Minimally invasive duodenojejunostomy for superior mesenteric artery syndrome: a case series and review of the literature

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Abstract

Background Superior mesenteric artery syndrome (SMAS) is a disorder characterized by vascular compression of the duodenum leading to mechanical obstruction. Surgical intervention is indicated in patients who fail standard nonoperative management, in which duodenojejunostomy is favored based on previous small series. Given the rarity of the condition, knowledge of the optimal indications for surgery, risk of postoperative complications, and prognosis of SMAS after minimally invasive duodenojejunostomy is limited.

Methods A retrospective chart review was performed on patients who underwent minimally invasive duodenojejunostomy for SMAS from March 2005 to December 2013 at our "healthcare system". We analyzed patients' presentations, work-up, surgical therapy, and outcomes.

Results A series of 14 patients with SMAS underwent minimally invasive duodenojejunostomy. All of these patients met clinical criteria of SMAS with radiological confirmation. Average weight loss before surgery was 10.7 kg. Depression and eating disorders were comorbid in 6/14 patients. The mean age was 39 years (19–91 years). Twelve operations were completed laparoscopically and two were performed with robotic assistance. Mean operation duration was 119 min and average length of hospital stay was 5.5 days. There were no immediate postoperative complications. One patient developed a delayed anastomotic stricture that improved with single endoscopic balloon dilation. Initial symptom improvement occurred in all patients and the improvement occurred in 11 patients (79 %) during the follow-up. At a mean follow-up of 20 months, two patients experienced complications, including one infection at a simultaneously placed J-tube site and one patient with dumping syndrome. Mean weight gain was 3.8 kg ($p < 0.01$).

Conclusion SMAS should be considered a potential diagnosis in patients who present with a history of persistent postprandial vomiting, epigastric pain, and weight loss and confirmatory radiographic findings. In well-selected patients, minimally invasive duodenojejunostomy is a safe and effective treatment for SMAS with excellent short-term outcomes.

Superior mesenteric artery syndrome (SMAS) is an unusual cause of vomiting and weight loss, but with an estimated mortality rate of 33 % [1]. It is characterized by compression of the third portion of the duodenum due to narrowing of the space between the superior mesenteric artery and aorta, and is primarily attributed to loss of the intervening mesenteric fat pad. It has been referred to by a variety of other names including Cast syndrome, Wilkie syndrome, arteriomesenteric duodenal obstruction, and chronic duodenal ileus [2, 3].

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Rokitansky first described SMAS in an anatomy text in 1842 [4], and since this time, understanding of its pathogenesis has increased. However, because of its rarity and insidious nature, diagnosing this disease is often made by a process of exclusion. There have been numerous case reports of this condition; however, diagnosis is frequently delayed resulting in ineffective symptomatic therapies and inappropriate investigations [5, 6]. Once the diagnosis is made, treatment is either non-operative or surgical. Non-operative therapy consists of the establishment of enteral access distal to the obstruction with improved nutrition and weight gain. Weight gain is believed to increase the fat in the mesenteric fat pad overlying the third portion of the duodenum, thus relieving the obstruction. For patients who fail non-operative measures or who require a more durable therapy after the correction of electrolyte abnormalities and a period of re-feeding, surgery is indicated. Surgical options include lysis of the ligament of Treitz, gastrojejunostomy, and duodenojejunostomy. Of these, duodenojejunostomy has shown superior outcomes based on results from small series in the literature [7].

Previously, open duodenojejunostomy was considered the most successful surgical intervention, but recent advances in laparoscopic techniques have made laparoscopic duodenojejunostomy a safe and effective option [8]. Given the rarity of the condition, knowledge of the optimal indications for surgery, risk of postoperative complications, and prognosis of SMAS after minimally invasive duodenojejunostomy are limited. In this study, we described our experience and outcomes of a case series of 14 patients with SMAS. This series represents the largest series of patients with SMAS treated with laparoscopic duodenojejunostomy at a single healthcare system.

