



Megabulbus in endoscopy; suspect for superior mesenteric artery syndrome in children

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ABSTRACT

Rarity of Superior Mesenteric Artery Syndrome (SMAS) and necessity of invasive tests to verify the diagnosis leads to patients receiving symptom-oriented drugs for a long period without any definite diagnosis. Diagnostic tests such as barium series, abdominal CT scan, abdominal angiography or magnetic resonance arteriography are used in patients with suspected SMAS. In pediatric patients, a non-invasive SMAS diagnosis may be considered easily with abdominal ultrasound performed by experienced hands. Megabulbus is used as a radiological term; however, reviewing the literature an endoscopic definition for megabulbus was not found. We decided to mention severely dilated pyloric ring and bulb as megabulbus. Megabulbus might be an indicator for SMAS. This is the first case of SMAS in adult and pediatric age groups presenting with megabulbus.

Keywords: Megabulbus, endoscopy, superior mesenteric artery syndrome

INTRODUCTION

Superior mesenteric artery syndrome (SMAS) is defined as compression of the third portion of the duodenum between aorta and proximal part of superior mesenteric artery (SMA) (1). Rokitsky published SMAS as an autopsy finding for the first time in 1842 (2). In 1927, Wilkie (3) reported a large series of 75 patients as Wilkie syndrome in surgical literature; however, the term "superior mesenteric artery syndrome" is widely used today. SMA is separated from aorta with approximately an average angle of 45° (38°-56°). Clinical SMAS manifestations appear if the angle drops below 20°. This angle measurement values are for adult patients; however, it is believed that values of this angle may be lower for pediatric patients (1-3).

CASE REPORT

A 14-year-old male patient admitted with postprandial epigastric pain, early satiety, nausea and vomiting. He had no important aspects in his past medical history; however a male sibling had been followed in our department with the diagnosis of intestinal pseudo-

obstruction for 3 years and died in an operation in another center. The parents were non-consanguineous. In physical examination; body weight: 39,9 kg (10-25 pr), height: 155 cm (25-50 pr) and all systems were normal. Grade A esophagitis and antral gastritis were detected in endoscopy. Histopathologic examination revealed HP (+) gastritis and esophagitis. Two years later, patient readmitted with abdominal pain, nausea and vomiting for the last one month. History revealed a loss of 11 kg of body weight in a month. In second endoscopy pangastritis and duodenogastric reflux were noted. Pyloric ring and bulb were severely dilated and bile mixed with food residues were found in the duodenum (Figure 1, 2). Barium series showed narrowing in the third portion of duodenum and no transition beyond the third portion of duodenum. Contrast-enhanced CT revealed small aortomesenteric angle (12.7°) and compression of the third portion of duodenum (Figure 3). The patient was diagnosed as SMAS and duodenojejunostomy operation was performed. During the 10-month follow-up period, the patient had no abdominal pain or vomiting.

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Received: September 12, 2012

Accepted: December 22, 2012

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DISCUSSION

The largest series in pediatric age group is 22 children diagnosed and followed up as SMAS reported by Biank et al (4). The vast majority of patients admitted had more than one symptom (64%). In this study, patients were diagnosed by gastrointestinal series 18 patients (82%), abdominal computed tomography 2 patients (9%) and laparotomy 2 patients (9%). The incidence of SMAS, which may be defined as a rare upper gastrointestinal obstruction is reported as 0,013% to 0,3% (5). Rarity of the disease and necessity of invasive tests to verify the diagnosis leads to patients receiving symptom-oriented drugs for a long period without any definite diagnosis. Diagnostic tests such as barium series, abdominal CT scan, abdominal angiography or magnetic resonance arteriography are used in patients with suspected SMAS.

The clinical severity of the disease varies depending on the degree of compression. However, the indispensable trio of the syndrome is abdominal pain, vomiting and weight loss. The first endoscopy of our patient was consistent with pancreatitis. Any pathological sign suggestive of gastrointestinal obstruction was not observed in endoscopy or medical history. In second endoscopy, megabulbus was noted, a hint for distal intestinal obstruction. During presurgical treatment in hospital, nutritional support was attempted but failed. We have two comments related with SMAS in our patient. First; SMAS might become obvious due to weight loss in an asymptomatic patient with congenital narrow aortomesenteric angle or secondly, angle narrowing due to decrease in mesenteric fat mass as a result of weight loss alone might reveal the clinical state. Reviewing the literature weight loss is mentioned; however, how much weight loss in how much time is not clear. In our patient the first comment may be more likely.

