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CASE REPORT

The First Clinical Application of the Spiral Intestinal Lengthening and Tailoring (Silt) in Extreme Short Bowel Syndrome

T. Cserni · B. Biszku · I. Guthy · F. Dicso · L. Szaloki · S. Folaranmi · F. Murphy · G. Rakoczy · A. Bianchi · A. Morabito

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Abstract

Aim Spiral Intestinal Lengthening and Tailoring (SILT) invented by our team is a new technique that offers minimal mesenteric handling and a more physiological result compared to the STEP procedure. Its feasibility has been tested in animal models and now we report the first successful human application in extreme short bowel syndrome.

Materials and Methods A 3-year-old girl suffered subtotal loss of her small bowel and ileocaecal junction as a result of midgut volvulus. Only 15 cm of jejunum remained intact. Parenteral nutrition (PN), gastrostomy feeding, controlled bowel expansion and SILT procedure were applied.

Results The length of the jejunum increased from the initial 15 to 22 cm during 12 months of PN and bowel expansion. Eleven centimeter of distended bowel was further lengthened up to 20 cm by SILT giving a total small bowel length of 31 cm. Oral and gastrostomy feedings were commenced 5 days postoperatively. There were no surgical complications 6 months after the procedure. The patient's liver function was preserved, she was weaned off PN, discharged from hospital, but remained on gastrostomy top up feeding. The net weight gain of the patient was 1,800 g 6 months after the procedure.

Conclusion SILT procedure is a safe and feasible technique for human intestinal lengthening and tailoring.

Keywords Spiral Intestinal Lengthening and Tailoring (SILT) · Short Bowel Syndrome · Parenteral nutrition

Introduction

Non-transplant surgical management of residual autologous bowel in patients who suffer from short bowel syndrome is a promising and rapidly developing option in lieu of or before intestinal transplantation. Success depends on intestinal adaptation with a sufficient increase in absorptive mucosal surface area and adequate bowel motility, providing enough time for absorption but preventing the stasis and bacterial overgrowth that is associated with dilated and poorly propulsive bowel.[1,2]

In clinical practice, intestinal tissue expansion is used to increase the residual small bowel in length and mucosal surface area thereby providing additional new tissue for reconstructive procedures. These include bowel tailoring and lengthening, which are designed to return the dilated adapted small bowel towards a normal propulsive diameter and function.[3]

At present longitudinal intestinal lengthening and tailoring (LILT)[4] and serial transverse enteroplasty (STEP)[5] are the techniques available with various success rates depending on reports from different centres. The Spiral Intestinal Lengthening and Tailoring (SILT) is a new emerging technique that has the advantage of minimal handling of the mesentery but does not dramatically alter the orientation of the muscle fibres like the STEP procedure.[6–8] The feasibility of SILT has only been tested in animal models, and this paper reports on the first human application in extreme short bowel syndrome.

T. Cserni (✉) · B. Biszku · I. Guthy · F. Dicso · L. Szaloki
Department of Pediatrics, Josa Andras Hospital Nyíregyháza, Teaching Hospital of Medical Health Science Centre University of Debrecen, Debrecen, Hungary
e-mail: tcserni@yahoo.com

T. Cserni · S. Folaranmi · F. Murphy · G. Rakoczy · A. Bianchi · A. Morabito
Department of Pediatric Surgery, Royal Manchester Children's University Hospital, Royal Manchester, UK

Patient Report

A 3-year-old girl, born prematurely at 24 weeks gestation, who was blind and had learning difficulties as well as failure to thrive, presented with a 24-h history of bilious vomiting, shock and an acute abdomen. Emergency laparotomy revealed a midgut volvulus with severely compromised small bowel. Following derotation and attempted reperfusion, only 15–17 cm of proximal jejunum was viable. The rest of the small bowel with the ileocecal junction and 6–8 cm of the ascending colon was resected. A primary end-to-end jejunocolic anastomosis developed a leak on the fifth postoperative day and she underwent a second laparatomy and stoma formation. At this time, at 8 kg (<fifth percentile) body weight, with informed parental consent and ethical approval of the National Research Ethics Service, controlled bowel expansion[3] was commenced with a view to intestinal lengthening with guidance from a specialist centre (Royal Manchester Children's Hospital).

