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Conservative treatment in the superior mesenteric artery syndrome in children: Case report

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Abstract
Superior Mesenteric Artery Syndrome (SMAS) is an upper intestinal obstruction due to the compression of the third duodenum between the Superior Mesenteric Artery and the Aorta.
We report the case of a boy aged 14 years, with a BMI 12.49 kg / m², who presented for 10 days of postprandial bilious vomiting with persistent epigastric pain. Radiological assessment allowed the diagnosis of Superior Mesenteric Artery Syndrome with a 9° angle between the Aorta and the Superior Mesenteric Artery. Conservative treatment combining parenteral and enteral feeding came up with good results: we noted a gain in weight of 1 kg in 4 days, oral feeding was started and all symptoms disappeared. The patient was released after a 10-day hospitalization.
Superior Mesenteric Artery Syndrome usually presents in its acute form, consisting of symptoms of upper intestinal obstruction. Severe weight loss is usually involved. CT scan allows the measurement of the Aorto-Mesenteric angle confirming the diagnosis. Conservative management is the first line treatment, if it fails, one can resort to second-line surgery.

Keywords: superior mesenteric artery syndrome, occlusive syndrome in children

Introduction
Superior Mesenteric Artery Syndrome (SMAS), or Wilkie Syndrome, is a rare form of duodenal obstruction secondary to extrinsic compression of the third portion of the duodenum between the Superior Mesenteric Artery (SMA) and the Aorta. It was first described in 1861 by Rokitansky, and the first large series of cases in adults has been reported by Wilkie in 1921 [14].
Several factors are involved, facilitating the occurrence of this disease. When associated with signs of upper obstruction, SMAS diagnosis can be suggested.
We report a case of SMAS observed in a boy and discuss its various epidemiological, clinical, paraclinical and therapeutic aspects.

Patient and Observation
We report the case of a male patient, aged 14.
The patient got to the emergency room with post prandial bilious vomiting lasting for 10 days, associated with intermittent epigastric pain and a weight loss of 3 Kg. Further investigation showed no history of abdominal trauma or fever. Patient's father and uncle were treated for Behçet's disease
General examination revealed a cachectic patient with a weight of 34 kg for a height of 1.65 m, slight dehydration folds and oral aphthous. Abdominal examination revealed a soft abdomen with mild epigastric tenderness, there was no abdominal contracture or mass.
The rest of the examination was unremarkable, there was no genital ulceration.
Laboratory tests showed mild hyponatremia and the rest was normal.
Plain radiography and abdominal ultrasound revealed a gastric and intestinal stasis. An esophagogastroduodenoscopy was performed showing a significant gastric stasis with a delicate crossing of the D2-D3 junction, the lumen was narrowed but there was no apparent mucosal lesion. An injected abdominal CT-enteroclysis (Figure 1) and CT-scan with gastrografin ingestion (Figure 2), revealed a dilation of stomach, first and second part of duodenum, and a compression of the third part of duodenum between Superior Mesenteric Artery (SMA) anteriorly and Aorta posteriorly. The Aorto-Mesenteric angle was of 9° and the Aorto-Mesenteric space of 5 mm, confirming the diagnosis of Superior Mesenteric Artery Syndrome.

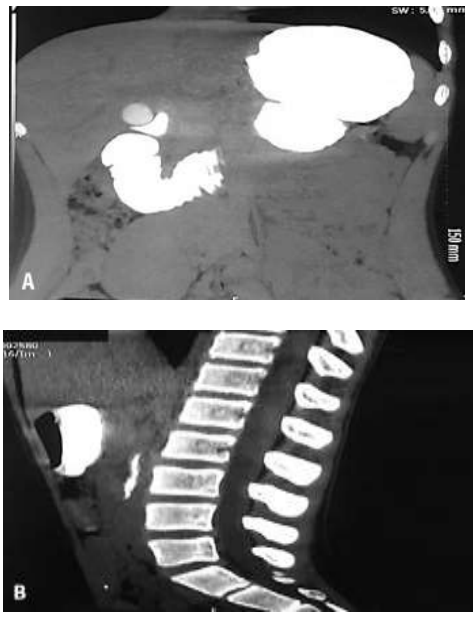

Fig 1: CT-Enteroclysis: Frontal view showing an obstructed PC D3 level (1-A), Sagittal view showing a narrowing of the third portion of duodenum (1-B)

