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Candy Cane Syndrome: A Review

Candy Cane Syndrome

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KEYWORDS

Candy cane syndrome, blind pouch syndrome, post-gastrectomy syndromes, side-to-side enteral anastomosis, end-to-side enteral anastomosis

ABSTRACT

Introduction: Candy cane syndrome (CCS) is a condition that occurs following gastrectomy or gastric bypass. CCS remains underrecognized, yet its prevalence is likely to rise due to the obesity epidemic and increased use of bariatric surgery. No previous literature review on this subject has been published.

Methods: To collate the current knowledge on CCS, a literature search was conducted with PubMed and Google Scholar for studies from May 2007, until March 2023. The bibliographies of the retrieved articles were manually searched for additional relevant articles.

Results: Twenty-one articles were identified (135 patients). Abdominal pain, nausea/vomiting, and reflux were the most reported symptoms. Upper gastrointestinal series and endoscopy were performed for diagnosis. Surgical resection of the blind limb was performed in 13 studies with resolution of symptoms in 73-100%. In surgical series, 9 complications were reported with no mortality. One study reported the surgical construction of a jejunal pouch with clinical success. Six studies described endoscopic approaches with 100% clinical success and no complications. In one case report, endoscopic dilation did not improve patient's symptoms.

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Discussion: CCS remains underrecognized due to lack of knowledge about this condition. The growth of the obesity epidemic worldwide and the increase in bariatric surgery are likely to increase its prevalence. CCS can be avoided if an elongated blind loop is avoided or if a jejunal pouch is constructed after total gastrectomy. Diagnosis should be based on symptoms, endoscopy, and upper GI series. Blind loop resection is curative but complex and associated with significant complications. Endoscopic management using different approaches to divert flow is effective and should be further explored.

KEY POINTS

- Candy cane syndrome (CCS) is underrecognized and likely to increase in prevalence.
- CCS can be diagnosed by symptoms, upper gastrointestinal series, and endoscopy.
- Blind loop resection is curative but associated with risk of complications.
- Endoscopic management to divert flow is possible, safe, and effective.

INTRODUCTION

It has long been recognized that when long, blind enteral loops are left in place after a side-to-side or end-to-side anastomosis, they can dilate and be the cause of symptoms that may appear many years later (1). The classic designation for this clinical condition is "blind pouch syndrome", although it is possible to find references under other designations, causing confusion (2,3). A particular case of blind pouch syndrome following gastrectomy or gastric bypass is called candy cane syndrome (CCS). CCS was first reported in a 2007 paper describing a series of patients with gastrointestinal symptoms associated with a long blind loop proximal to the gastro-jejunostomy after gastric bypass and creation of an end-to-side anastomosis to a jejunal loop (4). Few case reports and retrospective studies have described this condition. Yet, with the increasing prevalence of obesity and number of operations being performed worldwide, CCS is expected to become more frequent (5).

Probably, the pathophysiology of CCS is exclusively mechanical: a long, mispositioned blind loop preferentially directs luminal contents, increasing

pressure and causing dilatation, pain, regurgitation, postprandial vomiting, and weight loss (Figure 1) (4–7). Caquexia and spontaneous rupture of the blind loop are described (8,9). Given its nonspecific presentation, the diagnosis of CCS is often subjective and based on clinical symptoms in conjunction with the endoscopic and/or radiographic appearance of long and dilated blind jejunal limb proximal to the anastomosis, which is known as the candy cane sign (6,7,9,10). CCS can be prevented by the avoidance of an unnecessary elongated jejunal (blind) loop proximal to the anastomosis in the initial surgery (2–4). A blind loop of less than 3 to 4 cm is usually not associated with obstruction and therefore does not cause CCS. In addition, construction of a jejunal pouch after total gastrectomy prevents CCS and improves feeding, weight recovery, and quality of life (11–14).

For treatment, surgical resection of the dilated loop is curative but technically complex, due to previous surgeries and adhesions, and is associated with non-negligible morbidity (15,16). Endoscopic management of CCS using various approaches to divert the flow from the blind loop is possible, safe, and effective (6,7,17–19).

Knowledge of CCS is important to avoid delays in diagnosis and inadequate treatments. Thus, the goal of this study was to collate evidence on CCS symptoms, diagnosis, treatments, and outcomes.

To the best of our knowledge, no previous literature review on this topic has been published.

METHODS

A literature search was conducted using the PubMed database and Google Scholar, and by searching in addition to electronic links to related articles, from May 1, 2007, through March 31, 2023.

Search terms included candy cane syndrome, blind pouch syndrome, blind loop syndrome, afferent loop syndrome, Roux limb syndrome, post-gastrectomy syndromes, complications of gastrectomy, side-to-side intestinal anastomosis, end-to-side intestinal anastomosis, and symptoms (pain, reflux, regurgitation, vomiting, and/or weight loss) after gastrectomy. The latter terms were used in various combinations for the search. Language restrictions were not applied.