Vascular Access A 6.6 Fr Hickman line was inserted surgically initially trough the right external jugular vein. Line sepsis occurred on three separate occasions and was treated successfully with combined antibiotics, antifungal treatment and 70 % ethanol line locks. The central venous line was changed three times; once because of sepsis, once because of blockage and once because of external damage.

Parenteral Nutrition before Lengthening Procedure Low fat (<1.5 g/kg) hepatosparing parenteral nutrition (PN) was commenced after the first laparotomy. The liver function was monitored closely and preserved during treatment.

Oral and Gastrostomy Feeding before Lengthening Procedure Enteral feeding was introduced from the fourth postoperative day after stoma formation. Oral feeding was encouraged at all times; however, the patient refused oral intake for a long period despite of regular review by the psychologist and dietician. Therefore, a gastrostomy tube was inserted and hydrolysed semi-elemental formula containing medium chain triglycerides was introduced. Regular replacement of B_{12} vitamin and micronutrients was administered.

Controlled Bowel Expansion At laparotomy, the stomas were taken down, and a tube jejunostomy and colostomy were performed to allow safe recycling of bowel content. The jejunal tube stoma was clamped for increasing periods of time from 30 mins to 3 h and unclamped for 40 min. Fifty to seventy-five percent of the aspirates from the proximal stoma were slowly recycled into the distal mucus fistula.

After 2 weeks, the proximal stoma leaked into the abdominal cavity resulting in localised peritonitis and removal of the tube stomas with reestablishment of stomas. After recovery, 24 Fr Foley catheters were inserted and the proximal stoma was surgically narrowed at the level of the abdominal wall. After 4–5 weeks, the stoma diameter became more dilated and the catheter balloon was unable to provide an adequate seal, therefore, 2/0 Ethilon sutures were placed under local anaesthetic around the stoma. The bowel expansion was suspended when the patient developed severe skin erosion and infection around the stoma site. After a few weeks, the stoma narrowed allowing the catheter to provide a better seal and bowel expansion was recommenced. Dilatation of the bowel segment was controlled under fluoroscopic guidance (Fig. 1a, b).

Intestinal Lengthening

After 12 months, due to the fact that there was no further bowel dilatation, intestinal lengthening and tailoring was considered. At laparotomy, the jejunal length was found to have increased from 15 to 22 cm with a diameter of 4 cm for the distal 11 cm. This segment was further lengthened up to 20 cm by SILT to give a total jejunal length of 31 cm and with a diameter of approximately 2 cm.

The SILT procedure: Firstly, the dilated segment was mobilised and the spiral incision line marked, keeping the angle between 45° and 60°. Stay sutures were placed where the marking lines met the antimesenteric and the mesenteric borders. The incision was completed with monopolar diathermy using a Colorado needle. A 2–3 cm incision was made in the mesentery passing between the blood vessels, where the marked spiral incision line met the mesenteric border. A 2 cm diameter, silicon catheter was inserted and secured into the lumen to maintain the orientation of the bowel, which was subsequently lengthened and narrowed down to 2 cm in diameter.

The bowel loop was stabilised in its new shape using 5/0 Maxon interrupted sutures every 2–3 cm along the spiral line, then the suturing was completed with a continuous serosubmucosal suture (5/0 Maxon) (Figs. 2–3). After the completion of the spiral suture, the jejunum was anastomosed to the colon in an end-to side fashion to mimic the natural ileocaecal junction. Yates drains were left intraperitoneally and were removed after 3 days. The patient was on intravenous Co-amoxiclav, Gentamicin and Metronidazole for 7 days. Nasogastric tube aspirates diminished after 3 days, the patient opened her bowel on the fourth postoperative day and enteral feeding was commenced on the fifth postoperative day.

Parenteral Nutrition after Lengthening Procedure PN and close monitoring of liver function continued after the lengthening procedure. PN was discontinued 4 weeks after the procedure, at which point her enteral calorie intake had exceeded 100 Kcal/kg and weight gain was consistent.