Megabulbus of the duodenum may be a part of generalized megaduodenum or may be a localized condition. Megabulbus is used as a radiological terminology. Golden described an "enormously dilated" bulb caused by an anomalous obstructing band for the first time, but did not give the actual dimensions (6). Taylor (7) presented 2 cases of megabulbus associated with annular pancreas. The size of a normal duodenal bulb is debatable. Meschan (8) indicated that it may vary up to 5 cm in length and 3 cm in width; whereas Fischer (9) suggested that any duodenal bulb more than 4 cm in diameter is a megaduodenum of significance. Therefore, if a duodenal bulb reaches 5 cm in diameter, the possibility of underlying cause has to be considered. If a large bulb is seen, the radiologist must search for evidence of the following lesions: ulceration in the postbulbar segment of the duodenum, annular pancreas, duodenal obstruction such as bands or lymphoma and duodenal atresia, abnormal peristalsis or atony of the loop.

In our patient, megabulbus was viewed in second endoscopy. English and Turkish sources were evaluated and we could not

find an endoscopic definition for megabulbus. We decided to mention severely dilated pyloric ring and bulb seen in the figure as megabulbus (Figure 1). Radiological measurement is easy; however, endoscopically measurement for the hollow organ would not be practical. Usage of megabulbus term is not encountered as an endoscopic appearance in the literature. Both adult and pediatric gastroenterologists might meet such a view during endoscopy. Megabulbus appearance has to remind the physician to suspect a distal obstruction and consider further examination.

As noted above, in cases of abnormal peristalsis or atony such as scleroderma, megabulbus or megaduodenum may be monitored. Regarding the sibling's diagnosis and the fact that intestinal pseudoobstruction is a motility disorder, we may consider that our patient's diagnosis may be intestinal pseudoobstruction. After a full year without any symptoms, clinical follow-up and time will guide us the presence or absence of such a disease at the background.

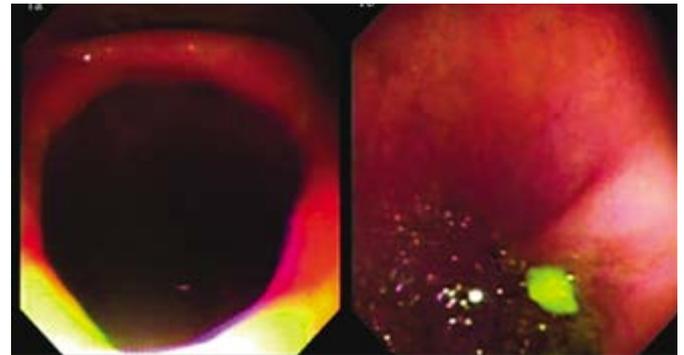


Figure 1. Large pyloric ring and bulb.

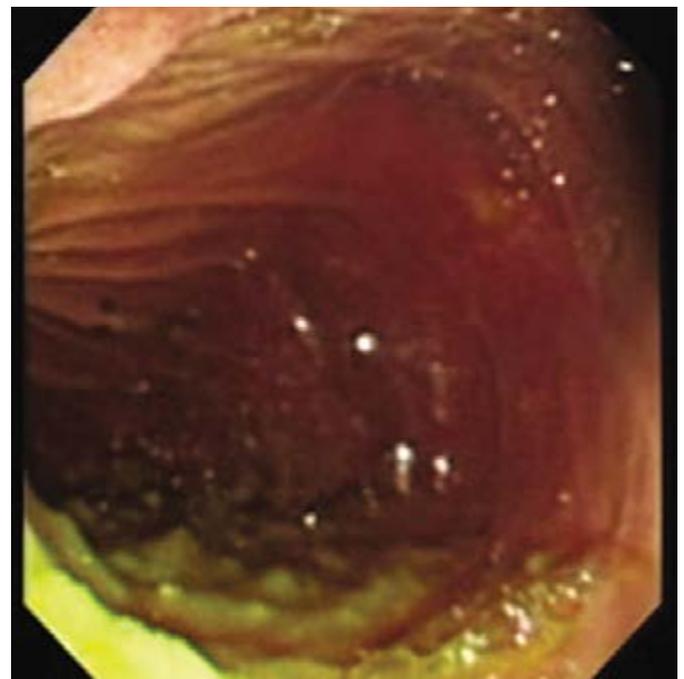


Figure 2. Food residues in duodenum.



Figure 3. Small aortomesenteric angle and compression of the third portion of duodenum.

As a result, SMAS is a rare disease and can be seen in pediatric age group. This is the first SMAS case in adult and pediatric age groups presenting with megabulbus. In the presence of highly enlarged pyloric ring and megabulbus during endoscopy, an intestinal obstruction such as SMAS has to be considered.

Ethics Committee Approval: Ethics committee approval was received for this study.

Informed Consent: Written informed consent was obtained from patient who participated in this study.

Peer-review: Externally peer-reviewed.

Author contributions: Concept - Ö.E.; Design - Ö.E.; Supervision - B.D.; Resource - Ö.E.; Materials - Ö.E.; Data Collection&/or Processing - Ö.E.; Analysis&/or Interpretation - Ö.E.; Literature Search - Ö.E.; Writing - Ö.E.; Critical Reviews - B.D.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

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