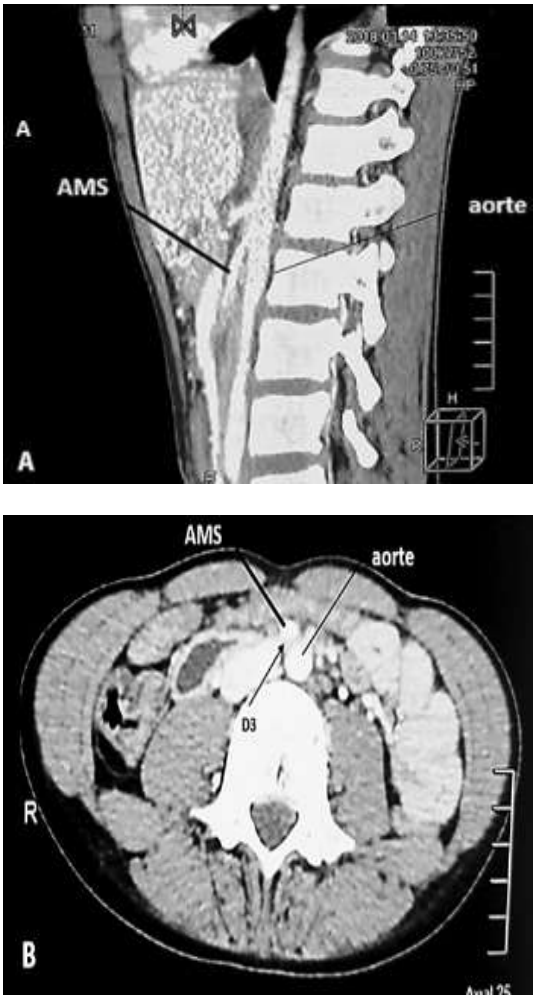

Fig 2: Abdominal CT scan: Sagittal view showing a narrowing of the Aorto-Mesenteric angle (2-A), transversal view through the third portion showing a duodenal D3 compression, SMA and Aorta (2-B)

A conservative approach was adopted combining parenteral - oliclinomel N4 (1000ml/day) - and enteral nutrition (2000 kcal/day) using a naso-jejunal tube placed by endoscopy beyond the obstacle. All associated with lifestyle and dietary measures: splitting meals and proper positioning of the patient in left lateral or post-prandial prone position.

In 4 days, the patient gained 1 kg. Vomiting and abdominal pain also disappeared. The naso-jejunal tube was removed and oral feeding along with lifestyle and dietary measures were initiated. After a 10-day hospitalization, the patient showed a complete resolution of symptoms and a weight gain of 3 kg in a total of 14 days.

Discussion

SMAS is a rare upper intestinal obstruction, with an incidence of less than 0.013-0.3% [1]. It generally occurs to young patients between 10 and 40 years with a female predominance.

Anatomically, the Superior Mesenteric Artery comes off the Aorta at an angle between 45 ° and 60 °. The duodenum is maintained by the ligament of Treitz and is surrounded by the retroperitoneal fat and lymph.

Reducing the Aorto-Mesenteric space to less than 8 mm or the Aorto-Mesenteric angle to less than 20 ° causes a duodenal compression and obstruction explaining the high ileus.

Some individuals are more prone to develop a SMAS than others. Anatomical and physiological factors (Table 1) have been incriminated, all causing rapid and severe weight loss leading to the reduction of the aorto-mesenteric angle. Some familial forms have also been described with a genetic component [2].

Our patient had a slender body, apart from that, no other factor was found.

Table 1: Main etiological factors of SMAS

Predisposing Factors
▪ A slender body ▪ A short or hypertrophy of the ligament of Treitz, ▪ A low origin of the Superior Mesenteric Artery ▪ A spinal lordosis
▪ Precipitation Factors
▪ Post-traumatic or post-surgery spinal deformities ▪ Anorexia ▪ Malabsorption diseases ▪ A hyper catabolic state due to burns, surgery, or an extended stay in an intensive care unit [3]

Clinically, SPAM may present in two forms: the acute form characterized by signs of upper intestinal obstruction; bilious vomiting and epigastric pain.

The chronic form usually presents with intermittent epigastric pain, nausea and vomiting often treated wrongly as gastroesophageal reflux.

Differential diagnosis includes anorexia nervosa, bulimia and other causes of duodenal obstruction due to peristalsis slowness [4] such as scleroderma, diabetes, dermatomyositis, systemic lupus erythematosus, amyloidosis, chronic intestinal pseudo-obstruction.