Methods

An institutional review board-approved retrospective chart review was performed on all patients with SMAS who had minimally invasive duodenojejunostomy from March 2005 to December 2013 in our healthcare system. For patients, an algorithm (Fig. 1) was used for preoperative evaluation and treatment decision. Image study is mandatory. Findings most consistent with SMAS are abrupt obstruction at the level of the SMA, dilated proximal duodenum, and recreation of symptoms during exams. Primary evaluation should be via gastroenterologists and psychologists with surgical evaluation for appropriate candidates. Other helpful studies can include CT scan of the abdomen which may demonstrate the same, with coronal reconstructions,

Fig. 1 Algorithm of preoperative evaluation
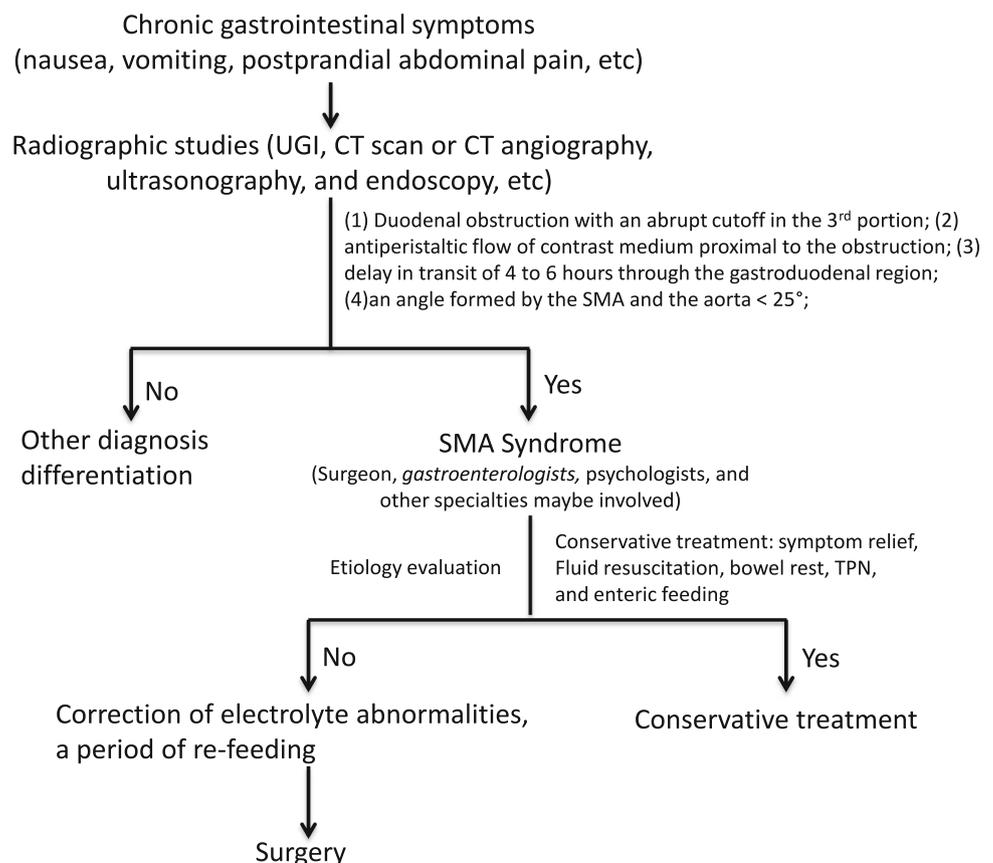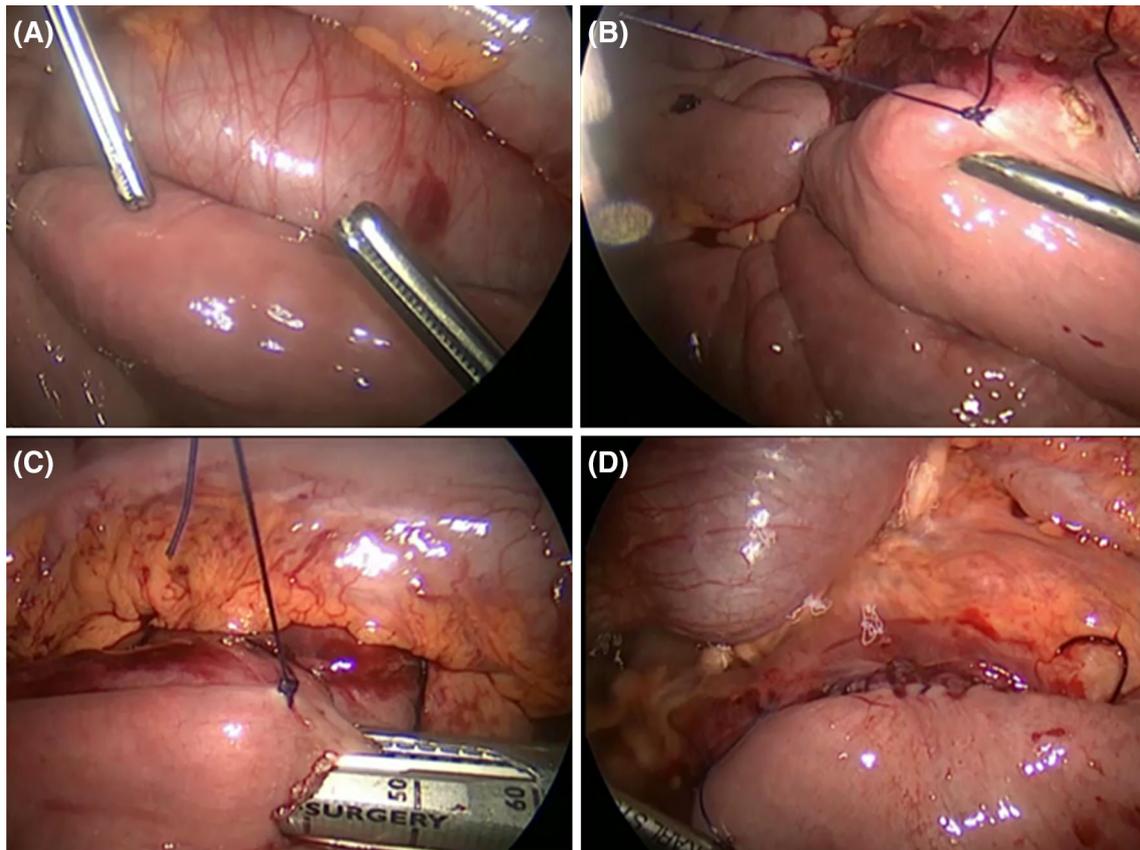
Fig. 2 A side-to-side duodenojejunostomy performed using a 60-mm linear stapler advanced to 40 mm when using a laparoscopic approach. **A** Laparoscopic view of duodenum and jejunum approximated prior to anastomosis, **B** Enterotomies made in jejunum and duodenum, **C** 60 mm linear stapler advanced to at least 40 mm mark, **D** Common enterotomy closed in running fashion

