Case Report

Success of Serial Transverse Enteroplasty in an Adult With Extreme Short Bowel Syndrome: A Case Report

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Since its introduction as an alternative intestinal lengthening technique, serial transverse enteroplasty has been increasingly used as the surgical treatment of choice for children with refractory short bowel syndrome, but there have been few reports about the adult patients. This report describes the case of a 71-year-old man with a short bowel after distal gastrectomy with Billroth II reconstruction for gastric cancer, followed by extensive intestinal resection. The serial transverse enteroplasty operation was performed and lengthened the small intestine from 49 to 67 cm. The patient tolerated the procedure well and weaned off total parenteral nutrition. Liver function also improved. This case shows that the serial transverse enteroplasty procedure increases intestinal length. This procedure should be considered a surgical option for adult patients with extreme short bowel syndrome.

Key words: Adult – Extreme short bowel syndrome – Intestinal lengthening – Serial transverse enteroplasty

Short bowel syndrome (SBS) is caused by the anatomic or functional loss of a large segment of small intestine and is characterized by the inability to maintain protein energy, fluid, electrolyte, or micronutrient balances when on a conventionally accepted normal diet.¹ Surgical management of SBS focuses on the principles of bowel conservation and restoration of intestinal continuity, which includes a variety of procedures designed to delay intestinal

transit, taper dilated intestine, lengthen the bowel, or replace the lost bowel with an intestinal transplant.^{2,3} However, conservation principles often give way to practical considerations at the time of the initial surgery, resulting in resection of dilated intestinal segments or ostomy creation. Serial transverse enteroplasty (STEP) is an alternative approach that offers the potential benefits of promptly estab-

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lishing intestinal continuity and avoiding further loss of intestinal absorptive area.⁴

The serial transverse enteroplasty procedure was first described in 2003 as an alternative intestinal lengthening technique for patients with refractory SBS.⁵ Similar to other bowel lengthening procedures, STEP lengthens the intestine and narrows bowel diameter with the intent of improving peristalsis and preventing small bowel bacterial overgrowth, malabsorption, and sepsis.⁶ In contrast, STEP has gained favorability as its novel approach is intuitive, relatively easy to perform, poses minimal threat to intestinal blood supply, and can be utilized for varying degrees of bowel dilatation.^{4,6} Accepted indications of STEP include intestinal failure and bacterial overgrowth refractory to maximal medical management, as well as neonatal intestinal atresias or obstructions with limited small bowel length and a dilated proximal segment. However, most reports of STEP were in children with SBS; there were few reports in adult patients. Herein, we report the successful clinical application of this procedure in an adult patient who suffered from proximal 85% of small intestine removed, remained total parenteral nutrition (TPN)-dependent, and was diagnosed with parenteral nutrition-associated liver disease (PNALD).

Case Presentation

This report describes the case of a 71-year-old man with a short bowel after distal gastrectomy with Billroth II reconstruction for gastric cancer, followed by extensive intestinal resection. On January 18, 2013, he was diagnosed with gastric cancer on gastroscopy and underwent distal partial gastrectomy with Billroth II reconstruction the next day (Fig. 1). The histologic examination revealed that the tumor was gastric adenocarcinoma.

However, he complained of abdominal pain, oliguria, lower blood pressure, and increase of intra-abdominal drainage fluid with no obvious incentive on February 7, 2013. Treatments with intravenous fluid, anti-infection, and TPN were applied, but there was no clear improvement. Emergency exploratory laparotomy was performed 2 days later. At operation, the patient was found to have approximately 500 mL hemorrhagic ascites. The gastrointestinal anastomosis healed well, and the proximal bowel was only 20 cm in length from the Treitz ligament with dilatation, and the distal bowel was 5 cm in length from the ileocecal valve, while the remainder of the small intestine was

Fig. 1 Sketch of prior surgery: the distal partial gastrectomy with Billroth II reconstruction.

nigrescent and necrotic. The mesentery appeared purple and the arterial pulse was weak, which indicated there might be mesenteric vein thrombosis, so we performed the resection of necrotic intestine. Histological examination showed extensive small bowel hemorrhagic necrosis. Treatments with anti-infection and TPN were applied immediately after the operation. Postoperatively, he received TPN support for 5 months due to his intolerance of enteral nutrition (EN), and then we performed a computed tomography angiography (CTA) examination, which showed multiple plaque in superior mesenteric artery and lumen stenosis.

Considering the poor tolerance to EN, we performed the STEP procedure and replaced Billroth II with Billroth I on July 17, 2013. Intraoperative exploration showed the patient had relatively even dilatation of the small bowel with 4 cm in width (Fig. 2), which prompted us to perform the STEP procedure. To take full advantage of the rest small intestine, we performed the Billroth I reconstruction. On exploration, the total length of small intestine was about 49 cm. we performed the STEP procedure using 45-mm linear cut staplers (Covidien Public Ltd. Co., Dublin, Ireland) on the proximal small bowel. As a result, the intestinal length was increased from 49 to 67 cm (Fig. 3). During the operation, the bowel was flattened, keeping the antimesenteric line directly in the middle while the

Fig. 2 The dilated small bowel with 4 cm in width.

stapler was applied leaving approximately 6 cm of bowel uncut at the end. The next firing of the stapler was approximately 6 cm distal to this, and the stapler was brought in from the opposite side. This was repeated 6 times along the length of dilated bowel. All stapler applications were kept perpendicular to the long axis of the bowel, thus preserving good blood supply to the intestines (Fig. 4). Postoperatively, TPN was continued but routine laboratory tests showed the patient suffered from PNALD (direct bilirubin concentrations was 3.68 mg/dL after 8 weeks of parenteral nutrition [PN]). It seemed that liver function was affected. We applied the combined use of parenteral and enteral nutrition

Fig. 3 STEP was performed using 45 mm linear cut staplers (Covidien).

