and AMCS was clear in our first patient, who presented with malnutrition due to reduced food intake following an appendectomy. In the three cases previously reported in Senegal, SMAS was primary in a 16-month-old infant [6], secondary to scoliosis surgery in a 25-year-old patient [7] and to a gastric tumour in a 46-year-old patient [8]. In our second patient, the malnutrition could be of multifactorial origin, with lack of intake linked to the loss of autonomy secondary to her neuropathy, and of malabsorptive origin attributable to the cholestasis syndrome [1] [13]. However, hepatic steatosis and axonal polyneuropathy could be complications of this malnutrition [13] [14]. Axonal polyneuropathy could also be related to vasculitis or systemic disease [15], which are also sources of malnutrition and SMAS due to hypercatabolism [12]. Management is initially medical, consisting of inserting a nasogastric tube to decompress the stomach and duodenum, placing the patient in the left lateral position, and, above all, compensating for hydroelectrolytic disorders and introducing a

high-calorie diet. Increased retroperitoneal fat may be achieved with parenteral feeding. Prokinetics and antacids were among the drugs occasionally recommended [16]. Nutritional supplementation helps to gain weight and restore aortomesenteric adipose tissue, which displaces the SMA in front of the aorta, thus avoiding duodenal compression. Meals should be diluted to the consistency of puree or soup. The success rate of medical treatment is around 72%, with recurrences at around 30% [5]. Surgical treatment is recommended if medical treatment fails, involving either a gastrojejunostomy or a duodenojejunostomy bypass [16]. Medical treatment is deemed to have failed if there is no improvement in symptoms. According to reports, the average length of medical therapy is around 45 days. Given that medicinal therapy exceeding six weeks yielded worse results, surgical intervention becomes a viable option. Nonetheless, a successful treatment lasting 169 days has been documented in a child [5]. The mortality rate of SMAS is 33% [1]. In children and adults with a short history, there is a reasonable prospect of success; however, in the chronic adult patient, conservative treatment is often a prolonged in-hospital therapy with a low success rate [17]. Consistent with this observation, patient 1, presenting with an acute clinical course, was diagnosed earlier than patient 2, who was effectively diagnosed only after six months of evolution during her second hospitalisation. The first patient exhibited a good clinical evolution after the initiation of medical treatment, whereas medical treatment failed in our second patient and she died before surgery.

4. Conclusion

The defining symptoms of AMCS are variable and nonspecific; it can present as an acute or, more commonly, chronic condition. Diagnostic delay is often associated with a poor prognosis. A positive diagnosis relies on an abdominal CT scan, which measures the aortomesenteric distance and angle. Malnutrition is the most frequently reported aetiological factor and is also a consequence of AMCS.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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