and CT angiogram, which further delineates the vascular anatomy and relationship to the duodenum. All patients in this study had clinical characteristics and confirmatory radiologic findings consistent with SMAS. Data collected from the charts included patient demographics, clinical presentation, operative data, and outcomes such as morbidity, mortality, and further therapies.

Surgical technique

The same technique was applied in this cohort. Entrance into the peritoneal cavity is typically obtained in the left periumbilical region with a 5-mm optical trocar. A 10-mm trocar is placed in the left upper quadrant for suture passage and a 5 mm in the left lower quadrant. Two 5 mm trocars are placed on the right side as well. Sequentially, the omentum and transverse mesocolon are elevated. The duodenum and jejunum are identified at the Ligament of Treitz. The peritoneum overlying the junction of the second and third portions of the duodenum is divided through the transverse mesocolon. The duodenum is gently mobilized off the retroperitoneum at this level. A segment of jejunum roughly 20 cm distal to the Ligament of Treitz is sutured to the previously dissected duodenum with a 2–0 absorbable stay suture. Immediately adjacent to this, a 2–0 non-absorbable suture is placed. Enterotomies are made in both the duodenum and jejunum in between the prior placed sutures. A side-to-side duodenojejunostomy is performed using a 60-mm linear stapler advanced to 40 mm when using a laparoscopic approach (Fig. 2). A fully hand-sewn technique was used during the robotic-assisted approach. The common enterotomy is then closed with the previously placed 2–0 absorbable suture and is tied to the tail of the non-absorbable suture. Proximal anti-obstructing stitch is placed. The anastomosis is then submerged in saline and an endoscopy performed to verify a patent and well-approximated anastomosis. The patient is admitted overnight with diet progressing from clear liquid to regular as tolerated.

