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Minimally Invasive Approach to Pediatric Superior Mesenteric Artery Syndrome (Wilkie´S Syndrome): A Case Report

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1. Abstract

Wilkie's Syndrome (WS) and Superior Mesenteric Artery Syndrome (SMAS) both refer to a rare cause of obstruction of second or third duodenum part due to its compression between the aorta and superior mesenteric artery. Presentation symptoms usually embracempostprandial crampy epigastric abdominal pain, vomiting, nausea, anorexia, and early satiety, sometimes mimicking pancreatitis, because of the features and location of pain. Characteristically, SMAS could appear after spinal surgery, psychological disorders, or rapid weight loss. Diagnosis is based on clinical features, but it frequently requires second level radiological studies. SMAS management may be accomplished through both medical or surgical approaches. We report the case of a 15-year-old boy presenting to the emergency department because of repeated episodes of biliary vomiting, always increasingly associated with bowel habits alteration. Anamnesis was characterized by massive weight loss and dorso-lumbar scoliosis. Once CT scan confirmed SMAS diagnosis, a primary conservative approach was chosen, with incomplete regression of symptoms. A mixed surgical technique encompassing laparoscopic dissection and trans-umbilical anastomosis was so reputed to be in the patient's best interest. Postoperative period was uneventful. No late complications were found after 15 months of follow-up. Mixed laparoscopic and trans-umbilical surgery resulted a safe and feasible approach to pediatric SMAS, providing both good aesthetic and clinical outcome.

2. Introduction

In 1927, Wilkie [2] described and named an obstruction of second or third duodenum part due to its compression between the aorta and superior mesenteric artery [1]. This condition is also known as Superior Mesenteric Artery Syndrome (SMAS). In 1957, Prouty and Waskow [3] firstly reported pediatric cases of SMAS [1]. Presentation symptoms usually embrace postprandial crampy epigastric abdominal pain, vomiting, nausea, anorexia, and early satiety [4-5]. Clinical features may mimic pancreatitis, because of the features and location of pain and the possible elevation of blood amylase [4]. Characteristically, SMAS could appear after spinal surgery, in patients afflicted by psychological disorders, or rapid weight loss [5]. Among known conditions which may result in the acute angulation between the aorta and the superior mesenteric artery are: (1) loss or significant depletion of retroperitoneal fatty tissues subsequent to weight losses; (2) ab-extrinsic compression by spica casts or belts; (3) congenital anomalies or anatomical defects as ligament of Treitz high insertion, scoliosis surgeries or intestinal malrotation [5-6]. According to Welsch et al. [6], patients presenting with a suggesting history and symptomatic court should undergo 2nd level radiographic studies with CT scan or MR angi-

ography to establish the diagnosis.

2.1. Radiographic Diagnostic Criteria Include

1. an aorto-mesenteric angle <20° (normal range 28–65°);
2. an aorto-mesenteric distance <8 mm (normal range 10–28 mm);
3. gastro-duodenal dilatation [6].

Wilkie's Syndrome management may be accomplished through both medical or surgical approaches. Usually, clinicians tend to start with conservative treatments as naso-gastric tube gastro-duodenal decompression and nutritional and electrolytes deficiencies correction [8-9]. In order to reach an early enteral nutrition resumption and to build up the lost retroperitoneal fatty pad, naso-jejunal tube may be positioned, allowing to bypass the compression site [8]. If enteral nutrition results insufficient or unaccomplishable, parenteral nutrition may be considered [6]. When medical treatment fails, surgery should be considered. The mainstay surgical principle is to bypass the site of obstruction through a proximal-distal anastomosis [8]. Several techniques had been described: duodeno-jejunostomy, gastro-jejunostomy, both with laparotomic or laparoscopic approaches [8, 10-11]. Other reported techniques encompass the Treitz ligament dissection with a compression release thanks to the mobilizations of distal duodenum parts [8,12]. In the SMAS-affected 15-years-old patient we are reporting, we decided to use a peculiar surgical technique which embrace both the advantages of laparoscopic and open approaches in order to resolve the anatomic anomaly with greater certainty and minimize recurrences risk. To the best of our knowledge, no witness of previous experiences is reported in literature regarding a mixed surgical approach encompassing laparoscopic dissection and trans- umbilical anastomosis, even though in paediatrics, more than one authors groups described mini-invasive surgeries for Wilkie's syndrome treatment [13, 14], including Roux-en-Y techniques [15], Ladd's procedures [16] and robot-assisted surgical approaches [8,17].