The bibliographies of the retrieved articles were manually searched for additional relevant articles. The articles were carefully read to identify only those exclusively focusing on candy cane syndrome.

RESULTS

In accordance with the search criteria, we identified a total of 21 articles (135 patients), including 13 case reports, 3 case series, 4 retrospective studies, and 1 prospective study. Among these studies, the most reported symptoms were abdominal pain, nausea/vomiting, and reflux. In addition, almost all studies performed upper gastrointestinal series and endoscopy for diagnosis.

Fourteen studies reported surgical resection of the excessive and/or dilated blind limb (13 studies, 111 patients) or construction of an enteral pouch (1 study, 1 patient) with resolution of symptoms in 73%–100% of patients (4,15,16,20–24). In one case, the surgical procedure was performed through thoracoscopy (25). These studies reported a total of 9 complications (1 biloma, 3 infections, 1 anastomosis ulcer, 1 enterotomy, 1 hematoma, 1 pneumonia, 1 hepatic infarction) with no mortality(16,20,22).

Seven studies, including 5 case reports, 1 case series, and the only prospective study available, described various endoscopic approaches: In two studies, a lumen-apposing metal stent was used to divert the luminal content into the efferent loop (18,26); in another two cases, a suture device was used to prevent the passage of food content into the blind loop (17–19). These approaches are technically complex and have low reproducibility; A case report and a prospective study used a magnetic device to cut the tissue between the blind loop and the efferent loop, creating a pouch and allowing the free passage of the food contents (6,7). In this case, the food is not retained in the blind loop and progresses unhindered to the efferent loop. All these endoscopic approaches led to resolution of symptoms in 100% of patients with no reported complications. One case report described candy cane syndrome treatment by endoscopic dilation, which does not divert the blind loop, without success (9).

DISCUSSION

CCS remains underrecognized and misdiagnosed due to a lack of knowledge about the condition. However, its manifestations have been described as

common after gastrectomy (27). In this review, we collected the current evidence on CCS symptoms, diagnosis, and treatment.

When the luminal contents preferentially pass into an overly long blind loop that retains food and distends, the characteristic symptoms of CCS appear, most commonly postprandial abdominal pain associated with nausea and vomiting. These symptoms can appear several years after surgery. Although CCS is a particular case of blind pouch syndrome, it has characteristics that justify being considered an independent clinical entity. As the obesity epidemic persists worldwide and the use of bariatric surgery increases, the prevalence of CCS will likely rise. Thus, CCS should be included in the group of post-gastrectomy syndromes and should be more readily recognized to avoid misdiagnosis, delayed treatment, and inappropriate interventions.

The differential diagnosis of candy cane syndrome should include other surgical complications such as anastomotic stenosis, dysmotility syndromes secondary to surgery, and recurrence in cases of an oncologic indication for gastrectomy.

Collective evidence indicates that the diagnosis of CCS can be suggested based on clinical history and symptoms and should be confirmed by endoscopy and dynamic fluoroscopy.

The characteristic finding in upper gastrointestinal series is a preferential filling of the blind loop followed by delayed passage of contrast into the efferent loop, the so-called "candy cane sign" (10).

Endoscopy in patients submitted to gastrectomy or gastric bypass should include the careful exploration of the blind loop and of the passage to the efferent loop. In these patients, access to the blind loop is usually easy and direct, and access to the efferent loop is difficult and done after passing through an angulation (21). When CCS does arise, effective treatment options are available. Surgical resection of the excessively long and/or dilated loop is curative. However, this method is technically complex, due to previous surgeries and adhesions, and is associated with serious complications in a significative number of patients. By contrast, endoscopic management of CCS using various approaches to divert the flow from the blind loop is safe and effective and should be further explored.

CONCLUSION

CCS is still an unknown diagnosis for most physicians, including gastroenterologists who are often the first clinicians to deal with these patients. Although it is underreported, the prevalence of CCS is probably higher than is commonly thought. Its diagnosis is based on clinical, endoscopic, and imaging findings. Symptoms such as dysphagia, pain, regurgitation, or reflux after food intake are relatively frequent in patients after gastrectomy or gastric bypass and should lead to a precise clinical investigation. Although surgical revision of the blind loop is an effective treatment, it is associated with complications in frail patients with comorbidities. Sectioning of the septum and marsupialization is nowadays the standard mini-invasive treatment for esophageal diverticula. The development of a simple and safe endoscopic technique, such as the blind loop marsupialization described in the only existing prospective study, will in our opinion be the preferred treatment of CCS in the future.

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Ricardo Rio-Tinto, François Huberland, Pauline Van Ouytsel, Cécilia Delattre et al. "Magnet and wire remodeling for the treatment of candy cane syndrome: first case series of a new approach (with video)", Gastrointestinal Endoscopy, 2022

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