Statistical analysis

Data are presented as mean $\pm$ standard deviation or as the range for continuous variables. Continuous variables were

Table 1 Patient demographics

Patient	Age	Gender	Comorbidities	Length of symptoms (months)	Diagnosis to surgery (months)	Length of F/U (months)
1	28	F	No clear reason (fibromylgia)	3	3	43
2	50	F	Multiple illness; scoliosis	6	1	31
3	19	F	Binge, fast	48	5	1
4	19	F	Anorexia	24	3	12
5	30	M	Psy/anxiety	24	1.5	13
6	27	F	Pancreatitis	12	1	1
7	37	F	Surgery/psy	36	7	10
8	25	M	Drug;/psy	18	1.5	12
9	74	F	Multiple surgery	11	7	10
10	52	F	Multiple surgery	17	10	40
11	50	F	Multiple surgery	36	2	14
12	91	F	Surgical; chronic	24	1	43
13	23	M	Anxiety; depression	36	1.5	43
14	20	F	Fetal alcohol syndrome; scoliosis	43	38	5
Mean	39	–	–	24	6	20

compared using unpaired Student's t test. Comparisons of categorical variables were performed using Fisher's exact two-tailed test. All tests were two tailed, and the results with a $p < 0.05$ were considered statistically significant.

Results

A total of 14 patients with SMAS (11 female/3 male) underwent minimally invasive duodenojejunostomy during the time period (Table 1). Mean age was 39 years (range 19–91 years). Comorbidities include psychosocial conditions (6/14), chronic disease (N = 2), and postoperative peritoneal adhesions (N = 5). One patient had no clear cause with only fibromyalgia identified in the past medical history.

Perioperative information is presented in Table 2. Presentation consistently involved persistent nausea (86 %), vomiting (79 %), epigastric pain (93 %), bloating (14 %), and esophageal reflux (21 %). The symptoms were chronic with the length of symptoms ranging from 3 to 48 months (mean 24 months) before surgery. The average period from confirmed diagnosis to surgery was 5.8 months (1–38 months). All patients had gastroenterologist review, and the diagnosis was confirmed with upper gastrointestinal series (UGI) and computed tomography (CT) scan (Fig. 3). Findings at UGI excluded intrinsic obstruction and CT scan included dilatation of the first and second parts of

Table 2 Perioperative information

Symptoms	Value
Nausea (%)	86 %
Vomiting (%)	79 %
Epigastric pain (%)	93 %
Bloating (%)	14 %
Reflux (%)	21 %
Weight loss (kg)	10.6 ± 8
Preoperative weight (kg)	56 ± 18
Postoperative weight (kg)	59.7 ± 17
BMI pre (kg/m^2)	19.9 ± 3.8
BMI F/U (kg/m^2)	21.3 ± 4
Operation duration (min)	119 ± 61
LOS (days)	5.5 ± 3.3
Morbidity (%)	14 % (2/14)
Mortality (%)	0 (0/14)

the duodenum, with or without gastric dilatation, duodenal obstruction with an abrupt cut off in the third portion and an aorta-mesenteric artery angle of <25°. After non-operative management in all patients, symptoms did not improve and subsequent surgical consultation was requested. All patients had weight loss from the development of symptoms to surgery ranging from 3 to 30 kg (mean 10.7 kg). Mean preoperative body mass index (BMI) was 19.9 kg/m^2 (range 14.5–28 kg/m^2).