3. Case Report

In 2021, a 15-year-old boy presented to the emergency department for clinical disturbances for some weeks, characterized by repeated episodes of biliary vomiting, always increasingly associated with bowel habits alteration (complete constipation in the last three days before the visit). A massive weight loss and a previous diagnosis of dorso-lumbar scoliosis were declared during anamnestic interview. Physical examination highlighted extreme thinness and skin pallor, hypertympanic epigastric area without peritoneal irritation signs. Abdominal CT scan was performed, showing significant gastric distension with air-fluid levels, dilation of first and second duodenal portions. A < 20° reduction of the angle between the aorta and the superior mesenteric artery was described (Figure 1, 2), so the suspicion of Wilkie's Syndrome was confirmed. Conservative treatment with nutritional amendments including parenteral supplementation was carried out for a month, with incomplete improvement of clinical condition. When our patient reached an acceptable nutritional status without regression of sub-occlusive intestinal symptoms, surgical approach was reputed to be in the patient's best interest. We proceeded with a video-assisted surgical approach. In a supine position, a first umbilical incision was made to reach the peritoneum, then three laparoscopic accesses were placed in the lower half of the abdomen. A 30° scope was used. The dilated duodenum was visualized and freed from ligaments. In order to perform a Roux-en-Y duodeno-jejunostomy, a jejunum tract about 30 cm far from the Treitz was identified and exteriorized through the umbilical wound, previously dilated with Alexis© wound retractor (Applied Medical, California, USA). Duodenal-jejunal bypass surgery with Roux-en-Y loop were performed (Figure 3,4). Postoperative period was uneventful, resumption of enteral nutrition on the seventh post-operative day with regular weight gain. No complications were registered during 15 months of follow-up (Figure 5).

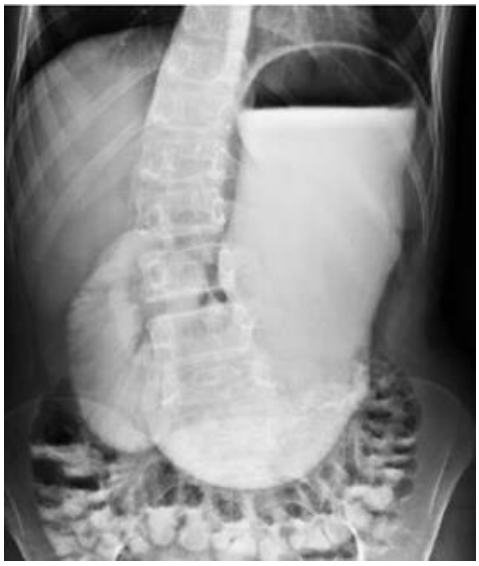

Figure 1: Barium study: Marked gastrectasia with air- fluids level and duodenal distension.

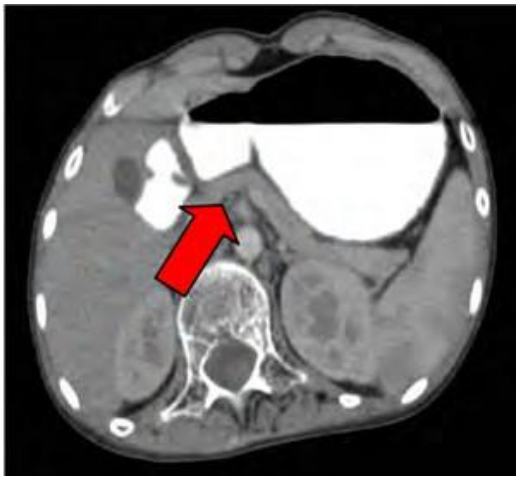

Figure 2: Abdominal CT: Presence of SMAS with aortomesenteric compass angle of 19°, minimum distance of 3mm between aorta and mesenteric artery. Compression of the III duodenal portion. Marked gastrectasia.

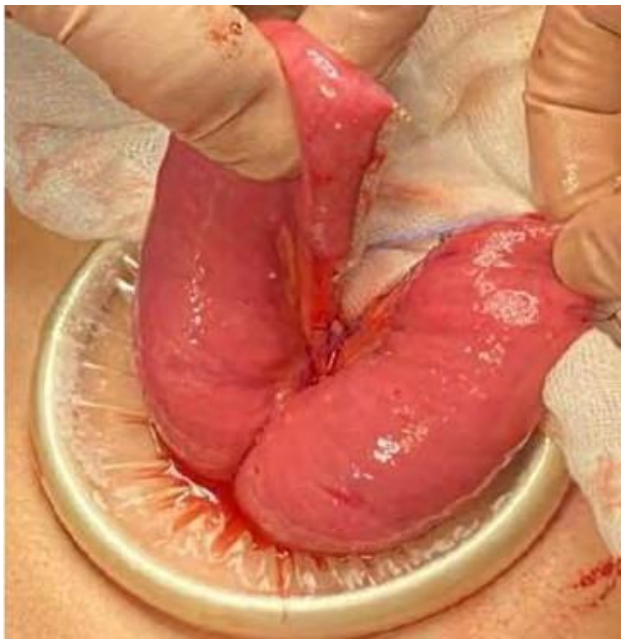

Figure 3: Surgical approach: Umbilical breach dilated with Alexis© wound retractor. The loop of jejunum is exteriorized.