The diagnosis can be considered in any patient with a rapid and severe weight loss presenting signs of upper intestinal obstruction with no history of trauma or infection.

Plain abdominal radiography demonstrates gastric stasis. Esophagogastroduodenoscopy shows difficulty crossing the third duodenal portion with no apparent mucosal injury. Upper gastrointestinal series can make positive diagnosis showing dilation of the first and second portion of the duodenum with retention of the contrast product in a characteristic vertical linear line.

The diagnosis is confirmed by a contrast enhanced CT scan. It highlights duodenal dilation upstream of the obstruction and allows the measurement of the aorto-mesenteric space and angle.

First line treatment of SMAS in children should always be conservative. After correcting the different electrolyte disturbances, it consists on enteral feeding using a naso-jejunal tube passed distal to the obstruction under endoscopy. It allows the recovery of mesenteric fat and therefore the lifting of the occlusion. If enteral nutrition is impossible, we can resort to parenteral nutrition associated with nasogastric decompression. Proper positioning in left lateral or prone position has also been effective relieving the obstruction. In our patient, an association of entero-jejunal and parenteral nutrition was established with good results.

Medical treatment is not always effective, especially in infants Invalid source specified. If there is no improvement after three weeks of conservative treatment Invalid source specified, surgery may be considered. Various procedures have been described: Ladd procedure [6] which releases the third duodenal portion by dividing the ligament of Treitz and reorienting the small bowel on the right and the cecum and

colon on the left, also digestive bypass such as duodeno-jejunostomy or gastrojejunostomy, then Strong procedure [7] based on mobilizing D3 by dividing the ligament of Treitz and placing the duodenum to the right of the Superior Mesenteric Artery. Anterior Trans positioning of the third part of the duodenum anteriorly to the Superior Mesenteric vessels has also been described [8].

Evolution is often favorable with conservative treatment alone: Weight gain can restore mesenteric fat and thus relieve various symptoms.

Table 2: Summary of observations reported in literature on Mesenteric Artery Syndrome in children

Author	Age	S	Context	Diagnostic	Treatment	Evolution
Khemakhem [9]	14 yo	F	Primitive form	Abdominal CT Scan	Conservative	Exit at D10
Reported by Khemakhem [9]	11 yo	H	Primitive form	Ultrasound, UGI	Conservative	Discharge at D6
	14 yo	F	Anorexia nervosa	UGI	Conservative	Good
Loeb [10]	14 yo	F	Spinal fusion for scoliosis	UGI	Conservative	Oral feeding at D18
	16 yo	H	Collapsed vertebra following an AVP	UGI	Conservative	Oral feeding at D11
Galli [5]	5 yo	F	Weight loss	Ultrasound, abdominal CT	Conservative	Discharge D15
	16 yo	M	Emery-Dreyfus Myopathy	UGI	Ladd intervention	Discharge at D7
	14 yo	M	Morphotype rangy	Abdominal CT Scan	Conservative	Discharge at D8
Mbaye Fall [11]	16 months	M	Primitive form	Abdominal CT Scan	duodeno-jejunostomy	Discharge D11
Ali Coşkun [1]	13 yo	F	Anorexia nervosa	Abdominal CT Scan	Conservative	Oral feeding at D15
Mosalli [12]	7 days	M	high insertion of Ligament of Treitz	surgical exploration	Surgical intervention Strong	Discharge D11
Li Bing [2]	6 months	F	Vomiting postprandial	UGI, abdominal MRI	laparoscopic Ladd intervention	Discharge D5
	9 months	M	Vomiting postprandial	UGI, abdominal MRI	laparoscopic Ladd intervention	Discharge D4
	9 months	F	Vomiting postprandial	UGI, abdominal MRI	laparoscopic Ladd intervention	Discharge D4
Bernotavičius [13]	12 yo	F	Operated pure thoracolumbar scoliosis		Conservative	Discharge D24
Abdul Rehman [14]	15 yo	F	Anorexia nervosa	Abdominal CT Scan	Strong intervention	Discharge D5

Conclusion

Superior Mesenteric Artery Syndrome is a rare entity that should be evoked in front of a picture of duodenal obstruction.

Its diagnosis is based on abdominal CT scan with injection which allows measurement of the Aorto-Mesenteric angle.

Medical treatment should be initiated early and is usually sufficient. Surgery is proposed in secondary intention.

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