Fig. 3 Upper gastrointestinal series (UGI) and computed tomography (CT) scan for diagnosis of all patients who had radiologist review
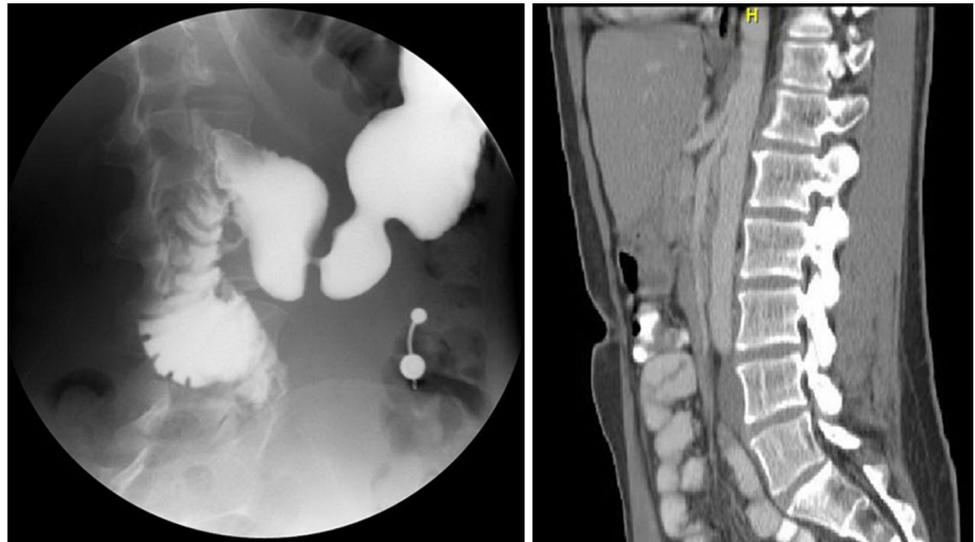
A minimally invasive approach was performed in all cases (12 laparoscopic and two with robotic assistance). Dilated stomach and duodenum associated with collapsed small bowel distal to the duodenum were observed in all operations. The mean operative time was 119 min (range 64–225), and the mean length of hospital stay was 5.5 days (range 1–14). All patients recovered uneventfully and no complications occurred during hospitalization. One robotically assisted patient developed a delayed anastomotic stricture that responded to single endoscopic balloon dilation. Follow-up endoscopy 6 months post dilation revealed a patent anastomosis. Two patients experienced complications (2/14), including one infection at a simultaneously placed J-tube site and one patient with dumping syndrome.

On follow-up, a non-standardized assessment of patients' symptoms (nausea, vomiting, pain, and bloating) was recorded by clinician interview with comparison to preoperative visits, baseline symptoms. All patients reported symptom improvement immediately after surgery. At a mean follow-up of 20 months, durable symptom improvement was achieved in 11 patients (79 %), while two patients still noted nausea and vomiting. One patient had less severe nausea and vomiting than before, but epigastric pain was still persistent. These three patients all had preoperative psychological conditions (anorexia, bulimia, anxiety, and depression). Postoperative imaging and endoscopy showed good emptying of the stomach and duodenum and no evidence of further obstruction. The mean weight gain of this group was 3.8 kg (0–10.7 kg, $p < 0.01$) at the last visit. Mean BMI gain of all patients was 1.38 kg/m^2 (range 0–4.2 kg/m^2, $p = 0.37$).

Nutritional status, characterized by need for supplemental nutrition (enteral or parenteral), was examined in all patients

Table 3 Nutritional status

Nutritional route	Preoperative	Use at F/U	p value
Enteral	1	1	0.6
Enteral and parenteral	1	–	–
Parenteral	1	–	–

(Table 3). Patients should be considered for supplemental nutrition if they demonstrate severe nutritional depletion, manifested as profound weight loss, greater than 10 % TBW, low acute and long-term nutritional lab parameters (albumin, pre-albumin, and transferrin). Before surgery, the use of enteral nutrition was required in three patients. At the time of surgery, the enteral access was given for severe nutritional depletion; manifest as profound weight loss, greater than 10 % total body weight; low acute and long-term nutritional lab parameters (albumin, pre-albumin, and transferrin). After surgery, one patient remained on enteral nutrition at last follow-up because of cyclical vomiting syndrome.