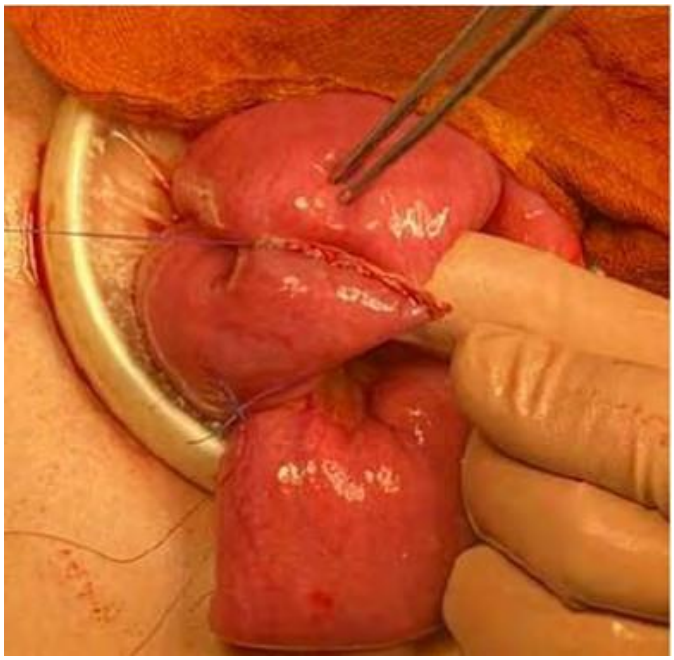

Figure 4: Duodenal-jejunal bypass surgery with Roux-style loop.

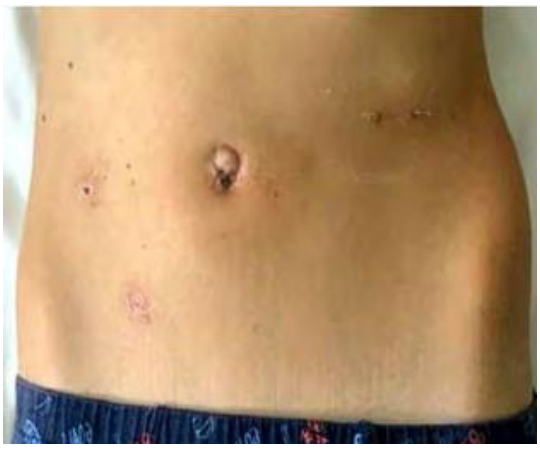

Figure 5: Post-operative aspect.

4. Discussion

Wilkie's syndrome is a rare condition leading to duodenal obstruction symptoms; its prevalence has been estimated between 0.013% and 0.3% amongst general population [18], but in children it remains uncertain [19]. Although different mini-invasive surgical procedures are nowadays well described [13-17], to our knowledge no previous literature does exist concerning mixed approaches encompassing laparoscopic section and manual anastomosis confection. The advantages of mini-invasive surgery are nowadays undeniable (e.g., shorter hospital stay, less painful post-operative period, smaller scars) and laparoscopic approach seem to be beneficial in most types of paediatric surgeries [20], and that is why we chose to employ it in the dissection steps of our procedure. In order to continue to grant minimum invasiveness and a minimum-scarred outcome while embracing the comfortability and the precision of an open-sewed anastomosis, we reputed adequate the bowel exteriorization through the Alexis©-dilated umbilicus, making anastomotic surgical gestures easier always maintaining thin the cutaneous breaches. No complication occurred neither during the seven-days long hospital stay nor in the 2021-2023 follow up. Aesthetic result was satisfactory for the adolescent.

5. Conclusion

Albeit the demonstration on a single patient and the necessity of further studies, mixed laparoscopic and trans-umbilical surgery results a safe and feasible approach to pediatric intestinal obstruction caused by Superior Mesenteric Artery Syndrome (SMAS or Wilkie´s syndrome), guaranteeing a good aesthetic and clinical outcome.

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