Fig. 4 Sketch of STEP

support at 10 days after operation and the patient was tolerable to EN with body weight gain. Total enteral nutrition support was applied at 1 month postoperatively, and the energy was 1500 cal/d. The frequency of defecation was about 4 times per day. Plain film examination and computed tomography 3D reconstruction showed that the gastrointestinal tract was unobstructed and with uniform expansion. Kjeldahl method showed that the patient was tolerable to EN at 3 months postoperatively (Fig. 5). Meanwhile, because of the usage of EN, the liver function improved (Fig. 6), and the risk of death caused by PN-associated liver complications was also reduced. He was discharged 2 months after the STEP procedure and tolerated EN, with normal liver function and mental development.

Discussion

Intestinal lengthening procedures have been shown to improve intestinal function in patients with SBS.

Fig. 5 Absorption rate of nitrogen pre- and post-STEP showed that the patient was tolerable to EN. Absorption rate of nitrogen were continuously measured on the 1st, 2nd and 3rd pre- and postoperative day.

The serial transverse enteroplasty procedure was developed in 2003 as a novel bowel-lengthening procedure, and has been shown to be safe and effective in the short and intermediate term, and as a repeatable procedure.^{5,7,8} Serial transverse enteroplasty allows for the creation of uniform intestinal caliber in patients with variable degrees of bowel dilation. Additional potential advantages of STEP include the absence of intestinal anastomoses, decreased risk of vascular compromise, and reduced technical complexity when compared with other bowel-lengthening procedures.

Patients with SBS often require long-term PN to maintain daily nutritional requirements until the residual intestine undergoes adaptation and nutritional autonomy is obtained.^{9,10} However, although PN is life-saving, its long-term use is associated with severe adverse effects,¹¹ including septic infection, metabolic imbalance, and hepatobiliary dysfunction.¹² The hepatobiliary complications of PN are now well recognized as PNALD, which is associated with increased morbidity and mortality.¹³ Approximately 40 to 60% of infants and up to 85% of neonates are receiving long-term PN for intestinal

failure, and approximately 15 to 40% of adults are receiving home PN.¹⁴ Due to the potential complications, cost, and quality of life issues, intestinal lengthening procedures such as Bianchi and STEP have been used more and more widely in clinical practice. The STEP procedure became popular as it is technically simple (relative to other lengthening procedures), does not require intestinal anastomosis. What's more, it results in significant lengthening of the bowel and can be repeated and performed even after other lengthening procedures.¹⁵

Several issues merit discussion in this case. First, prior STEP procedures were almost frequently applied to pediatric SBS and there were few reports about the adult patients, while this case was an adult patient that finally gained successful treatment. Second, this case was for an adult patient with gastric cancer who already underwent several operations, which resulted in extreme short bowel before the STEP procedure. In order to better utilize the rest of the small intestine, we replaced Billroth II with Billroth I and lengthened the rest of the small intestine from 49 to 67 cm. Third, we chose a cut edge of 1.5-cm deep in this case based on our

Fig. 6 Liver function pre- and post-STEP showed that the liver function improved. ALT, alanine aminotransferase; DB, direct bilirubin.

experience and 6 equal sessions according to the length and blood supply of the small intestine. Lastly, the patient weaned off the TPN and had a rather good tolerance to EN after the STEP procedure, which provided relief from PNALD. This indicated the STEP procedure can be performed as a primary bowel-lengthening procedure.

Serial transverse enteroplasty may also have deficiencies. Studies have revealed that patients underwent STEP may develop gastrointestinal hemorrhage.¹⁶ A single institution series report on the STEP procedure by Ching *et al*¹⁷ revealed that 3 out of the 16 patients followed had gastrointestinal bleed that did not require reoperation. The underlying cause for these bleeds was not described. A recent single institution series report by Wales *et al*¹⁸ and subsequently by Oliveira *et al*¹⁸ revealed that of the 12 patients followed, one was noted to have developed chronic intestinal bleeding due to staple line ulcers that were surgically wedged out. Pathological evaluation of these ulcers did not reveal an underlying causative factor. Although the site of these ulcers (distal small intestine) and the timing of the bleeding (8 months post-STEP) were noted in many cases, the exact cause of bleeding was not demonstrated.

This case shows the safety and feasibility of the STEP procedure in an adult patient with extreme short bowel. This patient had improved intestinal absorptive capacity after the STEP procedure and has successfully remained clinically TPN independent. As a result, PNALD was in remission. The patient has been observed for relatively brief periods during the follow up and, to date, there has been improvement of intestinal absorptive capacity and body weight. However, as mentioned above, additional studies will be required to determine the full physiologic impact of this operation on patients who suffer from SBS.

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