Discussion

In 1842, Rokitansky first described the SMAS in an anatomy text [4]. It involves the entrapment and obstruction of the third part of the duodenum between the SMA and the aorta. In 1927, Wilkie published the largest study on SMAS based on 75 cases in which he concluded that duodenojejunostomy was the treatment of choice [9]. In recent years, there have been numerous case reports of this condition. However, because of its rarity, the majority of reports of this condition are limited to very small series. Diagnosis of this condition is frequently delayed, relies on

a high index of suspicion, and is often made by a process of exclusion, resulting in ineffective symptomatic therapies and inappropriate investigations [10].

A total of 400 cases have been reported in the English literature with a slight young female (18–35 years) preponderance [11, 12], though it can occur at any age and in males. In our series, 79 % of subjects were female. The mean age was 39 years and ranged from 19 to 91 years old. Among these patients, 36 % (5/14) patients were older than 50 years. The oldest patient was 91 years at the time of diagnosis, which is also the oldest patient currently reported in the literature.

Any conditions that cause a decrease in the aortomesenteric angle can result in SMAS. This syndrome ultimately falls into two broad categories: those who develop symptoms following surgery and those associated with severe weight loss. The second category includes wasting conditions such as chronic diseases, malabsorption, cancer, AIDS, cerebral palsy, and burns. Another group includes those with psychological conditions and eating disorders such as anorexia nervosa and drug abuse. There is a significant psychological overlap in many patients who present with this condition [13]. About half of the patients in our study had a history of psychiatric conditions and eating disorders alone or combined with other causes. In fact, all three patients in our series with persistent long-term symptoms had a history of psychological and eating disorders. Therefore, the psychological management of these patients must be considered to ensure optimal long-term outcomes after the surgical correction of the physical obstruction. Also, the presence of psychological conditions may predict poor outcome with duodenojejunostomy for SMAS.

In adult patients, non-operative therapy is often a prolonged in-hospital approach with a low success rate with variation from 14 [14] to 71 % [15]. In one previous case series, patients had extreme weight loss from the development of symptoms ranging from 20 to 40 kg (mean 29 kg), with the weight loss ranging from 33 to 55 % of their body weight before surgery [16]. In our study, the average weight loss prior to surgery was about 10 kg (about 20 % of body weight). One possible reason we did not see such drastic weight loss may be due to a high index of suspicion in these patients with increased use of diagnostic imaging to confirm the diagnosis. Another reason is that, as a large tertiary referral center, our reviewed charts may not include all medical history of those patients who were referred for surgical management. In these cases, after correction of electrolyte abnormalities and a period of feeding, surgery was performed. In this study, the window between diagnosis and surgery was 5.8 months. Previous case reports and reviews reported a much higher interval between diagnosis and surgical therapy. Currently, there are no standard guidelines regarding indications for surgery in this rare syndrome. It is recognized that significant preoperative weight loss is associated with an increased risk of postoperative complications. Furthermore, regardless of the etiology, active disease may result in a self-perpetuating cycle of vomiting leading to further weight loss and vomiting. For these reasons, our healthcare system has adopted a more aggressive approach in terms of surgical management during the early phase of a confirmed diagnosis.