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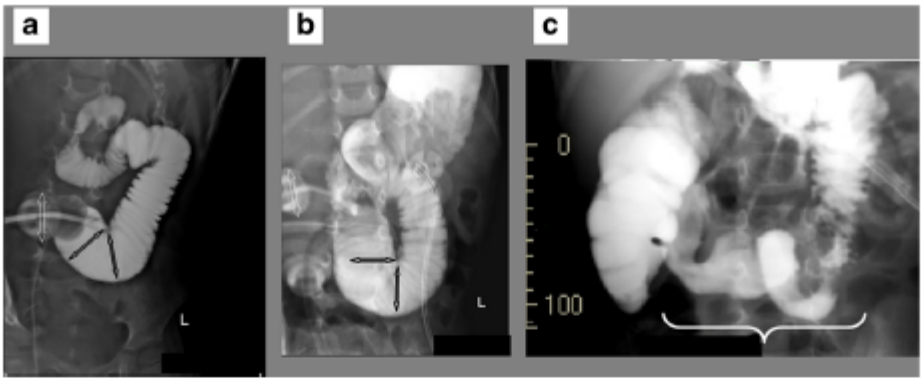


Fig. 1 Contrast studies demonstrating the small bowel size before bowel expansion (**a**) after bowel expansion (**b**) and after SILT (**c**). *White arrows* (marking the plough used for catheter fixation in the stoma) are used to equalize magnification between image **a** and **b**. They are the same size on both **a** and **b** images. The *black arrows* are representing the bowel diameter before bowel expansion on **a** and **b** images. Dilatation and some lengthening after bowel expansion can be observed in image **b**. Image **c** shows the tailored and lengthened segment anastomosed end to side into the ascending colon

Oral and Gastrostomy Feeding after Lengthening Procedure Enteral gastrostomy feeds with hydrolysed semi-elemental formula containing middle-chain triglycerides were introduced on the fifth postoperative day. The child refused oral feeds for a prolonged period, however, following major input from the psychologist and dietician, oral food tolerance finally improved and the gastrostomy was used only for top-up and overnight feeding.

Present Condition The patient, 6 months after the lengthening procedure, is at home with her family. She is tolerating oral diet and overnight gastrostomy feeding. Her present weight is 9.8 kg (<fifth percentile) and her weight gain after the bowel resection was 1.8 kg during an 18 months period. She is opening her bowel up to three times a day, and the consistency of her faeces is between 5 and 6 on the Bristol stool chart. The lengthened and tailored segments remain intact as demonstrated on a contrast study (Fig. 1c).

Medication

Proton pump inhibitors have been continuously used to reduce increased gastric and enteral secretions. Loperamide has been used to decelerate intestinal transit. Combined antibiotics and antifungal drugs were used only occasionally according to the culture and sensitivity microbiological specimens.

Discussion

Autologous intestinal reconstruction combined with controlled intestinal tissue expansion is a promising option even in severe short bowel syndrome and should be considered before transplantation. Our patient would not have met the selection criteria for intestinal transplantation because of the associated pathologies and limited family compliance.[9] The patient spent only 2 weeks in a supra-regional centre for the actual lengthening procedure, all other medical and surgical interventions were performed in the local department of paediatrics. This allowed the parents regular visits to the hospital and the opportunity to conduct their normal life.

Controlled bowel expansion follows a well-established protocol[3]; however, it proved to be difficult in our case.

In order to lengthen and tailor the dilated bowel, LILT, STEP and SILT were considered.

To avoid difficult mesenteric manipulation associated with Bianchi's technique, we decided to choose between STEP or SILT procedure. There is no difference in terms of mesenteric manipulation between STEP and SILT. Minor perpendicular incision of the mesentery is mandatory in both procedures. In STEP, it allows the zig-zag shaped bowel to stretch to a straight loop, whereas in the SILT procedure it allows the bowel to twist and lengthen.

Neither the STEP nor the SILT procedure will disrupt the propagation of myoelectric activity or alter the peristaltic reflex in the refashioned bowel. However, in the STEP-ed segment, the orientation of the muscle fibres changes dramatically as the zig-zag shaped loop straightens into a uniform loop: the circular muscle fibres become longitudinal, while the longitudinal fibres become circular (Table 1). The significance of this lies in the ability of the refashioned segment to propel the chyme. In a normal bowel, the peristaltic reflex is triggered by the food/chyme bolus in the lumen. At first, the circular muscle fibres contract proximal to the bolus narrowing the lumen down to stop the bolus migration cephalad; secondly, the contraction of the longitudinal muscle follows and as a result of this coordinated action the bolus progress in a caudal direction.[10]