Several surgical options have been described in the literature including Strong's procedure (mobilization of the duodenum by division of the ligament of Trietz) [17], gastrojejunostomy, and duodenojejunostomy. Strong's procedure has the advantage of maintaining bowel integrity, and has a success rate of 75 % [18]. Laparoscopic techniques of Strong's procedure have shown a similar success rate when compared with open laparotomy [19]. Gastrojejunostomy has been associated with complications related to failure to relieve duodenal obstruction, blind loop syndrome, and peptic ulceration [4]. Because of these success rates and potential complications, both of these procedures have been considered inferior to duodenojejunostomy [18]. Munene et al. [20] reported that the success rate of duodenojejunostomy is 100 percent based on their own one case report and previous 13 cases reported in the literature. Despite the high success rate, duodenojejunostomy does potential risk of blind loop syndrome [16]. To avoid this pitfall, a Roux-en-Y duodenojejunal bypass for SMAS has been described using the laparoscopic approach [21]. The Roux-en-Y duodenojejunal bypass left no blind loop, with free drainage of not only the duodenum proximal but also distal to the compression site at the SMA [22]. In our current study, no perioperative complications were identified. One delayed anastomotic stricture was found in our series, which responded to endoscopic balloon dilation. Interestingly, this patient had a robotically assisted operation with a completely hand-sewn anastomosis. The anastomosis was performed in a two-layer fashion with non-absorbable suture as the outer layer. One potential explanation for this stricture includes a non-calibrated anastomosis that resulted in narrowing of the anastomotic lumen following completion of sewing. All other cases used a standard 40 mm advancement of the linear stapler, which resulted in a uniform 40-mm anastomosis. This poor outcome has led us to abandon the robotic approach, especially given the superior results using a purely laparoscopic technique. The outcome also highlights the potential success of early endoscopic dilation when a stricture is identified.

All patients in this series reported improvement in symptoms immediately after surgery. Two patients complained of occasional nausea and vomiting and one had epigastric pain at the most recent office visit. All patients experienced weight gain, and mean BMI increased from

19.9 to 21.3 postoperatively. One of the largest reported series included 16 patients who underwent open duodenojejunostomy. These patients were followed for 7 years after surgery [23] and all experienced weight gain. However, symptoms were essentially unchanged with the exception of vomiting, which was significantly decreased. In our series, we observed improvement both in weight and symptoms. The predisposing medical and psychiatric conditions may correlate with the outcomes and in some cases, and symptoms may not resolve completely following the correction of abnormal anatomic findings [24]. In the follow-up, three patients still have symptoms, despite normal imaging. The possible persistence of symptoms after surgical bypass can be traced to the remaining prominence of reversed peristalsis in contrast to direct peristalsis, although the precipitating factor (the duodenal compression) has been bypassed or relieved.

A laparoscopic approach is certainly feasible and this advance has been reported by several centers [18, 25]. The advantages of a laparoscopic procedure include shorter operative times, improved optics, rapid recovery, and minimal need for postoperative analgesia. Both Strong's procedure and loop duodenojejunostomy have been performed using the laparoscopic approach with shorter recovery time and hospital stay when compared with conventional approaches by laparotomy [26, 27]. Compared with the most recent and largest series of open duodenojejunostomy done between the years of 2002 and 2007 where the reported LOS was 10 days (7–14 days) [16], the average length of hospital stay in our study is 5.5 days (1–14 days). With the low incidence of postoperative complications, the benefit of a minimally invasive approach is clear.

Study limitations

Because SMAS is an unusual symptom, the sample size may limit the power of the study result. Other limitations include the retrospective nature, in which the information collection is based on the description in the medical chart, and the symptom improvement was documented via outpatient follow-up and documentation, instead of a symptom severity score. Furthermore, as a large tertiary referral center, we do not have a total number of patients who were seen for the disease and those who were treated medically.

Conclusions

In this largest series of 14 patients with SMAS, we have shown that a minimally invasive duodenojejunostomy is a feasible, safe, and effective therapy in patients who have failed a non-operative approach. In our review, we also have some recommendations. First, SMAS should be considered as a potential diagnosis for patients at any age who present with a history of unexplained persistent postprandial nausea, vomiting, and weight loss. Second, for patients who have undergone multiple examinations and have failed to respond to medical therapy, early operative intervention should be considered once the diagnosis is confirmed, and electrolyte and fluid imbalances are corrected to prevent extreme weight loss. Third, the management of patients who had psychiatric conditions as comorbidity must be considered to ensure optimal long-term outcomes for patients.

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