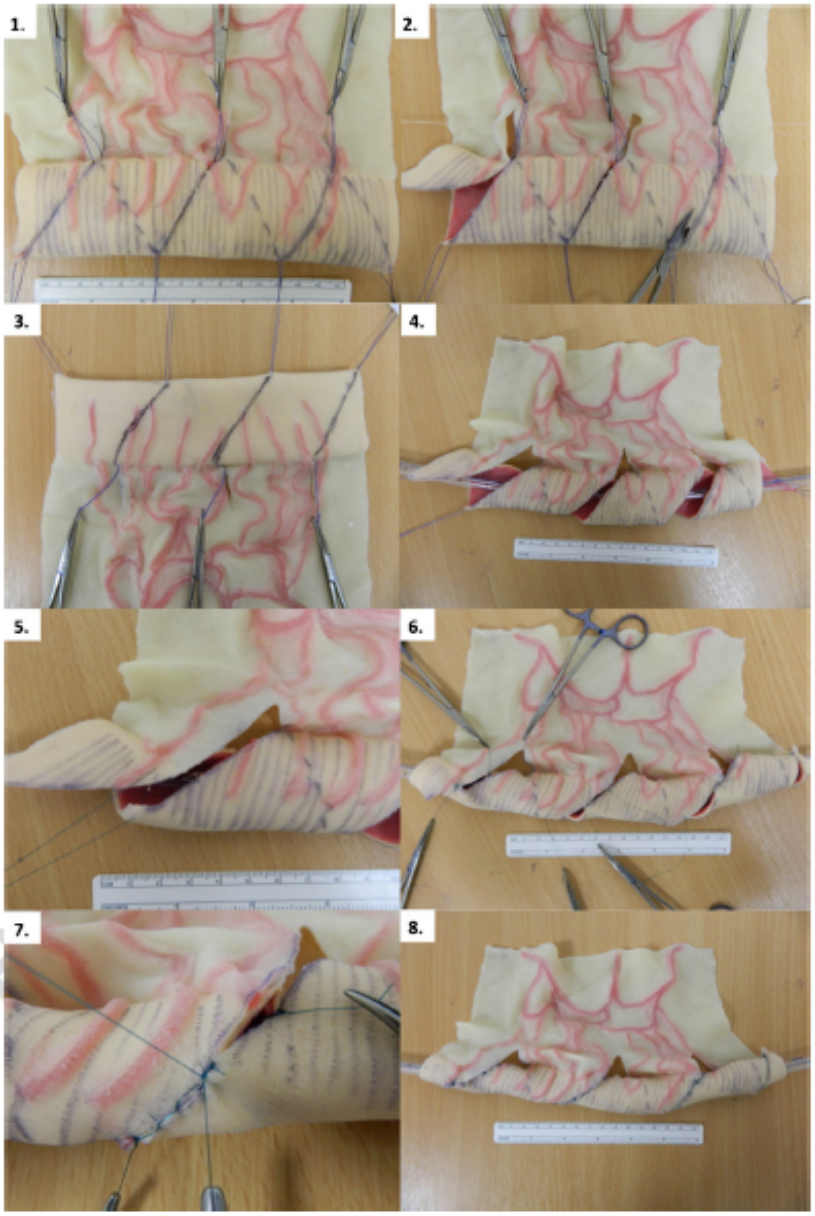


Fig. 2 *1–8* Figure shows the SILT procedure in eight steps on double layer bowel simulator with mesentery (designed for practicing by our team and Limbs and Things UK). *1* Incision line marked at approximately 60°, continuous line on the anterior, interrupted line on the posterior wall of the bowel. Stay sutures are placed where the line met the mesenteric and the antimesenteric line. *2* Incision on the anterior wall and on the mesentery. *3* The bowel flipped up to access the posterior wall, incision line marked. *4* The bowel flipped back after incision complete on the back wall, the intestine stretched along a large tube into a narrower but longer shape. *5* The first stitch gives the new caliber of the bowel. *6* The new shape is stabilized along the loop with interrupted stitches 2–3 cm apart along the spiral line. *7* Continuous locking serosubmucosa suture between two stay stitches, the suture tied to the interrupted stitches from time to time to prevent purse stringing. *8* The reconstructed bowel in narrower but longer shape. Note the position of the circular muscle fibres are close to normal

In a STEP-ed bowel loop, however, as the reflex circuit remains intact, the bolus first triggers the longitudinal (originally circular), than the circular (originally longitudinal) fibres. We believe this may lead to disturbed peristalsis or stasis.

After the SILT procedure, the orientation of the muscle fibres is altered compared to the normal anatomy and the Bianchi procedure. However, the oblique "circular muscle" fibres still have a significant projection perpendicular to the longitudinal axis of the bowel loop to narrow the lumen. The very mildly altered position of the muscle fibres after SILT can be clearly seen in our model (Fig. 2(8)). Minimal alteration was observed on the orientation of the mucosal folds and the relation of the nuclei of muscle cells on HE stained slides taken from the from SILT-ed bowel loop in our previous animal experiment.[7]

Considering this and that the main goal of the intestinal lengthening and tailoring procedures is to improve peristalsis, we decided to opt for the SILT procedure. The procedure was less challenging than LILT with regards to division of the mesentery. After stabilising the bowel in the new, longer, but narrower shape, it was easy to perform the final serosubmucosal continuous suture line in a safe fashion. Due to the spiral shape, the suture line in SILT is somewhat longer than in LILT; however, we did not feel this was significant. The

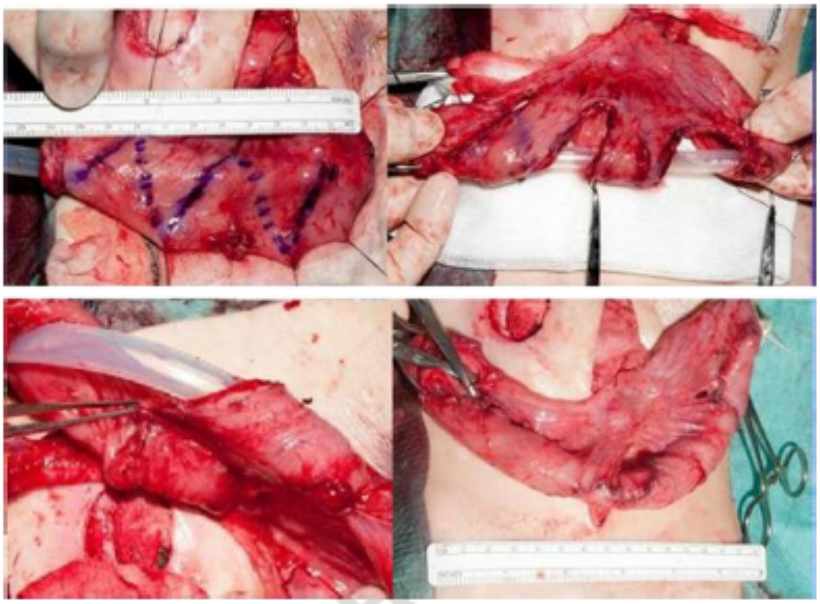
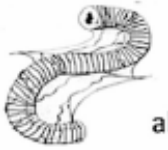


Fig. 3 SILT on human bowel: *Left upper* marking of the spiral incision on the dilated bowel segment. *Upper right* large silicon tube was stitched into the lumen to keep the orientation of the bowel segment. *Lower left* reconstruction of the jejunum in a narrower, but longer fashion with situation stitches first. *Lower right* the lengthened segment after sutures completed

application of a stapler theoretically makes the STEP procedure easier and quicker; however according to our experience, the staple lines, especially at their end, almost always require additional suturing to provide an appropriately safe seal.

With complex management, PN, bowel expansion and SILT, we were able to achieve enteral autonomy in a patient with extreme short bowel syndrome and severe associated anomalies.

t1.1 **Table 1** The table compares LILT, STEP and SILT procedures in terms of mesentery handling, adjustability of the lengthening and tailoring, and shows the change in the orientation of the muscle fibres

Procedure	Mesentery handling	Lengthening and tailoring	Muscle fiber orientation
LILT 	difficult	tailors diameter in half	 not altered
STEP 	minimal	adjustable	 fully altered
SILT 	minimal	adjustable	minimally altered 

Our case is the first evidence that the SILT procedure is safe and may be used as an alternative to the present techniques; however, comparison studies are required to confirm the expected advantages.

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